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An exploratory study of children with long-term ventilation care packages and their family narratives using a salutogenic-based approach

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An Exploratory Study of Children with Long-Term Ventilation Care Packages and Their Family Narratives using a Salutogenic-Based Approach.

By

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Abstract.

The number of children and young people receiving long term ventilation (LTV) care in the United Kingdom in their home settings has grown significantly (Wallis, Paton et al 2010:999). Due to the complexity, intensity and risk involved in this type of home care, many children and young people who are in receipt of LTV, receive nursing care support at home. Despite the significant cost of these care packages, several authors have noted the low levels of satisfaction of families receiving such packages (Dybwik et al 2011, Fine-Goulden et al 2015, Kirk et al. 2004). Conversely, little is known about what children, young people and their families find works for them in terms of coping with the complexities and care burden that LTV brings into the context of their daily lives. This novel study sought to fill this significant gap in the current research base, through innovative adaptation and application of Antonovsky's (1987) Salutogenic theory. Antonovsky's three concepts of *Comprehensibility, Manageability and Meaningfulness* (Antonovsky 1987) were adapted and used, along with arts-based methods to study children and young people's LTV care.

Firstly, data was collected by eight families completing a daily diary, scrapbook and memento box for one month, considering questions based around each of Antonovsky's three concepts. Secondly, families were then interviewed at home focusing on the approach. Following on, a new and novel method of participatory play-based scenarios was developed and used for the first time in this study to gain the views of 26 participating children aged four to ten. Analysis was then undertaken utilising two different types of content analysis known as explorative and theory-based content analysis (Hsieh and Shannon 2005). This approach allowed flexibility and depth in the analysis process.

This study identified three important themes. The first adapted from *meaningfulness* (Antonovsky 1987) was described as "the next big leap" and highlighted that for both the children and their families, meaningful personal or family aims were significant motivators for them to take on, and live with the issues, complexities, skills and care requirements impacted by a child with LTV needs. The second theme adapted from *Comprehensibility* (Antonovsky1987) was discussed as "a life of learning and adaption". High level findings identified significant daily life and knowledge changes for the children and their families. As the child developed, further complex adaption was required. The third theme was adapted from *Manageability* (Antonovsky 1987*)* and was discussed as "working together to make life and LTV manageable". It highlighted the need for consistent and flexible care, by staff who knew the child, young person and family well. Conversely, the professional support the children and their families experienced was planned around problems, had little or no teaching or case management support and varied greatly. The care and was frequently inflexible in terms of delivery of care.

It was the novel approaches adopted within this study, that helped produce new and important findings around what works for children and families in home-based LTV care. Importantly, this study also identified the differences between what children and their families want, and professionals aspire to provide. In a context of ongoing, unprecedented growth of LTV, and wide spread dissatisfaction with home care provision, both nationally and internationally, this study makes an important contribution.

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I wish to thank Professor Jane Coad for her support and inspiration throughout my research journey. Jane's research utilising Arts-based methods inspired me to undertake this study. Jane provided the expert debate, challenge and support I needed to move my practice-based ideas into a robust research study. I also wish to thank Dr Andree Dignon for her dedication and research expertise throughout this seven-year journey. Andree's attention to detail has been second to none and vital to the quality of this research study. Dr Rosie Kneafsey joined later in my study and has provided a fresh view and a constructive critical edge to all her reviews for which I am most grateful.

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Chapter 1. Introduction: The Emergence of the Study

This study was written in a time of rapidly increasing numbers of children and young people being initiated on long-term ventilation, in the United Kingdom (UK), and the wider western world (Dybwik et al.2011, Lima et al.2015, Wallis, Paton et al 2010:999). Research had demonstrated the significant difficulties that children, young people and their families faced (Lima et al.2015, Mendes 2013, Noyes 2000). However, there was also reported widespread dissatisfaction being expressed by children, young people and their families in the care they were receiving (Dybwik et al 2011, Fine-Goulden et al 2015, Kirk et al 2004). This frustration was felt not just by the children and families but also the teams that supported them (Maddox and Pontin 2013).

During this time, I was a Consultant Nurse for Children with Complex Needs and led two large care teams that increasingly had taken on supporting this group of children and young people. From this context and my professional experience, a research idea emerged which was to become this study. This idea focused around taking a very different approach that of Salutogenesis (Antonovsky 1987,1979) explored in Chapter Three as long-term ventilation (LTV) is a health need across all infants, children and young people. I will also use the term **Children** throughout to describe infants, children and young people up to the age of 19 years (the age limits for my study focus).

As this research will be focused on exploring views, I firstly set the scene about my own influence on the research and will therefore start with explaining my position and how this research topic and questions emerged for me before going on to consider the specific context of LTV in Children and their families. As a Consultant Community Children's Nurse, I work with Children and their families as they return home from hospital and resume their lives. The struggles they face are daunting but the ability of the Children and their families to adapt is inspiring. However, some cope much better than others. Community Children's Nurses have the privilege of being involved in supporting Children and families through this journey. During the first ten years of my career as a Community Children's Nurse (mostly working with Children with Type 1 diabetes), I focused on the traditional nursing approaches of assessing, treating, teaching, listening and supporting the child and their family. I became increasingly frustrated with the lack of impact from these approaches. In 1999, I was introduced for the first time to the theoretical model of salutogenesis (Antonovsky 1987,1979). The simple but paradigm changing approach of studying why people get well and adapt rather than why they get ill and struggle, turned my thinking on its head. My approach and practice changed as I developed my thinking through a Master of Science in Health Development thesis utilising this approach. I was also inspired by the impact of this change of focus in my clinical work. For me, it totally made sense because the children and families I saw get on and thrive were largely not motivated by avoiding complications but by mastering diabetes to lead the lives they wanted to lead.

Some years later, I moved into the field of complex care and long-term ventilation (LTV). Initially the complexity and risk involved in caring for this cohort of Children pushed me back into the comfort and certainty of traditional nursing approach. Problem-focused approaches were dominant in the intensive care treatment of this group of children and their families received prior to discharge. This problem focused approach then followed them out into the community and their day to day lives. Care rotated around risk mitigation and safety. As I became confident in the technical care of these Children and families and the literature supporting practice, I reflected back to salutogenesis; on further reading of the literature I realised the salutogenic approach (Antonovsky 1987,1979) appeared absent in Children's LTV research. There was wide spread dissatisfaction expressed by families with the care they received; both in practice and the literature, and I started to consider whether an approach developed and adapted from the salutogenic theory (Antonovsky 1987,1979) could provide a more empowering and effective approach to providing care for this group of Children and their families. There appeared a clear gap in theory and practice. As I pondered on this, I became involved in research studies undertaken by Professor Jane Coad. The participatory Arts-based approaches utilised in these studies was inspirational and I began to ponder on using a participatory Arts-based approach in a study to consider the gaps, I saw both in Children's LTV practice and research. My enthusiasm for returning to academia strengthened as an area for study, theoretical approach and participatory Arts-based methods came together. I went to see Professor Jane Coad and as we talked, I was inspired and knew that the time was right to embark on a PhD and following on I enrolled upon a part time PhD course at Coventry University.

As my study developed the overall study aim evolved to:

From the perspectives of Children and families what helped and enabled them to overcome the complexities that Long-Term Ventilation (LTV) brought into their lives.

To explore this aim, two research questions were honed and developed through the Children LTV literature and exploration of the salutogenic theory as outlined below.

1. What enables children and young people and their families to overcome the complexities that home long-term ventilation brings into their lives?
2. Does the salutogenic theory offer a new theoretical approach to underpin conceptualisations of the care for children and young people living at home with long-term ventilation support?

The study questions and the innovative mix of participatory Arts-based approaches taken during this study led me to identify several exciting new and novel findings. To me, the most important of which was "the next big leap" adapted from Meaningfulness (Antonovsky 1987). This theme identified the importance, to Children and their families, of regaining a focus on aims that were worth striving for them. This focus appeared to provide the motivation to start the parent's (and later the Child's) adaption to LTV. The motivation from this focus upon personal meaningful aims gave them the drive and focus to make sense of their new situation. The next theme "a life of learning and adaption" adapted from comprehensibility (Antonovsky1987), encapsulated the process of acquiring skills and understanding to enable initially parents to adapt LTV, (and later children), within the context of their lives at home within their community, and importantly the context of the child's stage of development. Working together to make life and LTV manageable, adapted from manageability (Antonovsky 1987) identified how the families (and later children) utilised support, both formal and informal to, resume their lives. This process appeared to repeat as the child developed or the context of the child or family's life changed. This seemed to me to be an important process to observe and capture.

When I related this initial observation to my own life, this resonated with my own personal drive to manage my dyslexia. I knew from the outset that this PhD would be the ultimate academic challenge and my deepest fear. But I knew this topic needed researching and would help me explore person-centred approaches, much needed if professionals are to support the growing needs of children and young people and their families with complex conditions. This drive to improve the care provided to, children and families living with LTV created in me the ability to overcome the significant challenges my dyslexia presented in the level of writing involved in undertaking this study. The third person writing style was chosen at the start of the study, this decision was made as I had been immersed in this style of writing since entering the nursing profession, making a change to the first person style was too difficult after 25 years of socialisation into a third person style (further complicated by the challenges of dyslexia).This decision on reflection, although pragmatic at the time, is a decision that I have moved on from and would reconsider in future studies. The third person style will now be used in the thesis until Section 7.8. In recognition of the importance of the children's voice in this study and to emphasise their contribution, I have also chosen to capitalise the words Child and Children from this point on. This leads me from my reflective introduction on to setting the scene about this group.

1.1 Context of Children and Young People's Long-Term Ventilation

Ventilation is the term used to describe mechanical assistance of breathing in the presence of respiratory insufficiency (inadequate breathing to maintain health) or an absence of a drive or ability to breathe. Long-term ventilation (LTV) is the term used when it has not been possible to discontinue (wean) ventilation. In this scenario, the ventilation becomes a long-term requirement. The failure to wean off ventilation is defined as, "any child when medically stable requiring a mechanical aid for breathing after an acknowledged failure to wean [off the ventilator], or slow wean, three months after institution of ventilation" (Jardine et al. 1998).

This inability to manage effective breathing without ventilation necessitates the use of ventilation in everyday life for an extended period and in many cases for the rest of the Child's lifespan. This dependence on ventilation can be just when the Child sleeps or for most or all of the twenty-four-hour period.

The underpinning conditions and causes that can lead to a failure to wean and a need for LTV are varied and there are three main categories of Children who require this support. The categorisations used are based upon anatomical systems: central nervous system; musculoskeletal; and respiratory system. However, not all long-term ventilated Children are as dependant on long-term ventilation. This level of dependency has a significant bearing on the day to day impact of long-term ventilation. Interestingly, level of dependency has been described in different ways in the literature. The term "life sustaining" is frequently used to determine the highest level of ventilation, but in cases of respiratory insufficiency this can be less clear, leading to different sublevels of dependence needing to be defined. The approach taken in this study was to adopt the levels outlined in The National Framework for Children and Young People's Continuing Care (2016). These levels are:

1. High - Can breathe unaided during the day but requires a ventilator for supportive ventilation. The ventilation can be discontinued for up to 24 hours without clinical harm.
2. Severe - Requires ventilation at night for very poor respiratory function; has respiratory drive and would survive accidental disconnection but would be unwell and may require hospital support.
3. Priority - Has no respiratory drive when asleep or unconscious and requires ventilation, disconnection of which could be fatal (DH 2016:32-33).

This study focused on level three (Priority) as this is the group of Children and families is where there is the most significant impact of the LTV.

The numbers of children and young people receiving long-term ventilation care in the UK in their home settings has grown significantly from one in 1975 to 933 by 2010 (Wallis, Paton et al. 2010:999). Due to the complexity, intensity and risk involved in this type of home care, Children, and families receive nursing care support at home. The levels of support identified in the literature (Margolan et al. 2004) varied from twenty-four hours a day, to a few nights per week, with trained healthcare support workers. Such care is referred to in this study as a care package. Despite the significant cost of these care packages, a number of researchers have noted the low levels of satisfaction of families receiving such packages (Dybwik et al. 2011, Fine-Goulden et al. 2015, Kirk et al. 2004).

Unlike many health interventions, the growth of the numbers of children receiving (LTV) has been closely researched since its inception. The studies indicated below have identified the significant increase in the numbers of Children receiving (LTV) at home.

1975 – One Child (Robinson 1990 cited in Noyes 1999:441),

1988 – 24 Children (Robinson 1990 cited in Noyes 1999:441).

1998 – 141 Children (Jardine 1999).

2010 – 933 Children (Wallis, Paton et al. 2010:999).

It is worth noting that these surveys were all dependent on practitioners in Tertiary Hospitals completing questionnaires at the request of the research teams. The weakness of this approach is normally with regard to response rates; however, the 2010 data achieved all but one UK centre completing the data collection. The increase in children living with LTV has been noted across the developed world (Dybwik et al. 2011, Lima et al. 2015). There is some evidence from Canada that the numbers of Children receiving LTV is plateauing (McDougall et al. 2013). Whether this trend is specific to Canada or more general is unknown. Updating of the UK incidence study, at the time of writing, was due to be carried out in 2020.

The dramatic rise in the numbers of LTV Children has had significant consequences for them and their families, the National Health Service, Education, Social Care and the wider statutory and voluntary sector. The reasons for the increase in numbers are complex and broad. Part of this change is related to Children surviving the initial critical times (such as immediately after birth) or the hours and days that follow a significant accident or illness. Advances in intensive care treatment are at the forefront of this change. Deaths on Paediatric Intensive Care Units (PICU) have decreased significantly in 2011 / 2013 when 96% of Children survived their intensive care stay. Mortality during PICU stays has never been lower and stands at 3.7% (Picanet 2014). Picanet data demonstrates a four percent a year growth in PICU admissions above the trend of an increasing birth rate in the UK. The report also identifies changes to ITU admission rates and, significantly for this study, a rising number of children with long-term complex needs (however the report acknowledges further research is required to evidence this).

When considering the rising numbers of Children going home on long-term ventilation, it is important to consider wider societal changes involved. Arguably, the biggest change has been the recognition that life can be happy even with extreme disability (Seear et al. 2016), marking a significant change of approach and upturn in the discharge of highly medically complex Children.

However, ethical decision making, and professional and legal frameworks have not kept up with the rapid changes in the ability to sustain life (Youngblut et al. 1994). Children, who have conditions where death in the first year of life was previously common, such as spinal muscular atrophy (SMA) type one, are increasingly being ventilated despite many reservations about the ethical appropriateness of the use of LTV in such conditions. This concern has centred on the on-going deterioration in muscle tone where children become increasingly locked in (i.e. unable to communicate through any means despite normal brain function) (Roper and Quinlivan 2010). Others have argued, however, that acceptable quality of life and improved physical health can be achieved with the use of LTV (Bach, Gupta, Reyna, Hon 2009). Medical advances in the treatment of SMA type one have occurred during the write up of this study and early trials of new treatment are leading professionals and parents to rethink their views, making this an area of constant debate (Finkel et al. 2016).

Other changes have also been linked to the increasing numbers of Children with complex care needs at home, for example, there are now many more medicines, therapies, and portable technologies to support health care needs at home (Social Policy Research Unit 2003). These changes have enabled more Children with complex care needs to move from hospital to community environments. The ability to keep Children's chests clear from secretions that they cannot clear themselves is an example of this change. Machines that can artificially generate a cough in a Child who has an absent or insufficient cough reflex have become widespread (Insufflation exsufflation machines) (Miske et al. 2004). Vibrating physiotherapy vests have also been widely used to achieve intensive chest clearance on automated cycle's (Miske 2004, Winfield NR, Barker NJ, Turner ER, Quin GL. 2014, Hull, Aniapravan, Chan 2012, Wang, Carsten, Rutkowski, 2010). Both technologies have become smaller and portable over the preceding ten years and are now increasingly common-place in home settings. These treatments are improving Children's quality and length of life and are of particular value to LTV Children.

The expectation of home care is widespread amongst clinicians and the public. The publicity afforded to new resources which have been made available to the National Health Service (NHS) has raised expectations of the level of care that can be provided, whilst not factoring in the increased costs of more complex treatments being required for longer. This expectation sets a context for ever more complex care moving out into the community. A comprehensive literature review was therefore required to explore what is known about the issue's that Children with LTV and their families face; and what makes these care packages more effective for Children and families.

1.2 Identification of Participants

To undertake this study, it required access to a group of Children with LTV and their families to offer them the opportunity to join the study. Although the cohort of LTV Children was growing nationally, the numbers are relatively low as outlined above. An additional complication was that the reasons for initiation of ventilation are diverse. Long-term ventilated Children are commonly classified within three main groups which are based on the underpinning reason for their dependency on LTV; these groupings include conditions which impact upon the central nervous system, musculoskeletal system and respiratory system.

To enable comparison across the participants, a group needed to be selected where there was similarity in their underpinning medical conditions. Three main areas of similarity were identified as important to inform the study questions. The first was that the Children's level of ventilation was similar in terms of their dependence on ventilation. Level three (described in 1.1:3) was identified as the level of ventilation with the highest complexity and severity. The second area of similarity that was identified as important was that there were a significant number of tracheostomy ventilated Children within the group. A tracheostomy is an opening created at the front of the neck, so a tube can be inserted into the windpipe (trachea) to enable easier ventilation. Tracheostomy management is complex in young children, as the small single lumen tubes required for children of this age can easily block and require frequent suction of the tube too keep it clear. In addition, continual observation of the child to keep them safe is required. The complexity of this interface and its impact on care needs were considered important for this study. The final area of interest considered important, was that that group had a significant care package giving similarity of their experience of home life and care packages.

Congenital Central Hypoventilation Syndrome (CCHS) is the largest single group in the tracheostomy-ventilated Children in the central nervous system category of which approximately 55 Children of which around 30 are tracheostomy ventilated. As such, this cohort of Children and their families had significant experience to potentially inform this study. CCHS is a genetic condition which affects the automatic control of breathing. This results in most CCHS children being unable to breath when asleep or unconscious. This necessitates life sustaining ventilation (level three ventilation) when the Child is asleep or unconscious. The condition also affects the Child's ability to increase their rate of breathing during sleep, illness or sport. The condition can also affect other areas of the body requiring autonomic control such as the bowel and heart. A small group will also have learning difficulties; however, this is not a common feature in this group.

Children with CCHS were identified as a priority group to approach as they are almost exclusively level three ventilated and are mostly, in the early years of life, tracheostomy ventilated. Although there is a spectrum of need present in this group, they have in general similar needs with few additional disabilities. This contrasts with the wider group of tracheostomy ventilated Children who often have a multitude of comorbidities and disabilities which create significant differences in their care needs. This group of Children also fit the criteria for children's continuing care (Department of Health and Social Care 2016) so were highly likely to have a care package.

Congenital Central Hyperventilation Syndrome UK Support Group (CCHSUKSG) was approached, via their Chairwoman, to see if they would be interested in hearing more about this study. The group agreed and became the focus of the research and were instrumental in the success of this study.

1.3 Overview of the Study

The literature review (See Chapter Two), critically examined the applicability of the current research base to identify further areas of research required to gain insight into Children's LTV. The literature identifies the devastating impact and restrictions of LTV on initially parents and as they grow older, the children themselves. The literature outlines vividly the life changing experiences families on diagnosis and through their prolonged hospital admission experience from witnessing an average 35 deaths on PICU (Briassoulis et al. 2004) to moving to a new house and having their privacy intruded upon by professionals for a significant part of the day and night. In many ways the literature reviewed was a close reflection of LTV practice and care with a focus on problems and adversity. However, a very small part of the literature identified some different aspects, those of hope and adaption. This review underpinning this study, therefore sets the scene critically to examine the applicability of the current research base, to identify further areas of research required to gain insight into these key areas.

The theoretical approach chapter considers and critiques the formulation and development of the salutogenic theory. Salutogenesis, as an approach, moves research and practice from why people get ill, to how people stay well (Antonovsky 1979). Antonovsky's conceptualisation of the sense of coherence (Antonovsky 1979, 1987) is explored and critiqued. Consideration is also given to the ways salutogenic theory has been used in research and its applicability to Children and qualitative research. Antonovsky's salutogenic theory is considered as a potential way to reflect upon Children's LTV anew. This change of focus is considered in the context of Children with LTV to explore its possible utility in this area. The chapter also explores and critiques the use of appreciative enquire and its theoretical fit with salutogenesis as a novel theoretical mix to add an aspect of visioning to the study.

The methodological approach taken in this study (see Chapter Four) utilised a mix of participatory Arts-based approaches. As a researcher with a background in children's nursing, the researcher was focused on gaining the child's, along with the adult's, perspective. An innovative Arts-based approach including a mix of diary, scrapbook and memory boxes followed up by interviews and focus groups, enabled a rich dialogue from parents, but largely failed to capture the Children's voice which was so important to this study. A novel new approach was therefore developed. This new approach was developed from the technique of vignette. In traditional vignette research, participants comment on a written or videoed scenario. For this study, a novel approach was developed where the Children, instead of commenting on scenarios based on the salutogenic theory and LTV from the outside, became actors within scenarios, through a new Arts-based approach developed for this study of participatory play-based scenarios. By utilising this approach, interesting new data was collected that focused on the child's perspective. Through the process of analysis, the Children's data was considered against, and with the adult data, through an unusual approach of using two different approaches of content analysis, within one analytical framework. This approach utilised conventional and theoretical content analysis approaches. The use of two different content analysis approaches alongside child and parent derived data set enabled triangulation and in-depth analysis of the data.

The findings in Chapter Five shed new light on how Children and their families adapt and live their lives with LTV. The methodological content analysis allowed a deeper depth and understanding of these findings and how they worked together. The findings are presented in the three conceptual areas of the salutogenic theory; comprehensibility, manageability and meaningfulness and identify important new findings in each of the three areas as explored in Chapter Seven.

The discussion (See Chapter Six), critiques and explores these findings in-depth identifying the strengths and weakness of the approach and importantly the applicability of using salutogenic theory throughout the study. Finally, Chapter Seven concludes the study with a consideration of the findings in relation to practice and goes on to consider the gaps and further research that could follow this study.

Chapters are organised within numbered paragraphs to allow easy cross reference between sections to prevent repetition, whilst allowing cross referencing with other linked material. Diagrams are used throughout the study to link ideas in a pictorial manner, alongside photographs taken with permission during the data collection, to illustrate the creative methods of data collection utilised during this study. The appendix contains launch presentations and other material pertinent to consideration of the methodology. This final section is concluded with a draft nursing assessment developed in reference to the findings of this study to promote further interest and discussion around the adapting of the salutogenic theory (Antonovsky 1987) in research and practice in the field of Children's long-term ventilation.

This study arose out of a gap in knowledge around Children's LTV community care provision, borne out of practical experience of working in this field. The unique approach taken in this study enabled a novel approach to be taken to an issue that was having significant impact on Children and families and the teams that cared for them. The findings were enlightening and have significant potential in moving forward future research and practice in this important and rapidly growing area. The following literature review will consider the existing evidence and gaps and areas for further exploration that underpinned this study.

Chapter 2. Literature Review

2.1. Introduction

As set out in Chapter One, the numbers of children and young people (Children) receiving long-term ventilation (LTV) in the UK in their home settings has grown significantly (Wallis, Paton et al. 2010:999). Due to the complexity, intensity and risk involved in this type of home care, Children and families receive nursing care support at home. The levels of support identified in the literature (Margolan et al. 2004) varied from 24 hours a day to a few nights per week with trained healthcare support workers. Such care is referred to in this study as a care package. Despite the significant cost of these care packages, a number of researchers have noted the low levels of satisfaction of families receiving such packages (Dybwik et al. 2011, Fine-Goulden et al. 2015, Kirk et al. 2004). A systemised literature review was therefore required to explore what is known about the issues Children with LTV and the issues that their families face and what makes these care packages more effective for Children and families. This review, therefore, will critically examine the applicability of the current research base to identify gaps and further areas of research required to gain insight into these key issues.

2.2. The Review

2.2.1 Determining the Need for a Review

To understand current research in the area studied, an initial search of systematic review data bases was conducted to identify systematic reviews in the study's area of interest. Three data bases were checked - the Cochrane Database of Systematic Reviews, The National Institute for Health and Clinical Excellence (NICE) and the NIHR Health Technology Program. However, no suitable reviews were identified from these databases. It was then decided that the best approach would be to conduct a literature review using a systemised approach, in order to best study the phenomenon (Grant and Booth 2009).

2.2.2 Process of Review

The literature review was led by the researcher, but as a systemised approach was chosen, quality mechanisms were set in place including drawing on the extensive expertise of the supervision team, a parent representative, and a clinical reference group. The study questions were developed and tested by parent representatives and the final questions are set out below -

1. Does the salutogenic theory offer a new theoretical approach to underpin conceptualisations of the care for children and young people (Children) living at home with long-term ventilation support?

2. What enables Children and families to overcome the complexities that home long-term ventilation brings into their lives?

2.2.3 Developing Criteria

From an initial review of the literature, it was evident that long-term ventilation (LTV) was the most prominent term used in studies covering this study's areas of interest. However, this term on its own did not capture the breadth of research pertinent to LTV Children. 'Technology dependant' is a significant term used to describe Children dependant on long-term ventilation and other medical technologies. This term is also not exclusive; technology dependant, medically fragile, complex care needs and chronic illness (Spratling 2015) are all titles used in the description of technology dependence. To further focus the search, Spratling (2015) offers the following sub classification based on her review of technology dependence, "Technology dependent and complex care needs" (Spratling 2015). Spratling identifies this group and differentiates them from those with "chronic illness, special health care needs, disabilities or the need for assistive devices" (Spratling 2015:648). Further criteria were that the group "may or may not require medical technology" (Spratling 2015:648). This approach was used in the exclusion criteria in the current review.

To gain an understanding about this group the following search terms were developed to focus on the research data most closely aligned to this group.

- Long-term ventilation and Children.
- Technology dependant and Children.
- Complex care and Children.

A search was then undertaken of the UK and international evidence base over the last 20 years [1996-2016]. The review focused on Children's long-term ventilation (LTV), technology dependence and complex care. Seminal literature in this area was collected. The Centre for Research Dissemination guidelines (Centre for Reviews and Dissemination 2009) were used in an adapted form to guide the review. The following exclusion criteria were developed.

- Articles not in English.
- Articles solely or mostly focused on young people or adults 19 plus (defined as 25% or less Children participants).
- Articles over 20 years old.
- Articles focused on hospital care.
- Articles where ventilation was not a significant focus (defined as less than 25% of participants were long-term ventilated).
- Articles that focus on the medical or technical aspects of ventilation at home.
- Articles solely focused on the process of discharge from hospital.

This process was then used to identify potentially relevant evidence around LTV and stakeholder perspectives in LTV and the demographics of this group. The review used electronic, bibliographic, database searching with subject headings and key words as well as hand searches of key journals along with using established public and personal networks.

Databases searched.

- Academic Search Complete Ebsco.
- Allied and complementary medicine databases (AMED) Ebsco.
- Applied social sciences index and abstracts (ASSIA) ProQuest Information and Learning Company. 1987
- British Standards online British Standards Institution.
- CINAHL Complete.
- Cochrane Library.
- Emerald Management.
- Family Health Database. 2013
- Health Management Database. 2013
- Health and Medical Collection. 2013
- Medline.
- MyiLibrary.
- National Library for Health.
- OTseeker.
- PEDro (The Physiotherapy Evidence Database).
- ProQuest dissertations and theses: UK and Ireland.
- ProQuest Nursing and Allied Health Source ProQuest Information and Learning Company.

Box 2.1 Databases Utilised in Systemised Review

2.3 Long-term Ventilation Review Articles

Overview

The initial search after reviewing abstracts produced –

Abstract area	Number of abstracts
Long-term ventilation and Children	23
Complex care	33
Technology dependant	45
Total produced	101

Table 2.1 Breakdown of Abstracts by Abstract Area

As indicated in this chapter, a systemised literature review was decided upon and undertaken to establish what was already known about this group of Children and possible gaps within the current knowledge base. A systematic process was designed to capture, explore and compare and contrast the current literature in reference to the research questions (Centre for Reviews and Dissemination 2009). The first level included the researcher reading each article and a process of critical reading and note taking was undertaken. Key concepts and areas of interest were noted with common and important issues being summarised on a grid. The grid was also used to identify themes of interest. Each theme was then carefully re-analysed to ensure it met the study overall aim and questions using the grid and are presented below.

Abstract area	Number of abstracts
Long-term ventilation and Children	14
Complex care	09
Technology dependant	15
Total for inclusion	38

Table 2.2 Included Studies per Abstract Area

The second level of the review process then included the supervisory team and parent adviser checking that the extractions were fair and representative. They made suggestions which the researcher responded to with reflection and review of the studies to be included.

The final stage involved the researcher re-visiting the extracted literature and synthesising the results together to ensure that a coherent account was the end result.

In addition to the explanation, the process and outputs are demonstrated in diagrammatic format below (figure 2.1).

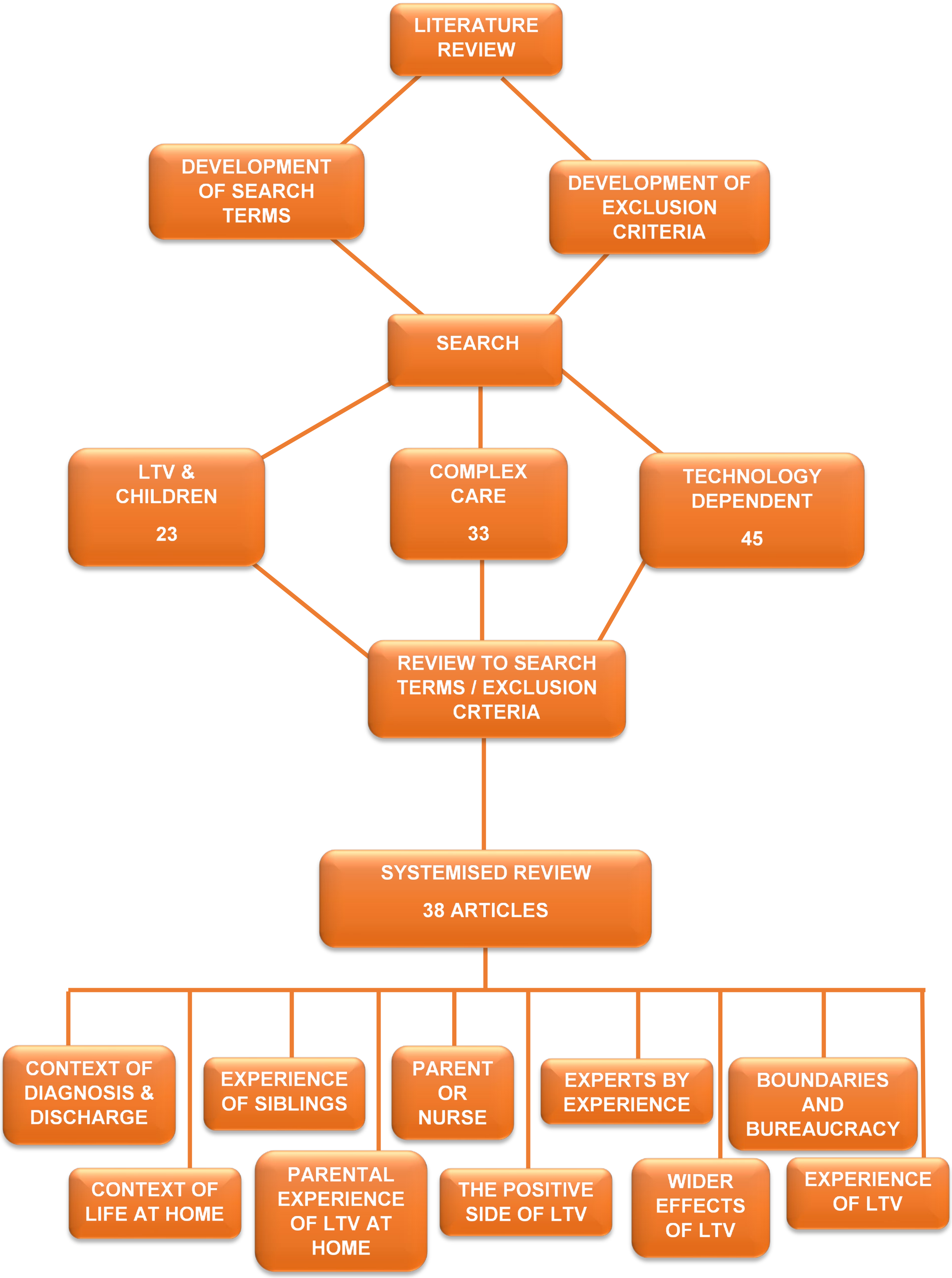
LITERATURE REVIEW
DEVELOPMENT OF SEARCH TERMS
DEVELOPMENT OF EXCLUSION CRITERIA
SEARCH
LTV & CHILDREN
23
COMPLEX CARE
33
TECHNOLOGY DEPENDENT
45
REVIEW TO SEARCH TERMS / EXCLUSION CRTERIA
SYSTEMISED REVIEW
38 ARTICLES
CONTEXT OF DIAGNOSIS & DISCHARGE
EXPERIENCE OF SIBLINGS
PARENT OR NURSE
EXPERTS BY EXPERIENCE
BOUNDARIES AND BUREAUCRACY
CONTEXT OF LIFE AT HOME
PARENTAL EXPERIENCE OF LTV AT HOME
THE POSITIVE SIDE OF LTV
WIDER EFFECTS OF LTV
EXPERIENCE OF LTV

Figure 2.1 Systemised Review Process

2.4 Findings of the Long-Term Ventilation Systemised Review

A total of 38 papers were drawn together. Within these papers 14 were based on data from the UK, whilst 24 were from the USA, Canada, Australia, Sweden, Norway and one from Brazil. The majority of studies (21) adopted a qualitative approach. Overall five studies were literature reviews and the remainder were stand-alone studies. The dominance of non-United Kingdom studies does raise questions around transferability of this data, however due to the low incidence of LTV, to gain an in-depth overview of current knowledge this necessitated that a multi-national approach was taken. During the review a careful focused approach was utilised to consider whether the wider contextual factors were significant, and this was highlighted when the data was used. The greater proportion of qualitative studies was notable and possibly derived from the exclusion of hospital-based studies. In deciding whether these exclusion criteria were trustworthy, reflection on the research questions was considered essential. This reflection took place through the research diary and supervision. It was concluded from this process that the studies selected to answer these particular research questions offered a trustworthy and representative collection of the available studies.

From the literature review ten themes were identified:

- The Context of Diagnosis and Discharge.
- The Context of Life at Home.
- Parental Experience of LTV at Home.
- Parent or Nurse.
- Experts by Experience.
- Wider Effects of Long-Term Ventilation.
- Boundaries and Bureaucracy.
- The Experience of the Ventilated Child.
- The Experience of Siblings.
- The Positive Side of Long-Term Ventilation.

Box 2.2 Themes Identified in Systemised Review

Each of these themes will now be outlined and discussed.

2.4.1 The Context of Diagnosis and Discharge

The literature identified the time of diagnosis and discharge as particularly important to families. The breaking of the news that your Child is going to be dependent on a machine to live is a defining point for families. There is evidence that the news is often not broken well (Lima et al. 2015). This event remains vivid to parents throughout their Child's life. There is some evidence that the way this news is broken, and the way subsequent significant news is shared affects future perceptions and interactions with professionals (Lima et al. 2015, Mendes 2013). The time taken to discharge a Child with LTV averages 9.7 months based on the finding of 39 cases in one large specialist English Hospital (Gilgoff and Gilgoff 2003). The long time spent in hospital (in particular PICU departments) can have significant detrimental effects on a Child. Parents have reported institutionalisation, loss of self-confidence, and behaviour issues (Noyes et al. 1999). The PICU environment is reported by Children themselves to be an environment where death is often witnessed (Noyes 2000). Based on the time of 14.7 months (the time taken to discharge in 1999 for Children ventilated by a tracheostomy), it has been calculated that Children spending all that time on PICU will see or be caught up in the emotionally charged events of 35 deaths. The Children also witnessed 433 Children arriving and going home (Noyes 2000).

A number of studies also noted that parents developed close friendships with other parents whilst their Child stayed on PICU. Whilst this brought benefits, the grief of friends and the mixed emotions of seeing Children and families they knew well going home, could cause trauma for them and worry about the psychological effect on their own Child. The detrimental effect of prolonged stays on PICU has been recognised and some progress has been made in speeding up the discharge process. The difference between these two cited estimations of time on PICU could show a quickening up of discharge and increasing use of transitional care units and high dependency units (HDU) to take a step down from the PICU environment (step down in a hospital environment is the process of supporting an LTV child on a HDU or ward prior to discharge to enable them to get used to lower support levels). Such a conclusion needs to be treated with caution as these figures were collected using slightly different approaches. For many Children and parents/families their PICU stay is prolonged and traumatising. For most Children dependant on LTV, this is not their last visit to PICU with readmission rates for LTV Children in one English centre calculated at 1.7 per year (Gilgoff and Gilgoff 2003).

The time in hospital instead of a time of preparation, planning and learning can be a time of negative experiences. In one study, only one in nine families felt involved in the discharge process with many feeling like bed blockers (Evans 2014). Studies have identified that bed availability in PICU is limited by medically stable LTV Children awaiting discharge (McDougall et al. 2013). This has led to national initiatives to reduce perceived bed blocking including speeding up of the discharge of LTV Children. The monitoring and need for beds created by these initiatives is apparent to parents and can make them feel unwanted. Some parents felt it was assumed by professionals that they would take on the highly complex care their Child required. Further, they felt this was not discussed openly, with the desire to discharge home being central to professionals (Kirk and Glendinning 2003).

There are many reasons that are cited in the literature as to why Children dependent on LTV are delayed in getting home (Noyes 2002). The National Framework for Children's Continuing Care (2016) sets a four-week time frame from referral to a funding decision for home care packages. However, there are anecdotal accounts that few organisations achieve this timeframe. The recruitment and training of staff is complex and can take many months (Margolan et al. 2004). The issues involved in terms of housing are often complex and time consuming, to resolve these issues will now be outlined and considered.

There is no specific literature defining what space is required in a family home to accommodate the child's care needs, equipment, and supplies. Work undertaken in hospital design for single rooms indicates that a three-meter square room is needed to fit the adapted bed, carer, and space to provide care (NHS Estates 2008). The space to store consumables and equipment is significant and intrusive in the family home (Jardine et al. 1999). Many homes therefore are deemed unsuitable or require major adaptions. Availability of adapted (or adaptable) council housing is limited, and private rented homes lack the security for long-term care. There is often a reluctance to allow adaptations of the property to take place or for local authorities to fund adaptations in the private rented sector (Wilson 2018). For families owning their own home they may have to upsize both their home and the mortgage used to pay for it at a time when they may be on sick leave or one partner may have left work to care for the Child on PICU (Edwards et al. 2004). However, it is not just a practical issue of moving or home modifications. This is a time of the family home changing into a medicalised environment. During this long process, hope has been identified in research as a tool parents of LTV Children use, to get them through these difficult times. However, a seemingly indefinite process can make hope difficult to maintain (Wilson et al. 1998).

Hospital is for many families, where all the teaching required to look after their Child took place. The taking on of the care of their Child with so much additional technology can be a daunting task for parents, requiring them to accept great responsibility and the daily possibility of their Child's death (Gilgoff and Gilgoff 2003). However, despite all the risks involved in ventilation at home, accidental death is rare with one UK centre identifying seven deaths out of 39 but none due to home care issues (Gilgoff and Gilgoff 2003). The training of parents' pre-discharge was variable from some parents reporting feeling competent, as well as confident, to deliver care, with others feeling unprepared when they got home (Coad 2013). In the best examples, there were follow up visits at home, but for the majority there was no ongoing training, while they faced the complexities of adapting their learning which had taken place in PICU to the context of life at home (Lima et al. 2015, Wang and Barnard 2008:506). These two studies are from different health care systems but do seem to reflect the mixed experience reported in UK studies. Other studies have reflected on the lack of psychosocial elements to training (Hewitt-Taylor 2004, Kirk and Glendinning 2003), which may in part explain difficulties in adapting training given in the clinical PICU environment to the very different ongoing and changing reality of life at home.

The experiences from diagnosis and the complex agreements and arrangements required to leave PICU have been described as setting the scene for the ‘fight for home’. Many families felt they had to fight to get their Child ventilated and this continued into getting home. Fighting for a care package, housing, equipment, and benefits can become daily life. This sense of the battle for home is a strong feature in the literature (Alexander et al. 1999, Dybwik et al. 2011, Noyes et al. 2002). The feeling of having to fight, and a context of needing to get home, set a background for the high intensity issues and relationships which will continue out into the home and care package. Some parents developed their approach to the fight with a persistent assertive approach being identified as most effective (Wilson et al. 1998). Others, however, remained angry or frustrated with the process of getting home.

There is evidence of reduced Quality of Life indicators in both Children and parents who live with LTV (Noyes 2006, Redouane et al. 2016). The diagnosis and time pre-discharge is significant both as an event and how its experience contextualises life at home. However, as LTV continues for many years at home, life experience at home will gradually take precedence in determining quality of life.

2.4.2 The Context of Life at Home

Research in the home environment has identified the environmental impact of LTV. The dependence of a Child on ventilation and the staff, equipment and supplies that are needed to support a ventilated Child at home has been described as transforming the home into a “mini hospital” (Kirk et al.2004). All parents in one study noted the volume of equipment and how it took over and changed their home (Kirk et al. 2004). Parents attempted to integrate or conceal equipment to normalise their home (Gilgoff and Gilgoff 2003) but the changing of the home space was compounded by the amount of time health care staff were present. The presence of health staff has been linked to the loss of privacy, a life full of relative strangers and the opening of a family’s private space to observation and judgement (Fine-Goulden et al. 2015). Parents have described this as turning their home into a “public place” (Kirk et al. 2004:460). However, many families through negotiation and complex reorganisation managed to return this modified space back to a place that had personal meaning to them (Gilgoff and Gilgoff 2003).

2.4.3 Parental Experience of Long-Term Ventilation at Home

The literature has identified the scale of transformation LTV brings into a family’s home life. Once home, the reality of life at home rapidly becomes apparent. The reality of how different their Child and therefore their life is going to be becomes clear. This period has been described by families as getting over the hump (Alexander et al. 2002) which describes the overwhelming change, challenges, and adjustments they must make in these early months at home. The setting of a new reality involves the creating of schedules and routines of care, allowing little time for relaxed family life (Wilson et al. 1998). The fight remains a significant theme in the literature with families fighting bureaucracy, resources and care provider-imposed limitations in terms of managing their Child's care (Alexander et al. 2002, Dybwik et al. 2011, Whiting 2012). Interestingly this data comes from three different studies in three different countries. There is evidence that there is an unfillable gap between parental expectations and what community providers can provide even with unlimited resources (Dybwik et al. 2011). An unfillable gap appears to lead to conflictual relationships, poor relationships with local care providers and sometimes led to disengagement or depression in the family. Studies have therefore shown reduced resourcefulness in families and increased markers for the risk of depression. Mothers' levels of depressive symptoms have been shown to reduce family functioning and were found to be double in mothers of LTV Children compared to the general population (Toly et al. 2012). Physical health indicators have also been found to have been affected. This was exemplified in one Canadian study which identified 50% of parents of ventilated Children had chronic health conditions and 25% rated them as severe (Seear et al. 2016).

The primary provider of the technological care Children require at home has been identified as mothers (Fernanda de Lima et al. 2015, Heaton et al. 2005) with just 8.35% of mothers in one study being in any type of employment compared to 21.30% of the fathers holding down a job. Fernanda de Lima et al. (2015) identified this trend as a disinterest in fathers being involved in care of the LTV Child. (This study however was conducted in Brazil and could reflect gender and cultural beliefs). Interestingly, UK studies provide evidence of more involvement of fathers in care, however, they do show males continuing their full-time work patterns. Mothers, however conversely often identified that they were unable to work. The main issue stopping them finding employment appeared to be the difficulty of finding a job which would be flexible enough to fit the complex care and supervision requirements their Child with LTV needed (Heaton et al. 2005).

Families with Children who have a dependence on LTV, have been observed as striving to achieve as much normality as possible to enable their Child to achieve the best they can. When this was achieved, life started to make sense. However, the instability of many Children with LTV and complex needs meant that families continually had established routines destabilised. This instability unsettled or derailed their attempts to achieve a sense of normality for the Child and wider family (Alexander et al. 2002, Gilgoff and Gilgoff 2003).

2.4.4 Parent or Nurse?

Within the literature it is evident that the performance by parents of invasive care tasks normally carried out by nurses in acute settings can have significant ramifications for parents. Parents moved from providing care and nurture to being the person carrying out an unpleasant or painful procedure. This transfer of role, even for some of the day, made some parents question their whole contextualisation of parenting. Parents wanted to be seen and defined as parents, not as clinical staff (Kirk et al. 2004). The more complex and unstable the Child's condition the more clinical tasks infringed on every parenting experience (Kirk et al. 2004). This context meant that when parents performed a clinical procedure it had a different meaning from a nurse carrying out the same procedure. As stated by Kirk et al (2004:462), "this process of transferring technical care has been described as so significant that it alters the meaning of parenting itself" thus, creating difficulty for parents in identifying their social identity (Wang and Barnard 2008:506).

2.4.5 Experts by Experience

In six of the studies reviewed parents saw themselves as experts in their Child's care (Heaton et al. 2005, Kirk et al. 2004, Kirk and Glendinning 2002, Kirk and Glendinning 2003, Maddox and Pontin 2013, Mendes 2013). It has been identified that parents, due to this expertise, expected nurses and professional carers to acknowledge their skills and to respect their wishes (Mendes 2013). Research with health professionals offers some support to this finding with the identification of parental expertise honed by experience being considered and recognised by professionals as being greater than their own (Kirk et al. 2004, Kirk and Glendinning 2002). Parents valued this recognition, but when this expertise was challenged or ignored, there was a high risk of a conflictual relationship developing (Kirk and Glendinning 2002). Parents felt a key nursing role is to support the strategies parents use (Kirk et al. 2004). Research has reported that the parent's personalisation of accepted practice had in some cases moved away from conventional norms (Maddox and Pontin 2013) making professional challenge more likely. Where these disagreements were present, trust was likely to be undermined affecting the relationships between the parent and care or professional team (Maddox and Pontin 2013). Conversely, trust and openness about professional skills deficits, and positive feedback on the care of their Child, enhanced relationships (Kirk and Glendinning 2003). The need to remain in charge of care continued back into the acute hospital when the Children were readmitted. Parents felt that their Child's complex care was provided better by themselves, due to a lack of skills and understanding of the child's care regimen by ward staff and inadequate staff cover (Heaton et al. 2005).

2.4.6 Wider Effects of Long-Term Ventilation

The literature has also identified wider detrimental effects of LTV. The demands in terms of time required to look after LTV Children has been studied in detail by Heaton (2005:448). Heaton (2005) identified not only that extensive time was required to deliver the care but also that the nature and timing of this care resulted in “difficulties for families, limiting their inclusion in school, work and social life in general” (Heaton 2005:448). Another study identified that social isolation can be compounded by people’s reactions to complex medical technology and procedures and lead to frustration and a feeling of being outside society (Toly et al. 2014). In a study of technological care (Kirk et al. 2004) found that the continual and ongoing process of care going on in the home, with high levels of professional support in the presence of friends and relatives could medicalise relationships and alarm friends and relatives, leading to changes within the social relationships of the family. Studies have identified a gradual reduction of family and friendship group support in families with a Child who requires complex technological support (Kirk et al. 2004). Similarly, Edwards et al. (2004) has described the profound isolation experienced by Children with LTV (Edwards et al. 2004) (however caution needs to be taken with this finding as it was undertaken in an American rather than UK cultural environment). One study noted that whilst increasing nursing support might increase ‘contact’ between families, Children and the outside world, it did not tend to fill the gap in a meaningful way and could have unintended additional complexities (O’Brien 2002). For example, greater levels of home care nursing have been linked with declining normalisation scores of parents, defined in (Toly et al. 2012) paper as the incorporation of a complex condition into day to day life created isolation. This isolation was felt most by mothers who did not work (Gilgoff and Gilgoff 2003).

2.4.7 Boundaries and Bureaucracy

Within a number of the studies explored it was clear that the ability of parents to take control of their Child’s condition and their family lives was often hampered by bureaucracy and rules of health providers supporting them at home (Dybwik et al. 201). This has been described as “threatening parenthood” by Lindahl and Lindblad (2013:495). Parents were extremely concerned by a lack of involvement in the care staff recruitment process since it was possible that without their input, carers could be recruited who did not fit the family’s views or values (Lindahl and Lindblad 2013). Indeed, in the study by Kirk et al. (2004) it was identified by parents that it was important for care staff to demonstrate genuine affection for their Child. Conversely, it is often acknowledged that care provider boundaries are created to prevent professional carers becoming over involved with individual Children.

A further aspect of this theme is reflected in studies by Lindahl and Lindblad (2011) and Kirk and Glendinning (2003) who have identified that the medicalisation of care can infringe on family life, and the importance of adapting hospital care routines to the home environment. Health care professional's visits were not adapted to daily life, often excluding the Child from activities they wanted to take part in. The lack of coordination between professionals compounded this leading to parents often having to take a lead in the coordination of professional care provision. The structure of home care providers has been described as "seldom designed to achieve what children wanted" (Lindahl and Lindblad 2011:250). In a study by Kirk and Glendinning (2003) the complexity of community services, overlapping roles of professionals and a lack of clarity around responsibilities led to confusion and a sense of being overwhelmed by professionals. Clear responsibilities and a single lead professional helped clarify and coordinate professional support and was considered as positive by parents (Kirk and Glendinning 2003).

2.4.8 The Experience of the Ventilated Child

A re-occurring theme across the studies highlighted that the experience of Children who require LTV is significantly more positive than their parents. In one Canadian study the adverse effects of home LTV were scored as mild by Children but moderate by parents (Seear et al. 2016). However, health related quality of life scores among LTV Children have been found to be significantly lower than comparator groups of school Children and Children with complex needs (Noyes 2006). Noyes (2006) examined Child and Parent derived quality of life scores and identified a crossover except in her category of "disease and relationships with friends" where the parents scored lower than the Child. More recent studies have found similar quality of life scores, but have only focused on parental perceptions of a Child's quality of life (Redouane et al. 2016), which, in the context of Noyes (2006) study needs to be treated with some caution.

As discussed earlier, (Sudbury and Noyes 1999) time spent on PICU can be traumatising and research with Children who had been long-term hospitalised, described issues of bonding with parents and care teams due to parental absence or limited uninterrupted time with parents. In terms of nurses, the number of nurses encountered, and the efficient performance of nursing care, regimens did not facilitate meaningful relationships with Children (Noyes 2000). Another barrier related to meaningful relations with Children pertained to communication. Communication issues enforce dependency, stifling a Child's attempts to let their views be known. This issue was not focused on in hospital leaving Children feeling isolated and unable to interact with staff. Overzealous health and safety restrictions was noted to restrict Children's freedoms and an absence of meaningful education and rehabilitation activities were apparent and had far reaching effects on Children which were evident long after discharge (Noyes 2000).

One notable criticism is that little research has been undertaken to establish the voices of long-term ventilated Children within the context of their daily lives at home. One American study

(Sarvey 2008) took a phenomenological approach and sought the first-person experience of Children at home. The study identified Children's experiences of living with ventilation. The "other" described the continual presence of a skilled care giver who was primarily a mother or a nurse and the "mechanical other" care giver was the ventilator which most of the Children were positive about as it made them feel better. However, the Children identified that they were never alone - they were always with the mechanical, and normally the human other (Sarvey 2008).

Some Children noted the freedom they could experience if in the care of experienced others on a camp for ventilated Children or other experiential settings (Sarvey 2008). However, in another study, some Children were noted as not wanting to go on another weekend in a school setting as they would feel different to their school peers (Gilgoff and Gilgoff 2003). Children noted and were distressed by the reactions of others; particularly other Children. Some developed ways to respond, others struggled more (Sarvey 2008). Restrictions of care were significant and a can-do approach of doing it differently was valued. The need to be normal like other Children was strong, sometimes resulting in resentment of siblings and an overwhelming desire to normalise care (Sarvey 2008).

2.4.9 The Experience of Siblings

Not surprisingly, given the context of LTV described above, siblings have also been identified as having significant difficulties in adapting to the medicalisation of home life. A number of studies have detailed how protracted discharge processes resulted in siblings being apart from at least one parent for significant periods of time. In addition, the workload involved in LTV care results in siblings becoming involved in significant amounts of family support including housework, laundry, and preparing meals. Care often included caring for other siblings and for older siblings that included getting them to school, or social activities. Care for many Children included caring at some level for the Child with LTV. In one study (Heaton et al. 2005) it was found that an older sibling provided respite when the parents were away. The consequences of providing this care included reduced attendance and academic performance at school or college. Siblings were also exposed to the stressful environment of complex technological care created in the home environment (Heaton et al. 2005). Tension exhibited between parents could be difficult, distressing and personal space was frequently infringed. The unpredictability of life due to the instability of the effected Child, therefore, had a significant effect on siblings (Heaton et al. 2005).

This pressure has raised concerns around the effect on sibling relationships. One Brazilian study noted a parent's distress where a sibling told her he didn't like his brother anymore (Fernanda de Lima et al. 2015). A Canadian study supported this finding by identifying parental concern about the resentment of siblings (Gilgoff and Gilgoff 2003). Studies conducted directly with siblings however, have noted siblings being ambivalent about their life situation (Lindahl and Lindblad 2011). There is some support for siblings, both in local authorities and children's hospices, with many UK children's hospices having developed support programs for siblings (Dempster, Cooper, Petty 2011). Local authorities and community charities for example have developed young carer activities which have been identified as helpful by siblings (Lindahl and Lindblad 2011). Care staff training around siblings and the issues they face has also been identified as a potentially helpful strategy (Hewitt-Taylor 2004).

2.4.10 The Positive Aspects of Long-Term Ventilation

The burden and complexity of long-term ventilation (LTV) however, is only part of the picture identified in the literature. Living with a Child dependent on long-term ventilation is described as "daily living with distress and enrichment" (Carnevale et al. 2006). To conceptualise life with a child with a ventilator as entirely negative was challenged in a small number of studies. Parents have reported having a good quality of life. There is pride and joy on their Child's achievements (Trowbridge and Mische-Lawson 2014). The drive to achieve the best for their Child is immense and has been described as "absolute involvement" (Wilson et al. 1998). The positive side of the LTV experience for parents has also been linked with parents noting personal growth, identifying that they feel they are better people with a can-do attitude, more empathy and more personal resilience (Wang and Barnard 2008:506). Children who are LTV have reported feeling better and more able to do things they want to do (Sarvey 2008). However, this positive story comes at a cost which the LTV literature is mostly focused upon. This deficit focus has been described as overwhelming and leading to an incomplete picture (Trowbridge and Mische-Lawson 2014). However, this conclusion was based on a purely quantitative review of the literature. The qualitative body of research does identify positives, as explored above, but this review has identified this research to be limited and lacking in breadth in this area.

The next chapter will explore salutogenesis and the salutogenic model's approach to health, to determine if a different approach could be used to refocus research with Children with LTV and their families (and to strengthen the knowledge base in this area of study). The literature review described above found no studies fitting the inclusion criteria for this study that have focused on salutogenesis or the salutogenic model.

2.5 Conclusion

The literature review overall highlighted that the use of LTV in Children is growing rapidly (1.1). There is evidence that the consequences of this highly complex type of care on Children and families is immense. Quantitative and qualitative studies have identified and explored these issues in depth. However, there is very little published around how to make services more effective for this group of Children and their families. Moreover, the incidence of LTV in Children is growing rapidly (1.1); without urgent action families, Children and services face being overwhelmed by the complexity and need created by LTV.

There is an interesting area identified (but not generally focused upon), around how Children and families get on and thrive despite the complex issues that they are faced with and what helps them to do this. This area is seldomly considered in the LTV literature. This gap is important to understand given the low levels of satisfaction reported by families around packages of home care (Dybwik et al 2011, Fine-Goulden et al 2015, Kirk et al. 2004). Rectifying problems identified in the literature, although helpful, is in the light of the limited data around the positive experiences of Children and families despite LTV, unlikely on its own to improve reported satisfaction of families. What may be required is a different approach altogether. A change of focus onto what Children and families consider important is urgently needed by Children, families and services alike. The study presented in this thesis will therefore aim to refocus the debate on precisely this area.

Chapter 3. Underpinning Theoretical Frameworks

3.1 Introduction

As identified in the proceeding systemised literature review, a significant gap in the understanding around the role of positive adaption of children, young people (Children) and their families to LTV at home is apparent in the current literature. This chapter explores the potential of the writings of Aaron Antonovsky (1979, 1987) and his concept of 'The Salutogenic Theory' to adapt for a study in this area. This is believed to be unique. As a result, it is thus pertinent to thoroughly explore the origins of his work using Antonovsky's original and now seminal literature on salutogenesis to underpin and critique this approach. The chapter will therefore commence with a theoretical review and critique of Antonovsky's two books and the wider writings that have emerged around Antonovsky's salutogenic theory (Antonovsky 1979, 1987). The chapter will then explore in depth, the applicability of this approach within this study by progressing on to systemised review and critique of qualitative salutogenic approaches in research studies focused on Children and their families.

3.2 The Beginnings of Salutogenesis

Aaron Antonovsky was a medical sociologist/anthropologist who had an interest in stress theory. Whilst undertaking research into women's adaption to the menopause in different ethnic groups of women living in Israel in 1970, he made a discovery that led to the creation of salutogenesis and the salutogenic theory. A group in the study was made up of European women who would have been 16-25 years of age at the start of World War Two. One of the questions asked of the group was "were you in a concentration camp?". Of the group who answered yes more than a few women among the concentration camp survivors were deemed to be well adapted "no matter how adaption was measured" (Antonovsky 1979). Despite going through the most unimaginable horror of the camp and being displaced from their home countries, as well as relocating to a new Israeli state and going through two further wars, these women were healthy, happy, had Children and were in good community and friendship groups (Antonovsky 1979). This finding fascinated Antonovsky who then went on to consider "The Origins of Health" (Antonovsky 1979).

3.3 Health, Stress, and Coping: Antonovsky 1979

Health, Stress, and Coping was Antonovsky's first seminal piece of work on salutogenesis, published in 1979. This book describes the concept of salutogenesis, which is described as the study of the origins of health and what enables people to be well (Antonovsky 1979). The word salutogenesis was formulated by Antonovsky from two Greek words genesis-origin and salutehealth (Antonovsky 1979:vii). Whilst a more positive approach to human health was not new or unique (as there are traces of this approach noted back to Aristotle), (see Mittelmark et al. Eds. 2017), what was new was a complete concept that acknowledged the ever-present threat from stressors, but approached the problem from a different direction to the dominant pathogenic approach (Mittelmark et al. Eds. 2017). Instead of a focus on what makes people ill, the focus for Antonovsky was on what makes people well.

Antonovsky did not see people as well or ill. He saw health as a dynamic process “along a healthy/dis-ease continuum” (Antonovsky 1996:14), along which people move during their lives. Antonovsky had explored numerous health data which he claimed supported his view that a third of the United States of America’s population (or more) were ill at any given time. Therefore, he argued that the pathogenic approach of a simple split of well and ill, does not work when such a large part of the population is sick at any given time. Antonovsky argued that decline into illness is inevitable and trying to study individual causes for illness or disease could never cover the vast complexity and interplay of illness. The issues which needed studying from Antonovsky’s perspective were those factors that cause health rather than those which cause specific diseases (Antonovsky 1979:16). Antonovsky also noted that the public’s perceptions of how healthy they were, was not in line with the pathogenic approach (with 87% of participants responding to the National Health Interview Survey) (1973, cited in Antonovsky 1979:34), rating themselves in good or excellent health. This paradox interested Antonovsky (despite acknowledging the weakness of this type of survey) and added to Antonovsky’s evidence that a different approach was required.

Salutogenesis has been described as a unifier of this type of, health rather than illness, focused approach into a single classification, Antonovsky wanted to move beyond the healthy/sick debate but still used (although hyphenated) the word dis-ease to describe the low health side of the continuum. Some have argued that a different term such as quality of life would have differentiated salutogenesis better (Strumpfer 1995 cited in Bonmatí-Tomás 2016:3). The use of a pathogenically related word (dis-ease), even hyphenated, has encouraged some to locate Antonovsky’s salutogenic theory (Antonovsky 1979, 1987) as the opposite of pathogenesis (Mittelmark et al. Eds. 2017). Antonovsky, however, viewed salutogenesis as a different concept rather than an opposite pole. Further, Antonovsky’s description of health, as a continuum with an emphasis on the positive pole, has led some advocates of the positive health approach to health to link the sense of coherence (SOC) to the theories of positive health (Mittelmark et al. Eds. 2017). Conversely, Antonovsky disputed salutogenesis being clearly located as a positive health approach, as in isolation positive health lacks the dynamic reference to the stresses present in life which is a key component of salutogenesis (Mittelmark et al. Eds. 2017:36). The following table compares the pathogenic and salutogenic approaches;

Pathogenesis.	**Salutogenesis.**
Start Point = Disease or Problem.	Start Point = Health Potential.
About avoiding problems and its causes.	About approaching potential and its causes.
Works to eliminate risk factors.	Works to create health (salutary) factors.
Reactive - react to signs, symptoms, and indications of disease.	Proactive - create conditions of physical, mental, and social well-being.
Disease or infirmity is an anomaly.	Humans flawed and subject to entropy.
Idealistic perspective - treat disease.	Realistic perspective - go get health.
Focus is to prevent pain or loss.	Focus is to promote gains or growth.
Prepares or helps prepare one to live.	Enhance capacities and potential so can live fully.
Wants to help avoid or prevent a person from being pushed backward.	Wants to help or enhance a person's ability to move forward.
Against Disease and infirmity.	For Health.
For those who need healing cures.	For those who want better health.
Primary focus - Prevention of negative health.	Primary focus - Promotion of positive health.
Secondary benefit - Health Promotion.	Secondary benefit - Prevention of disease and infirmity.
Outcome - absence of problem.	Outcome - presence of a gain.
Keep from making situation worse.	Continuous Improvement.
Minimisation of problems.	Optimisation of potential.

Table 3.1 Perspectives on Health (Becker et al 2010:32)

From the pathogenic perspective, Antonovsky's approach could be critiqued as non-scientific, as it originates from theoretical and qualitative research rather than double-blind research trials. The focus in the pathogenic approach, however, is reductionist. The search is for the answer to a hypothesis and the aim is to find a concrete outcome, such as a course of illness or a cure or treatment for a particular disease. In salutogenesis, the focus is not on illness, but focuses on the story of the whole person and an effort to move all people towards the positive end of the health continuum. In pathogenic terms, such a goal is too wide and therefore is not measurable (which could be viewed in pathogenic terms as unscientific).

Salutogenesis presents an orientation towards looking for what makes people well and enables them to overcome adversity. This coupled with a belief of a continuum of health between ease and dis-ease is the approach of salutogenesis, which involves looking for health assets or ways to help a person move positively on the health continuum. Antonovsky credits a large number of theories around stress and coping in enabling him to develop the idea of salutogenesis. Salutogenesis is commonly considered as a classification of these types of approaches and is depicted below:

Figure 3.1   Salutogenesis an Assets Approach Reproduced in Mittelmark (2017)

3.4 Salutogenic Theory

Antonovsky (1979) developed the salutogenic theory as his answer to the question posed by salutogenesis (what the origins of health are?). Within this theory, he described some of his previous research into the way people cope with stress as an element of salutogenesis (An example of this was the idea of generalised resistance resources). Antonovsky also described his new concept of the sense of coherence (SOC) which will be explored further below. This section will explore how the existing elements of salutogenesis and the SOC came together in *Health, Stress and Coping* (Antonovsky 1979) to create the new theoretical framework of the salutogenic theory.

3.5 General Resistance Resources

In looking for health assets which move a person towards the “ease” end of the health continuum or at least maintains them on their current location, Antonovsky’s work on stress led to the creation of the idea of generalised resistance resources (GRR). A GRR according to Antonovsky is “any characteristic in the person, the group or the environment that can facilitate effective tension management” (Antonovsky 1979:99). These resources and the extent that they are available to a person, (if the person has the ability to select and use them effectively), will move them along the health continuum towards the ease end. A GRR can be physical, biochemical, artificial, cognitive, emotional, a value, interpersonal or microsocial (Antonovsky 1979:103). Antonovsky theorised that a GRR worked by making sense out of the continual onslaught of stressors, which was different to the pathogenic view which contends that stressors affect the body and make a person ill. Antonovsky conversely theorised that stressors can be negative and positive. What mattered was how they were perceived and how able a person was to muster a GRR or a number of GRR to stop or make sense of the stressor.

3.6 The Sense of Coherence

The sense of coherence (SOC) was to Antonovsky the core of salutogenic theory. The ability to effectively use GRR to move positively on the continuum, Antonovsky believed, comes from an innate SOC. The SOC is theorised as coming from a loop where positive feedback comes from successful use and accumulations of GRR which in turn lead to an on-going belief that when affected by a stressor, things will work out as well as could be expected. This belief then makes even complex new situations or stressors seem understandable or at least possible to overcome. This to Antonovsky explained the remarkable resilience of the twenty-nine women identified in his 1971 study.

The SOC in *Health, Stress, and Coping* (Antonovsky 1979) was defined by Antonovsky for the first time as a “global orientation that expresses the extent to which one has a pervasive, enduring, though dynamic feeling of confidence, that one’s internal and external environments are predictable and that things will work out “as well as can be reasonably be expected” (Antonovsky 1979:123).[1]

Whilst inviting others to critique, for Antonovsky the SOC provided the single central answer to the question “what causes health”. This clear direction in his writings (including the last before his death, Antonovsky 1996) set the focus of research into the salutogenic theory on the effect of SOC rather than its conceptual existence. This has led to little challenge or critique of Antonovsky’s belief that the SOC is the answer to the salutogenic question of what causes health (Mittelmark et al. Eds. 2017)**.**

3.7 Contextualising Salutogenesis, the Salutogenic Model and the Sense of Coherence

The terms salutogenesis and sense of coherence (SOC) have become undifferentiated in much of the literature. Salutogenesis is the approach of investigating the causes of health, the health continuum, and a view that movement along the continuum is determined by the successful use of health assets. The salutogenic model is Antonovsky’s answer to the salutogenesis quest to find what causes health. In the salutogenic theory assets are “generalised resistance resources” (GRR) and the way they are made available and used effectively by a person is related to their SOC.

3.8 Unravelling the Mystery of Health, Aran Antonovsky 1987

Antonovsky undertook qualitative research to further develop the salutogenic theory. The revised theory was expanded upon and published in ‘*Unravelling the Mystery of Health’* (Antonovsky 1987). To develop the theory, Antonovsky (1987) undertook unstructured interviews with 51 people described as having undergone severe trauma, ranging from being in a concentration camp to severe disability. All of them were thought to be functioning remarkably well by the referring person.

The age range of the participants was between twenty-one and ninety-one and this group also included four young people. Thirty respondents were men and 21 were women. The participant’s country of origin included 12 from Israel, 19 from European countries and 20 from North African and the Middle Eastern countries. There was a mix of family status and occupations ranged from unemployed to senior official/professionals. Fifteen were from the highest economic groups (but this may come from the bias of the referees, a point which is

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[1] Within this definition for the first time two of the conceptual areas of comprehensibility and manageability which were to become clearly articulated central conceptual components in the final version of Salutogenic theory published in *Unravelling the Mystery of Health* (1979) were identifiable.

acknowledged by Antonovsky, 1987:65). The interview was very loosely structured with only one question being asked, "Tell me about your life?", (Antonovsky 1987:65) making for an explorative qualitative research approach. The respondents were interviewed in their own homes, helping to gain relaxed respondents. All interviews were written up verbatim after the interview. Considering most of these interviews went on for one hour this does pose some questions of trustworthiness. For example, Antonovsky could have had clearer recollection of the interviews that fitted his hypothesis as they would be more memorable.

From this research, the two main concepts of the SOC (coherence and manageability), identified in the original definition in *Health, Stress, and Coping* (Antonovsky 1979) were added to by a third concept "meaningfulness" (which had only been mentioned in passing in *Health, Stress, and Coping*, Antonovsky, 1979). Meaningfulness thereafter became seen as one of the three central components of the SOC. The expanded and final underpinning concepts of the SOC were therefore:

Comprehensibility: – A person with a high SOC sees confronting stimuli as making sense, in that they will be expected or if unexpected, they will be ordered or explicable. This includes undesirable stimuli such as death of a loved one or war (Antonovsky 1987:16-17). .

Manageability: – "the extent to which one perceives that the resources at one's disposal are adequate to meet the demands posed by the stimuli that bombard one" (Antonovsky 1987:17). This may include resources of the family, friends, or spiritual resources.

Meaningfulness: – A person with a strong SOC spoke of areas of their life that were important to them, that they were very much cared about and that made sense to them. Antonovsky saw meaningfulness as the motivational component. This component also includes the significance of being involved in decisions. In addition, it encompassed the emotional component with a link to "life making sense emotionally" (Antonovsky 1987:18).

Antonovsky (1987) pulled these three concepts together into an updated definition of the SOC - "The SOC is a global orientation that expresses the extent to which one has a pervasive, enduring though dynamic feeling of confidence that (1) the stimuli deriving from one's internal and external environments in the course of living are structured, predictable and explicable; (2) the resources are available to one to meet the demands posed by those stimuli; (3) those demands are challenges, worthy of investment and engagement". (Antonovsky 1987:19). All component parts are closely intertwined but Antonovsky did speculate that people would have different combinations of each of the three components. Antonovsky identifies these different possibilities and speculates on the long-term outcome on the sense of coherence (see table 3.2).

Antonovsky (1987:21) went on to note "the three components of the SOC, though all necessary are, of unequal centrality. The motivational component of meaningfulness seems most crucial"

(Antonovsky 1987:22). The importance of meaningfulness was identified in triggering change "which leads one to seek to order the world and to transform resources from potential to actuality" (Antonovsky 1990:79). Despite the addition of meaningfulness however, and his growing sense of its importance to the SOC, Antonovsky maintained the written ordering of the component part of the SOC as comprehensibility, manageability, and meaningfulness.

The linkage between these three components in terms of SOC is summarised in the following table, which identifies the importance of meaningfulness in maintaining or increasing a person's SOC.

The following table (3.2) considers all the possible combinations of Antonovsky's three concepts when scored using the SOC questionnaire (described in 1.7) consider how they work together and the overall predicted outcome.

TYPE	Comprehensibility	Manageability	Meaningfulness	Prediction
1	High	High	High	Stable
2	Low	High	High	Rare
3	High	Low	High	Pressure to move up
4	Low	Low	High	Pressure to move up
5	High	High	Low	Pressure to move down
6	High	Low	Low	Pressure to move down
7	Low	High	Low	Rare
8	Low	Low	Low	Stable

Table 3.2 Dynamic Interrelatedness of the Sense of Coherence Components Antonovsky (1987)

3.9 Development and Sense of Coherence

Antonovsky (1987) theorised that the development of a person's SOC start from birth with the environmental, social, and psychological factors that the baby, infant, child and adolescent experience. These factors will shape their SOC. Antonovsky went on to consider how each of his three concepts would develop through childhood and adolescence which is summarised below.

Comprehensibility develops if the child starts to see the world and the daily bombardment of stimuli and stressors they experience as consistent and routine. However, the child can only find this world if it is there to find. Constantly changing or contradictory expectations will undermine a child's growing comprehensibility.

Manageability comes from a balance of not having so much to overcome that it becomes overwhelming; for example, expecting a child to do something before they are developmentally ready, or under-stimulating a child where no new activities are encouraged or enabled to stimulate their rapidly developing brain.

Meaningfulness is underpinned by the ability of the growing child to have involvement in decision making and the response they get from undertaking the activity is gratifying, resulting in them feeling that they matter. Conversely, if they are never allowed to have an opinion or derided when they express a view or want to do something that is important to them, meaningfulness is undermined.

When reaching adolescence, the individual experiences a time of significant physical, psychological, and social change. At this point the individual is developing their sense of reality, (comprehensibility), their mastery of the world around them (manageability) and individuality (meaningfulness). During this rapid period of change there is significant opportunity for the SOC to grow or diminish (Antonovsky 1987:102).

3.10 Family Sense of Coherence

When looking at Children's health, welfare and development, Children's development is very closely linked to their upbringing and family life. In *Health, Stress, and Coping*, Antonovsky (1979) described, almost exclusively salutogenesis as an individual feature. However, he made the statement "a strong SOC can characterise any social unit from the Jones family to a neighbourhood" (Antonovsky 1979:136).

The two hypotheses that underpin this statement include that firstly the construct could be translated from an individual to a group construct and secondly that that family SOC is related to family adaption. Both these hypotheses were considered in a study by Antonovsky and Sourani (1988) (published the year after the publication of *Unravelling the Mystery of Health* Antonovsky (1987). This study looked at whether there was agreement or not between individual spouses, SOC scores and whether that affected adaptation after a family crisis. This work concluded "that by and large there is a substantial degree of spouse agreement" (Antonovsky and Sourani

1988:79) and “families with a strong SOC are more likely to have reached a high level of reorganisation after a period of crisis” (Antonovsky and Sourani 1988:86).

At a group or family level, Antonovsky expressed the view that SOC scores would have a positive correlation, although he accepted that this would not always be the case. There may be a family member who may feel for them their experience is not coherent, though they are confident that for the rest of the family it is. In this way Antonovsky saw the group SOC taking precedence (Antonovsky 1987).

3.11 Sense of Coherence and the Life Span

In *Health, Stress, and Coping* (1987) Antonovsky hypothesised that “it is unlikely that one’s SOC once formed and set will change in any radical way” (Antonovsky 1987:188). He also added a hierarchical system to the theory as outlined below.

A strong SOC. Even the person with a strong SOC through life’s bad experiences will face challenges to their SOC, but the person with the strong SOC will “suck orderliness from the environment, which counterbalances the pressures towards disorder, from the internal and external environments” (Schrodinger 1968). This ability to see order and to have the view that the best possible outcome will occur leads the person through difficult periods of life. When these events are seen more as “challenges” than crushing blows, the strong SOC will remain over the lifespan (although ever challenged).

A moderate SOC. On taking on adult responsibility SOC becomes increasingly challenged, “One’s life is more like a spiral than a vicious circle, but the spiral is downward” (Antonovsky 1987:122).

A weak SOC is similar to that of the moderate SOC but has an ever quicker downward spiral. Therefore except for those with a strong SOC, Antonovsky sees life as becoming a downward spiral for SOC. “The loser continues to lose, and life becomes more chaotic, unmanageable, un-meaningful” (Antonovsky 1987:122). In 1987 Antonovsky’s theory remained an untested theory based on previous work and a small scale Qualitative study. Antonovsky’s initial position of SOC as a fixed or declining asset was subject to significant debate. Indeed, Antonovsky noted that view made him almost an “illich” of the helping professions” (Antonovsky 1987:124).

As more research was undertaken, Antonovsky’s (1987) view of the un-changeability of SOC was questioned (Eriksson and Lindstrom 2005). For many people meaning may not come till later in life, with a special relationship, children or even a return to higher education. This assumption of permanence and a fixed point of SOC could be argued to clash with the uniqueness of the human being. In later writings Antonovsky talked about the possibility of therapy. Life events “add a little strength and in some cases, create an opening for the beginning of a major change in circumstances" (Antonovsky 1996:16). Other researchers have gone further "it may be better viewed not as a fixed asset once gained, but as something to be recreated over and over in the course of life" (Hintermair 2004:17). In a systematic review of 458 scientific papers (Eriksson and Lindstrom 2005) it was suggested "after 10 years SOC seems to be comparatively stable, but not as stable as Antonovsky initially assumed. SOC tends to increase with age" (Eriksson and Lindstrom 2005:1). Others have found that SOC strengthens right through the life span in both male and female participants (Nilsson, Leppert, Simonsson, Starrin 2009). It has been questioned whether the sense of coherence is a fixed asset or something that has to be constantly replenished (Hintermair 2004). As a researcher with a background in the helping professions there was risk that the researcher would be more sensitive to studies that were suggestive that SOC could be developed. To counter this potential risk a significant additional review of the literature was undertaken. However, after this analysis it remained clear that Antonovsky's view of SOC being fixed or declining had not been borne out in research utilising Antonovsky's theory (Nilsson, Leppert, Simonsson, Starrin 2010).

3.12 The River Metaphor

Antonovsky uses a river metaphor to describe and position the salutogenic approach. The pathogenic approach is depicted as the "professionals" from the shore directing more and more complex ways of saving the poor unfortunate people who end up in the river. Despite becoming ever more effective in the rescue, the pathogenic experts never think about going upstream to explore why the people are getting into the river and therefore difficulty to necessitate rescuing in the first place (depicting the approach of health promotion).

To Antonovsky everyone is in the river including professionals, and those they work with. There are two key factors in how well we navigate the river. The first is the SOC which is what focuses and shapes our ability to swim and helps us adapt to river conditions by using internal and external resources to help us (GRR). Such resources enable us to adapt and swim confidently as we progress down the river. The second is our route down the river as some branches are rougher and more difficult to navigate than others. Therefore, to improve the plight of those in the river we need to study why some swim so much more confidently and effectively than others as well as the safest route down the river, rather than just trying to save those who are drowning, in the most effective manner. Antonovsky's river is illustrated below (Figure 3.2) in a PowerPoint slide used to describe salutogenic theory to participants at the launch event (see figure 3.2 below, Appendix 1 and 4.10).

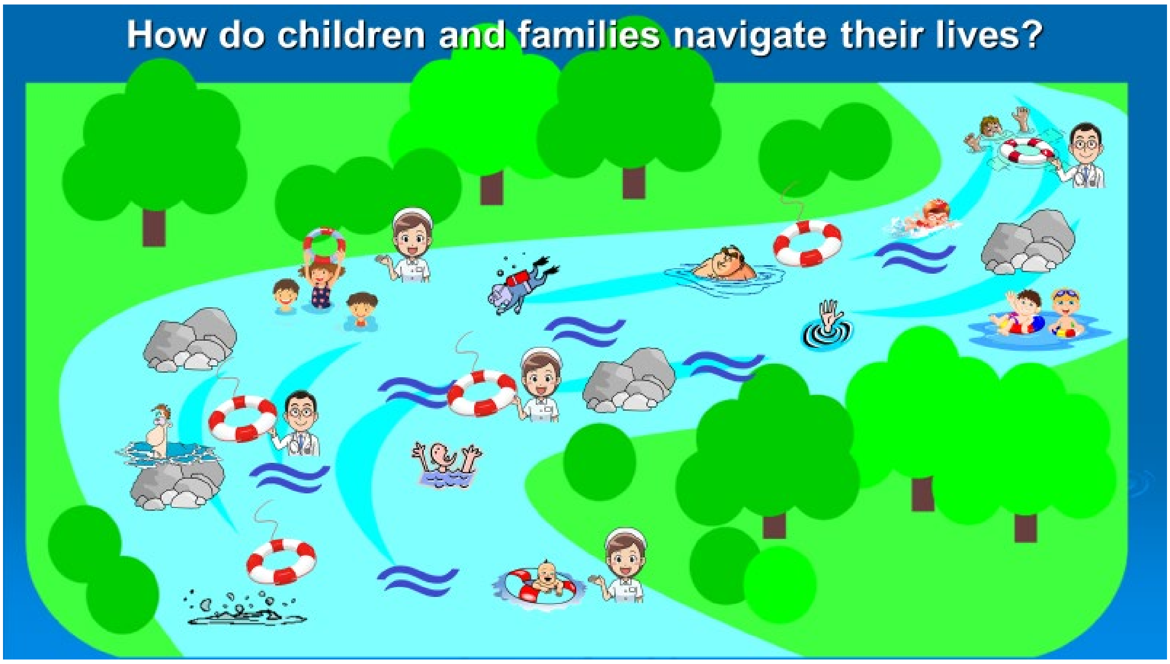


Figure 3.2 A Salutogenic Interpretation of the River Metaphor based on Antonovsky 1987

3.13 The Concept of Boundaries

Antonovsky (1987) postulates that not everything that a person is exposed to will affect them. Boundaries are theorised by Antonovsky as a way people limit what they are affected by. A boundary is the ability to narrow or widen the part of the world that affects a person. Therefore, that which is inside the boundaries will have to be comprehensible, manageable, and meaningful, for a high SOC. That which is outside these boundaries will not affect the SOC. For some people, international politics or the politics within their workplace may be unimportant to them, therefore, they see them as outside their personal boundaries and as such they do not have to be comprehensible, manageable, and meaningful. They do not affect the person's SOC, as they have not internalised such factors. Antonovsky however also expresses the view that you cannot narrow the boundaries so much as to make insignificant major areas of one's life, such as the death of a loved one and other inevitable factors. The concept of boundaries does not mean that a person can remove everything from the sphere of what will affect them, i.e. a diagnosis of cancer directly affects the person so becomes a major activity and cannot be put outside of a person's boundaries indefinitely, and thus how a person copes with the diagnosis of cancer will be affected by that person's sense of coherence. Overall, the concept suggests however, that outside factors can often be put beyond boundaries by the individual thus protecting them and limiting the areas that need to be comprehensible, manageable, and meaningful. Nevertheless, inner feelings, major activities, close interpersonal relationships, and existential issues cannot be put outside of the person’s boundaries.

3.14 Predictability

Salutogenesis as conceptualised by Antonovsky (1987) places significant emphasis on predictability as a key underpinning of a high SOC. However, “lack of predictability is not necessarily an unpleasant, or unhealthy, phenomenon in life, since we believe that disruption in life is constant in human experience” (Flensborg-Masen et al. 2005:767). This critique contends that lack of predictability is often linked to a meaningful life. Therefore, a lack of predictability may not always be negative and indeed may be perceived by an individual as positive. However, in terms of LTV the initial unpredictability experienced by Children, and their families is considerable in every area of their lives (as considered in Chapter 2.4.1-2.4.9). Consequently, for this study sensitivity to the issues related to unpredictability was seen as important to an underpinning model.

3.15 Health Professionals Influence on Sense of Coherence

Antonovsky (1987) is sceptical about the abilities of health professionals to promote change in SOC “without very considerable, quite radical change in the institutions, social and cultural settings that shape people’s life experiences. It is utopian to expect an encounter or even a series of encounters between a client and clinician can significantly change sense of coherence” (Antonovsky 1987:124). With regard to the life course, Antonovsky (1987) views childhood and adolescence as a time when a strong SOC can be impeded or facilitated. Antonovsky accepts that in adulthood, there may be fluctuations around the mean caused by major life events, but ultimately one returns to their mean. The diagnosis of a major condition may leave the person unable to make sense of an event for days or weeks; Antonovsky, however, views this as a well-structured encounter that is consistent, balanced and meaningful as temporarily boosting SOC. Therefore, interactions that are of a high quality are at least temporarily meaningful. His view was that “interventions such as these do enable people to begin to do more than reinterpret these experiences in that they equip people to seek out, within the scope of their lives, what I would call SOC enhancing experiences" (Antonovsky 1987:126). However, they “do not, and cannot, seek to change the life situations of the people that shape their experiences” (Antonovsky 1987:126). Nevertheless, as discussed in the LTV literature review, (See 2.4.1) these initial encounters with health care professionals were vivid and strong many years after the event, raising some hope of a sustainable impact (or the risk of sustained negative impact). The nature of care packages in LTV is extensive and long-term often including rehousing and many hours a week of direct care. This level of intervention/intrusion does change life situations, making LTV an interesting area to study the impact of health care interventions.

The focus of research on the sense of coherence appears to have reduced the focus on intervention studies utilising the salutogenic theory. A literature review by Dietscher, Winter, Pelikan (2017) into physical health studies utilising Antonovsky's work found 178 papers looking at the salutogenic theory and patients, 89% of these papers looked exclusively at SOC rather than the wider salutogenic theory. This focus on the SOC was found to lead to the majority of studies purely exploring the link, rather than explicitly recommending work to improve or support SOC or one of the concepts underpinning the salutogenic theory. Correspondingly there is little research measuring the impact and long-term effects of healthcare on SOC and even less looking at the wider salutogenic theory. This review noted the interesting paradox that SOC particularly in physical health studies had become a proxy risk factor, with treatment related to remedying the risk rather than promoting health, thus turning a salutogenic approach into a pathogenic measure (Langeland and Vinje 2017).

There is a small but growing body of evidence in mental health studies suggesting the "feasibility and effectiveness of taking a salutogenic orientation" within some of the physical conditions most closely related to the psychological domain such as palliative care (Millberg and Strang 2007, Millberg and Strang 2004, Strang and Strang 2001). In Children some developmental studies have suggested SOC can be developed an example of this approach can be found in (Margalit and Kleitman 2006).

3.16 The Salutogenic Model and Health Promotion

The health promotion movement after World War Two took early steps towards changing of focus away from its routes in disease prevention. The World Health Organisation (WHO) defined health as "not only the absence of disease but a state of complete well-being in a physical, mental and social wellbeing" (United Nations Department of Public Information 1948). Although widely criticised for being idealistic this definition moved away from purely a medical stance. However, the implicit inference of the definition remained a well or ill approach. The Ottawa Charter for Health Promotion (1986) pulled together a range of theories and thinkers from the public health world including Antonovsky. The charter called for a new public health where "health was created in people's everyday lives" (Mittelmark et al. 2017).

The WHO held a seminar in 1992 which Antonovsky was invited to speak at; The outcome of the meeting was a significant change of approach from a disease focus to one of health (Eriksson and Lindstrom 2008). Health promotion whilst still very diverse in approach now had a significant focus on health assets and resources for health. The overall salutogenic approach to health, when defined as a unifier of these approaches (see figure 3.1), was now a main tenant of health promotion. This need for health promotion approaches to be the central approach to NHS care have been a popular mantra of Secretaries of State for Health "For too long health has been seen simply in terms of hospitals and bed numbers. NHS stands for the National Health Service not the National Sickness Service and we want it to live up to its name," Ms Hewitt said (Gaines 2007). However successive governments have not achieved the transformation eluded to. However, a clear aim to move the NHS from a sickness to wellness service remains in policy (Department of Health and Social Care 2018).

3.17 Settings and Sense of Coherence

Antonovsky had been concerned about the risk of the SOC being seen purely in an individual context. As a sociologist, this had concerned him, Antonovsky had therefore moved to looking at the family sense of coherence (3.10). Work has continued since his death widening the understanding of this concept still further moving beyond the family setting in to communities and other social or work settings.

Studies have been undertaken focusing on settings defined as “the place or construct in which people engage in daily activities in which environmental and personal factors effect wellbeing” (WHO 1998:153). The transition into new settings such as school are important areas for addressing in research with Children. These transitions are a key feature of life with a complex lifelong condition and of childhood. From a salutogenic point of view these are times of threat to the load balance between stressors and GRR. As a result, SOC could change and become unbalanced. It has been identified that schools need to accept the Child for who they are and what they bring to school community, this is vital component to the approach of health promoting schools and the salutogenic approach (Jenson, Dur and Buijs 2017:226).

3.18 Using Salutogenic Theory in Research

Antonovsky when considering what methodology to use to develop his salutogenic theory was clear there were multiple “powerful methodologies in science” (Antonovsky 1987:63). As a researcher who had focused on survey research throughout his career, he set out to establish a closed questionnaire and scale to enable the SOC to be measured. This decision closely aligned the salutogenic theory to quantitative approaches.

3.19 Sense of Coherence Questionnaire

Antonovsky as an appendix of “*Unravelling the Mystery of Health how people manage stress and stay well*” (Antonovsky 1987) went on to formulate a questionnaire to assess SOC. The questionnaire had 29 items comprising of: 11 for comprehensibility; Ten for manageability; and eight for meaningfulness. This instrument was tested in Hebrew and English. Antonovsky notes that the people who administered the questionnaire found respondents enjoyed it and rarely missed a question.

The majority of research conducted using the salutogenic model has been undertaken using Antonovsky's original questionnaire or derivatives of it. The success of this tool has been impressive with over 400 studies (458) having been carried out using this questionnaire or a derivative of it (Eriksson and Lindstrom 2005:1). The questionnaire appears to have been extensively validated (Eriksson and Lindstrom 2005, Eriksson and Lindstrom 2007, Eriksson and Lindstrom 2008, Flensborg-Madsen et al. 2005, Flensborg-Madsen et al. 2006,). Researchers have largely deemed it to be a rigorous tool (Eriksson and Lindstrom 2005:4 and 441). The focus in research towards SOC questionnaire-based studies is therefore extensive and the widespread use of the questionnaire has been used to demonstrate the trustworthiness of the salutogenic approach. However, the focus on testing the SOC scale, rather than the underpinning theory, has led to a weaker evidence base for utilising the wider theory within the literature.

Some researchers suggest the SOC scale does not address the physical health domains of the salutogenic model (Endler, Haug, Spranger 2008, Flensborg-Madsen, Ventegodt, Merrick 2005, 2006). In '*Sense of Coherence and Physical Health. A review of Previous Findings*' (Flensborg-Madsen, Ventegodt, Merrick 2005) a review was carried out of 50 papers which have looked at the SOC scoring tool. This review found a strong correlation with psychological aspects; however, this did not appear to be the case for physical health. The researcher speculates that this comes from the questions used, in particular the questions based on predictability, which is a contentious area of the salutogenic theory (3.14). However, this critique is solely based on the SOC scale. The researchers go on to argue that it is not the theory that does not address physical health, but the questions within the SOC questionnaire designed by Antonovsky to score physical aspects of health (Antonovsky 1987:189-194). The weakness of physical health questions is a challenge for using the SOC scoring tool in an area such as LTV where there is a significant physical impact of the Child's condition.

The overwhelming predominance of the SOC questionnaire in research studies has raised concerns that the wider aspects of the salutogenic theory are becoming lost, due to the dominance of the SOC scale (Langeland and Vinje 2017). The utility of salutogenic theory in qualitative research will be considered in detail in the following section.

3.20 The Use of Qualitative Approaches in Salutogenic Research

3.20.1 Overview

Notwithstanding the predominance of quantitative salutogenic studies, Antonovsky (1987:63) nevertheless asserts that “we can only learn and advance by the use of different methodologies” (Antonovsky 1987:64), including qualitative approaches. Antonovsky (Antonovsky 1987) articulated his acknowledgement of the myriad of approaches to research and specifically identified grounded theory (Glaser and Strauss 1978) as an approach he would encourage in research into the salutogenic question. Antonovsky used qualitative research in his development of the salutogenic theory and to establish the questions for his closed questionnaire. Antonovsky maintained through his writings that” there are many alternative ways of measuring SOC and testing the model “(Antonovsky 1987:63). This approach, despite the overwhelming predominance of the research focused on the SOC scale, has continued to be developed and expanded upon “the use of the SOC questionnaire is not the same as being guided by the salutogenic perspective, salutogenesis… is a much broader concept than simply measurement via SOC” (Eriksson and Lindstrom 2008:191).

The majority of research linked to the salutogenic theory has been quantitative and based upon the use of the SOC questionnaire. The research questions in this study are however explorative and inductive in nature which suggests that for this study a qualitative approach is appropriate. To determine the applicability of qualitative methods, based on an adapted salutogenic theory derived framework, this review will go on to consider the use of Antonovsky’s salutogenic theory in qualitative studies undertaken with Children.

3.20.2 Undertaking a Review of Studies using Salutogenics

To consider the applicability of salutogenic theory in the current study a systemised literature review was carried out set out in diagrammatic format below.

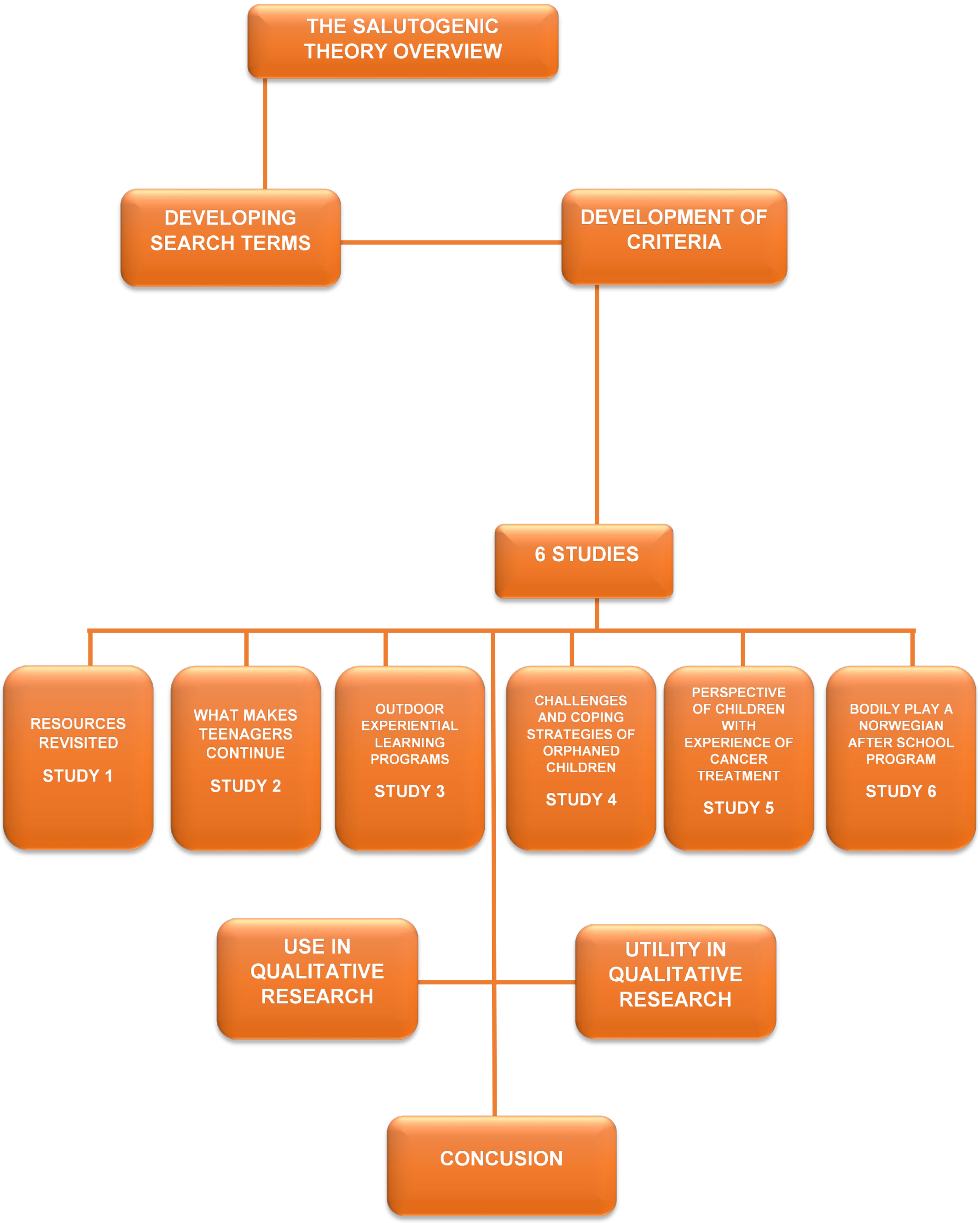


Figure 3.3 Systemised Review Process for Children's Qualitative Salutogenic Studies

This systemised review looked at studies conducted with Children that had taken a salutogenic and qualitative approach. The following search terms were developed to guide the review:

Salutogenesis and Children, Salutogenesis and Adolescents, Salutogenic Theory and Children, Salutogenic Theory and Adolescents, SOC and Children, SOC and Adolescents.

An exclusion criterion was developed which included: Studies not entirely focused on Children and families or adolescents and families, quantitative studies, studies where salutogenic theory was only used in discussion were excluded. In addition, studies over 20 years old (to keep to studies within the current generation of families), were excluded as well as any studies not published in English (Figure 3.3 above).

3.21 Overview of Included Studies

Six studies were identified that met the inclusion and exclusion criterion. Only one study was identified within this group that was focused upon chronic illness. The remaining studies cover a wide breath of Children experiences. The studies used a number of different approaches for using salutogenic theory in research; the first was focused on family adaptation from a health promotion perspective and GRR.

The first resources revisited were *Salutogenesis from a Lay Perspective* (Cowley and Billings 1999). This study involved semi structured interviews with 50 families with young children. The study focused on the GRR that parents employ, these GRR were considered to ascertain a sense of how this group of families maintain and develop their health. The study identified the usefulness of a resource-based approach in this group of families, in particular the identification of health as a concept intertwined with everyday living which has particular resonance with Antonovsky's (1987) river analogy. The practices of health visitors were considered alongside family resources to identify helpful and unhelpful approaches. The results were used to propose an alternative approach to health visiting practice based on determining and building upon GRR. This study used salutogenic theory to conceptualise the research, within both methods' analysis and in the final discussion.

The second study explored the motivation for non-elite young people continuing with sport or taking up new sports, at a point in their development where the dropout from active sports is high and recruitment is low, this study focused on the three underpinning concepts of salutogenic theory comprehensibility, meaningfulness and manageability. *What Makes Teenagers Continue? A salutogenic approach to understanding youth participation in Swedish club sports* (Jakobsson 2014). The study involved conducting 18 semi structured interviews with young people who had continued with sport despite not being elite members of the club to which they belonged. This study was inspired by the works of Antonovsky. Salutogenic theory was used within the conceptualisation and analysis, not directly as an analytical, tool, but in the words of the researchers, as a theory that was "good to think with" (Jakobsson 2014:242). This approach continues on into the discussion. The findings link meaningfulness, comprehensibility and manageability together and propose a more inclusive less competitive and varied approach to sport to continue the engagement of young people in sport.

The next study considered the benefits of outdoor experiential programs on expanding the resources (GRR) of young people. *Exploring salutogenic mechanisms of outdoor experiential learning on youth care farms in the Netherlands: untapped potential* (Schreuder et al. 2014). Within this study nine young people reflected upon and discussed their six-month experience on a youth farm. The data collection was based upon semi -structured interview questions. The analysis was conducted “in reference to the SOC framework” (Schreudera et al. 2014) with the findings presented within three domains of the salutogenic theory comprehensibility, manageability and meaningfulness. The findings were suggestive that youth farms contributed to personal development by exposure to a resource and stress rich environment, this experience was found to increase the young people’s perception that their SOC was increasing with them feeling their life was comprehensible, manageable and meaningful.

The following study used Antonovsky’s salutogenic theory with a highly challenging subject of orphaned Children in a third world context and also focused on GRR. *Challenges and Coping Strategies of Orphaned Children in Tanzania who are not Adequately Cared for by Adults* (Daniel and Mathias 2012). This interesting and challenging paper describes and contrasts two studies in Tanzania. The first study was undertaken with orphaned children living in households headed by children in rural areas. The second study involved 15 children who had been trafficked to a city. This paper is situated in a very different context to the United Kingdom (UK) making transferability low. Despite this, the study was included as the purpose of this review was to help explore qualitative uses of the salutogenic theory rather than to utilise the findings. The context is given significant weight by the researchers. The study involved phenomenological studies with the two groups. The salutogenic theory was used to gain GRR and to analyse the data through a SOC standpoint from a qualitative perspective. The findings are presented with and without the theoretical lens of the salutogenic theory. The salutogenic theory was described as useful, gaining an insight into the resources Children had, and how they used them to cope with the extreme circumstances they found themselves within.

The next study considers cancer in Children, in particular, the effects of treatment on the future of long-term survivors of cancer. The improvements in cancer treatment for many Children have changed cancer into a long-term condition, with an ongoing possibility of relapse giving this study a close connection with the experience of families with LTV Children where treatment can interrupt childhood and sudden deterioration remains an all too real possibility. This study uses an overall salutogenic approach rather than an aspect of salutogenic theory.

Friendship Relations from the Perspective of Children with Experience of Cancer Treatment: A focus group study with a salutogenic approach (Einberg et al. 2015). 15 children took part in focus groups to look at friendships and relationships from a salutogenic perspective. This study used salutogenic approach to determine what health promotion approaches could be applied to help children maintain or develop friendships during and after cancer treatment. Salutogenic theory is used as an approach and in discussion in this study. The study determined that underpinning friendship for this group of children was an “equal and mutual commitment that evolves over time and with interactions face to face and digitally.”

The final study looks at the importance of physical play in the development of the SOC and used the shortened 13 question SOC scale to guide the formulation of open questions. *Children’s Lived Experience and their Sense of Coherence: Bodily Play in a Norwegian After-School Program* (Londal 2010). The children were video recorded during play and field notes were collected. These were viewed to gain lines of enquiry for phase two; 36 children were involved in the first phase of this study. In the second phase, nine children undertook semi structured interviews where the 13-point shortened SOC questionnaire was used to formulate questions with prompts amid a wider conversation which was encouraged to develop around the answer to each question. In addition, the key lines of enquiry collected in phase one were explored further by additional interviews. The children were followed up for four months in total. The study used salutogenic theory as a holistic concept and also adapted the shortened SOC questionnaire (SOC 13) into a qualitative semi structured interview. No SOC scores were collected. The salutogenic theory was also used in discussion of the results. The study demonstrated the potential of bodily play in developing the sense of coherence in childhood.

3.22 How Salutogenic Theory has been used in Qualitative Research

All the studies bar one (Einberg et al. 2015), went beyond the overarching approach of salutogenesis and into Antonovsky’s salutogenic theory. The three conceptual areas of Antonovsky’s salutogenic theory were used in the study design (Jakobsson 2014) and the focus was on GRR in Daniel and Mathias (2012) and Cowley and Billings (1999). A different approach was taken in Londal (2010) with the SOC questionnaire being used in question formulation (with the wider theory being used in discussion of the findings). In the other studies the focus was an overview use of the theory. This was justified by the intertwined nature of the three concepts and GRR. One of the studies described the process of explaining the underpinning theory to the users and found that it’s positive focus allayed concerns around research bringing up difficult memories (Einberg et al. 2015). For the other studies this background design was not explained to parents. Salutogenic approaches were also identified as helpful in helping health and research professionals to “work in consultation with parents” (Cowley and Billings 1999:1002).

Looking at things salutogenicaly i.e. from a resource’s viewpoint, was seen as a helpful strategy to see things differently often and more from the view of the person and in order to find new solutions (Daniel and Mathias 2012, Einberg et al. 2015). The use of the three conceptual areas (i.e. comprehensibility, manageability and meaningfulness) was equally found helpful as a way to find new ways forward for children and families (Schreuder et al. 2014).

It is interesting to note that it is only in the last ten years that qualitative studies have started to be published in any number around the salutogenic theory. This may be in response to concerns about the dominance of the SOC scale in research utilising salutogenic theory.

3.23 Utility of the Salutogenic Theory in Qualitative Approaches

From this systemised search there is a small but significant evidence base that a qualitative approach in using Antonovsky's SOC theory can identify new and novel ways forward in research with Children and families by influencing overall study design, questions and in the analysis of findings. The salutogenic approach also seems to be empowering and non-threatening to participants.

These studies drawn together here are all inductive studies and have used the SOC theory differently from the strong focus on the SOC questionnaire that had dominated the research application of Antonovsky's theory at the time of this study. This different qualitative approach has led to new ways forward in the diverse areas considered above. The SOC theory has been identified as a useful part of the debate around how health is created, but to date research is limited in applying salutogenic theory to practice (Cowley and Billings 1999). The qualitative research presented in the current thesis aims to add to this data.

3.24 Conclusion

This chapter was important in that whilst Chapter Two had highlighted the literature around the topic in question (LTV in Children), the theoretical framework also needed an in-depth exploration in order for the reader to understand the perspective taken. This is believed to be a strength of this study. Overall, it is thus felt that Antonovsky's (1987) focus on what causes health is an interesting and novel way to concentrate upon the gap in LTV research with Children and families identified in Chapter Two. Moreover, no studies have been identified in this review that have used the SOC theory in Children's long-term ventilation research.

However, the researcher was very aware that the areas of permanence, of SOC and the importance of predictability needed particular consideration and care in using this theoretical framework. For example, the use of the SOC questionnaire (Antonovsky 1987) as an approach has a significant underpinning research base, however, this approach would not be in keeping with the research questions in the current study. In this study the research questions were explorative, inductive and focused on new ways of working, necessitating a qualitative, explorative and inductive approach.

This review has identified a small interesting cohort of qualitative studies undertaken with Children and their families, which have identified new ways forward in using and adapting Antonovsky's theory in a qualitative manner. These studies have identified strengths and some risks in this approach, and although they are not focused on LTV or complex care, they identify potential ways of adapting Antonovsky's theory, to use in a qualitative manner, with Children and families. These studies on their own did not describe fully a way that salutogenic theory could be utilised in this study. Therefore, the researcher based on this review, developed a novel new way of adapting and utilising the salutogenic theory (Antonovsky 1987). The close utilisation of theory and its development into questions and an analysis tool had risks. The main risk was that of being seen as theory driven. However, the gap in the existing data required a change of approach to study and flexibility was considered essential to develop the body of knowledge. This risk was considered and is reflected on extensively throughout this study 4.3,4.4, 4.13.2, 4.13.8, 6.2, 6.4. This reflection formed part of the quality review (Flick 2009) explored in Chapter Four. The two-phased analysis process was designed to help identify differences that could have occurred from this approach in analysis. As a further part of the quality review these areas were then considered, in an extensive process of member checking.

Chapter 4. Methodology and Methods

4.1 Introduction

In this chapter, the philosophical and ethical basis for the study is set out. Following on, the development and experience of data collection is outlined and critically reflected. The decisions made concerning ethics, sampling and approaching participants are explored, as are the issues of sample size and data saturation. To enable transparency, in each section methodological experience of the study is reflected on to add critique, context or support to the decisions that were made by the researcher. As such, this chapter forms the core of the underpinning quality framework as outlined by Flick (2009). To illustrate the quality control measures undertaken the key methods are demonstrated in Appendix 13.

4.2 Philosophical Approach

In any research study, it is important to understand the philosophical basis for the work (Flick 2009). In short, there are two very different ontological paradigms that need careful consideration when undertaking research, but it is important to consider the underpinning history and development of these approaches. The quantitative paradigm which originates from a positivist scientific approach seeks empirical measures, that denote truth and can be replicated. The quantitative researcher is frequently viewed as separate to the research subject and takes great care not to influence or be influenced by it (Sale et al. 2002).

It is interesting to note, that the quantitative approach developed in the empirical sciences which had moved away from the mystical thinking of the middle ages and opened the way for industrial, scientific and medical revelations. Indeed, historically, medicine and health care have been largely dominated by quantitative, experimental or positivist approaches to research, these being defined as the systematic collection of numerical information, usually within controlled conditions, using deductive processes of knowledge attainment (Streubert and Carpenter 1999). In this way, it is the ‘cause and effect’ relationship between variables that drives the work and the ultimate choice of statistical analysis. In nursing, however, phenomena that are of interest, such as attitudes, opinions and perceptions, do not always lend themselves to experiments. Indeed, the adoption of the physical science approaches has not been without critique. Dilthey (Smith 1983) questioned the possibility of separating the study of human emotions and interactions from those undertaking the study. Dilthey (Smith 1983) thus advocated that an interpretive approach was needed, one which accepted the influences of the researcher. Consequently, late in the 19th century, as social researchers started to develop approaches to study human interaction, they started to derive methods that were non-experimental, and the emergence of qualitative approaches evolved. Pioneers at that time included Durkheim, Mill, Comte (Smith 1983).

Using the qualitative approach, data can be collected from within natural environments and includes the social, cultural, experiences and feelings experienced by an individual in any given society (Streubert and Carpenter 1999). In the case of qualitative work, Denzin and Lincoln (2008) suggest that it is often criticised as being anecdotal, unreliable and invalid. Denzin and Lincoln (2008:10) add that qualitative research is often given the label of 'soft science' whereas quantitative work is considered to be ‘hard science’ and is subsequently viewed as more ‘scientific’. They further propose that this endorses a view that carries with it 'negative inferences' for those undertaking qualitative projects.

The dilemma for the researcher was which framework could be used to answer the question. In the context of this study, the question to be researched was felt to be open and was not suitable to develop into a hypothesis to test. The area of interest has little written about it, as reported in the literature review (Chapter Two) therefore, there was little insight and understanding from Children and their families that the researcher could draw on. To gain these insights requires gaining an in-depth understanding of participants’ lived experience and more specifically aiming to understand how Children with Central Congenital Hypoventilation Syndrome (CCHS) live their lives. The research base in Children’s long-term ventilation (LTV) is predominantly based on the quantitative paradigm. Examples are largely drawn from the experimental scientific approaches observed in the literature based in Paediatric Intensive Care Units (PICU) in the UK (Gilgoff and Gilgoff 2003, McDougall et al. 2013, Redouane et al. 2016). This was not the chosen approach in this study which wanted to explore real lived experience in the day to day lives of children with LTV needs and their families.

In this study, as outlined in the introduction (Chapter One), the aim was a unique exploration into the lives of Children living with LTV needs and their families. In particular, the study examined what approaches worked for them as families and supported them in day to day life. Therefore, following much consideration of the quality issues behind this decision, (Flick 2009), the researcher decided to base this study on a qualitative approach (De Vaus 2002). It was felt that there would be lessons and ideas to be found from the socially constructed meanings of Children and families in the study, which could help the researcher to explore this group.

Furthermore, the literature review (Chapter Two) identified a deficit in the knowledge about *what currently works* in the delivery of care packages for this group of Children and their families. In turn, this supported the choice of a qualitative methodological approach, which allowed for a quest for the unknown, rather than a narrow quantitative testing of a known or hypothesised approach. The researcher considered all families with Children with LTV needs to be unique and different due to the diverse social, cultural, environmental and spiritual backgrounds they encompassed. Therefore, there was no one “positivist” social structure within which a family with a child or young person receiving LTV existed. The researcher sought to understand the reality of what life was like for the families and this required more than observation, but rather interpretation by the Children and their families themselves. Different approaches were required to capture and explore such diverse reality, and it was important to understand the constructions and meanings of what was going on for the families (rather than to attempt to uncover one simple objective truth). Such issues underpinned the researcher's decision that a qualitative approach was the most suitable approach to underpin the study and the quality framework underpinning the decisions made (Flick 2009).

Further positivist researchers question the generalisability of qualitative studies based on the low numbers and lack of a highly-standardised approach (Sale et al. 2002). The researcher felt that the overall aim of this study was to generate new ideas, to challenge and test current practice rather than to specify an exact new approach. Therefore, a positivist approach would not have achieved this aim. By keeping Children and their families central at key points, such as diary design and in interviews, and through using person-centred interview questions derived from their own diaries, this study would fit well to the qualitative research approaches. The researcher also felt that this study would challenge and debate contemporary practice and an exploratory qualitative design was best suited to do this.

Finally, an inductive approach was taken in this study to analyse discourse with families and observations throughout the research. Therefore, this was an inductive study, which accepts a level of inherent uncertainty but is credible when viewed against the evidence collected. A key facet of the qualitative approach is to accept that the researcher has an influence on the outcomes of the research.

However, it is inaccurate to assume that qualitative research is always subjective whilst quantitative research is objective. All research has the potential of researcher bias, but the two paradigms of research approach the minimisation of bias in different ways (Scriven 1972 cited in Morrow 2005:254). The approach taken in the qualitative paradigm is to embrace the researcher effect on the data collected and its interpretation and to be “rigorously subjective” (Jackson 1990:154 cited in Morrow 2005:254). The importance of this to the approach requires a reflective and thorough analysis of the researcher's thoughts and feeling through the research process and an openness about how these may have influenced decisions reached (Bannister 1966). To address this concern, the researcher placed significant weight on examining personal thoughts and feelings towards the research, data and analysis. This was achieved by maintaining a detailed research diary throughout the process and analysing this diary to address the researcher's own subjectivity and bias throughout the research process, taking a reflective approach. This was a key component of the quality approach (Flick 2009) and was considered throughout the study. The researcher took the view that understanding values and opinions and being explicit about them reduces the risk of bias (Steier 1991:7). There is also a view that being open about personal views regarding the study subject from the start reduces the risk of preconception bias (Morrow 2005).

4.3. Using Salutogenic Theory

The conclusion of the theoretical review (Chapter Three), outlined the salutogenic theory and critically debated the use of salutogenic theory and the sense of coherence (SOC) questionnaire (Antonovsky 1987) as the framework tool for using salutogenic theory. The researcher concluded that the SOC scoring tool, although widely used, was not suitable for this study as it moved away from the explorative qualitative stance that this study has set out to take. Having considered extensively the risk of being theory driven and deciding that it was necessary to be flexible to develop the Children's LTV research base, the researcher explored the qualitative use of the salutogenic theory. After much deliberation around the quality basis for this decision (Flick 2009), it was decided to focus the research questions on the three underpinning domains of the salutogenic theory (previously explained in full in section 3.8) as follows:

- Comprehensibility
- Manageability
- Meaningfulness (Antonovsky 1987).

The study utilised and adapted the river analogy (Antonovsky 1987:89) which is fully described in 3.12 (Antonovsky 1987:89) but as a recap this analogy describes and critiques the "downstream," focus of much of the investment put into health. Using this analogy in this study kept the descriptions rooted in the theoretical base of Antonovsky (1987) and subsequently allowed the researcher to explain the underpinning methodology of the study. The explanation of the theoretical underpinnings of this study using this analogy with Children was a unique aspect of this study and had not been undertaken in any of the Children's qualitative studies considered in the literature at the time of the review.

4.4 Using Appreciative Inquiry

Appreciative inquiry (AI) was also chosen as a secondary approach to supplement salutogenic theory by gaining an insight into participants' views of what an LTV care service should aim to achieve (Cooperrider et al. 2009). AI is based on a social constructionist approach. In this approach, the way people, groups or organisations are organised are only limited by people's imaginations and what they can create. This methodology was designed "to symptomatically and deliberately appreciate everything of value" (Cooperrider, Whitney, Stavros 2009:5). This approach aims to capture the ‘best things’ that are happening in the current context for the individual or groups, rather than trying to learn from what is going wrong. By using such an approach, the value is in making the individual or group feel comfortable in what is known and trusted. Traditionally, four stages are set in the approach as below:

1. Appreciate what is.
2. Imagine what might be.
3. Determine what should be.
4. Implementation, create what will be (Cooperrider, Whitney, Stavros 2009).

Appreciative Inquiry (AI) has been extensively used in service redesign. The original four steps have now largely been superseded by a five-step approach. This extra step acknowledges the mind set change that may be required for many organisations and people to express themselves and think in a positive manner (this becomes step one). The five steps, as outlined, were chosen for this study as they have the advantage of clearly locating this model in the field of salutogenesis.

Figure 4.1 The Essentials of Appreciative Inquiry

https://www.centerforappreciativeinquiry.net/more-on-ai/the-generic-processes-of-appreciative-inquiry/

Choosing AI with salutogenic theory was a unique approach of this study, but was felt to be a good fit, as AI was chosen as an approach to underpin the primary theoretical approach of salutogenic theory. Whilst novel, both salutogenic theory and AI locate into the broader beliefs of salutogenesis as they both look for what works for people, making them well, happy and enabled to achieve and overcome adversity. Both approaches link to positive moments of life and stories that capture those moments. Arguably, AI goes a step further than the salutogenic theory as it is focused on creating a shared vision. However, fundamentally AI is an organisational change tool and the salutogenic approach is a theory. This had the potential to create a confused approach therefore, in selecting AI, it was important to consider the theoretical alignment with salutogenic theory as this was novel to this study. The focus on *what is going well* resonates well in the overall category of salutogenesis. Whilst AI could be suggested to cross into Generalised Resistance Resources (GRR) in terms of identifying life giving forces, the salutogenic theory's main tenet is the SOC. The SOC is a more complex concept of this theoretical approach which supports the notion that all humans or people have a sense of coherence which is our innate ability, when faced by a challenge to firstly believe we will overcome a challenge and secondly pick the appropriate GRR to overcome it.

Appreciative Inquiry (AI) is an outcome focused positive change process, whereas the salutogenic theory is an explanation of the question of what causes health. A purist view of salutogenic theory is arguably more guarded, even pessimistic than AI. For example, in writings by Antonovsky, it was purported that once a SOC was established from early adulthood there may be little prospect of strengthening or changing it (Antonovsky 1987). Conversely, in AI there is a clear sense of optimism which can change over time from person to person and across organisations (Carter et al. 2007). Interestingly, this original fixed view of SOC lessened in Antonovsky's later writings and has been largely disproved (Mittelmark et al. Eds. 2017).

However, fitting in the same overall belief systems of the broader meaning of salutogenesis does not make these two theories totally compatible for use together. The researcher liked the notion of having mixed approaches and indeed a limited number of other studies have used both approaches including physical education (Enright et al. 2014), gerontology (Naaldenberg et al. 2011) and studies centred on compassion (Wasserman and McNamee 2010). The approach in all these studies was to use AI as a data collection method and salutogenic theory as a theoretical approach, to inform the approach and the discussion. However, none of these studies adapted salutogenic theory, or its key concept of SOC for use in the data collection phase.

To really explore this for the first time the researcher then felt it would be useful to compare the three main areas of the SOC to AI in detail. (NB to illustrate this process each area of the SOC is outlined in brief in italics followed by a comparison to AI this was undertaken to inform the decisions around quality and ultimately the approach taken).

Comprehensibility: – "*A person with a high SOC sees confronting stimuli as making sense in that they will be expected or if unexpected they will be ordered or explicable. This includes undesirable stimuli such as death of a loved one or war" (Antonovsky 1987:17)..*
In AI there is a level of crossover with comprehensibility in that of taking the best of what currently works forward into any new model. This may be best practice experienced in care environments or things that work particularly well for a family in the context of their life. Therefore, there is a level of familiarity which aids a family to adapt to the new context that they find themselves within. However, AI does not recognise the SOC as the regulator; i.e. the belief that things will work out as well as could be expected, and an ability to effectively use GRR so there are fundamental differences in this regard.

Manageability: – *"the extent to which one perceives that the resources at one's disposal are adequate to meet the demands posed by the stimuli that bombard one" (Antonovsky 1987:17). This may include resources of the family, friends, or spiritual resources*
In AI there is less of an emphasis in this area and any crossover comes from identifying the "life giving forces" (Cooperrider, Whitney, Stavros 2009), that enable a person to get through, although this is slightly less functional than having resources available to meet issues and burdens which is implicit within manageability.

Meaningfulness: – *A person with a strong SOC spoke of areas of their life that were important to them, that they were very much cared about and that made sense to them. Antonovsky saw meaningfulness as the motivational component. This component also includes the significance of being involved in decisions. In addition, it encompassed the emotional component with a link to "life making sense emotionally" (Antonovsky 1987:18).*
Meaningfulness appears closely aligned with AI. In the AI approach an appreciation of all that is of value and recognition of the motivational effect of maintaining that which has proved helpful or effective is central. The aspect of visioning also aligns well with the aspirational components of meaningfulness. Where AI goes further is in visioning a future, which although arising from meaningfulness, takes this consideration an important additional step.

Overall, whilst AI and salutogenic theory have similarities and sit well in comparison, the approaches are divergent in focus in different areas. Salutogenic theory aims to explain why some people survive and indeed thrive in the face of adversity, where others struggle and succumb. AI is primarily an organisational change model that aims to take the best of what is currently happening forward in a collective positive vision of the future. However, this was useful as when considering the research questions posed in this study and the study group it was felt both salutogenic theory and AI together with their differences would give a complete approach to enable the research questions to be fully considered. Whilst this seemed a sensible pragmatic approach, there was always a risk by using two similar, but different approaches, that the underpinning methodology might become muddled and confused. The researcher was aware of this and managed risk by carefully recording and reflecting through the research diary and supervision. Through this process it was decided that the use of AI would be limited to use in the final interview question in the interviews and focus groups. These data collection points focused on a key element of the research question which was asked encapsulated in the following question:

If there was no service for LTV and you were advising on what the perfect service would be what would it look like and how would you describe what should be put in place?

On reflection, such an overall approach was effective and kept a clear focus on an adapted salutogenic theoretical approach, reducing the risk of theoretical confusion whilst using AI in a tightly defined way to answer an element of the research question. This novel and imaginative approach worked very effectively; enabling the research question to be answered but also meant the important pulling together of what worked for the children and families in this study was captured.

4.5 Ethics and Governance

Ethical approval was sought and gained from Coventry University Ethics and Governance Committee. It is vital in any study to explore further the context of ethics and governance. Further ethical decisions are a key component of the quality framework and are therefore presented in detail (Flick 2009).

In the aftermath of World War II and the atrocities enacted by the Nazis under the name of research the Nuremburg Code (1947) was developed. This code set the basis for “willing unpressured consent” (Fraser et al. 2004:97). However, it was not until the “1970s and 1980s, that research ethics committees (and, in the USA, institutional review boards for medical research), began to be introduced widely” (Saks, Alsop 2013:300). In the UK, the Alder Hey Hospital organ retention scandal illustrated that ethical violations are still a feature of some research (Fraser et al. 2004). Within the healthcare professions ethical practice has become integral to professional codes and as a nurse the researcher was bound by the *Nursing and Midwifery Council’s (NMC) Code of Conduct* (NMC The Code 2013). In this study, the NMC code was strictly adhered to which states:

- “Make the care of people your first concern, treating them as individuals and respecting their dignity.
- Work with others to protect and promote the health and wellbeing of those in your care, their families and carers, and the wider community.
- Provide a high standard of practice and care at all times.
- Be open and honest, act with integrity and uphold the reputation of your profession”.

(NMC 2013).

4.5.1 The Concept of Voluntary Participation

Voluntary participation was a component of the *Nuremburg Code* (1947) and in some studies, this can be difficult to achieve (such as studies, which involve observation of large groups in their natural context, for example field research). The issues for this study around voluntary participation included the position of the researcher. The researcher is a nurse who is an expert in the field and well- known to practitioners and some families. This in itself could be seen as pressure to participate. To reduce this influence there was a clear process for approaching users and a carefully worded information sheet generated, both for the user group and all the possible participants. The researcher approached possible participants through a national support group (rather than directly giving an extra level of distance from the researcher to assist families to make a free and unbridled decision).

Within this study, consent was gained from each participant, where old enough to consent. This was achieved by using registered Children’s nurses to make this judgement and to work with the researcher to take consent from parents and Children. There was an open-door policy where the Child could, accompanied by an adult, return to their parents or re-join the session at any time. Each member of the research team was a children’s practitioner, trained and fully enabling of this approach. All Children had full access to the research/playroom and could join in the research or just play.

For all the potential participants in the study there were many potential influences (for example peer pressure from other members of the CCHSUKSG). The researcher made the concept of voluntary participation clear in the participant information sheet (Appendix 5, consent form Appendix 7) and launch presentation (Appendix 4) on the day and to each Child on arrival in age appropriate language. Although signup was high, one family declined to be involved and a further four subsequently chose to leave the study which helps to support the view that freedom of participation was achieved. This family choice was totally respected.

4.5.2 No Harm to the Participants

The Nuremburg Code (1947) concentrated on potential physical harm to participants. However, in qualitative research the danger of psychological harm is just as real. Participants, for example, may recount painful memories or reveal behaviour or attitudes that may be uncomfortable and at worst highly emotionally distressing. The research process may make the participants think about deeply suppressed worries and concerns. This may be of particular relevance in LTV where the importance of learning complex practical skills often forces parents to bury the devastating emotional consequences of the diagnosis to get on with life-saving care. The use of the CCHSUKSG was particularly pertinent in this area. If a participant was distressed and required further support, the support group could have been called upon by the researcher or by the research participant. However, this did not occur during the study. As the CCHSUKSG program and the Children's research went on in parallel rooms', parental support was available at any time and Children were returned to their parents at any time they wished. As two of the children who attended the playroom were under three and others were nervous, it was important that they had a sense of not being separate from their parents. The CCHSUKSG meeting was running in a room across the corridor. The researcher wanted to achieve a sense of them just playing in another room. This was achieved by the young children and the more nervous older Children initially moving between the playroom and the room across the corridor frequently, (supported by one of the team) and as they became more confident, they stopped longer. By the afternoon, when the vignette scenarios were up and running, the Children were confident and stopped for the whole session. By running a playroom where parents were not present there were medical and practical risks to consider. The playroom was separately risk assessed and had nurses trained in CCHS present at all times. All staff were employed, vetted and trained by a local NHS trust. All equipment used in the room was cleaned and maintained in accordance with the local NHS trust's policies. A checking in and handover process was established, and all Children had name badges with a sticker identifying Children with CCHS. A nominated nurse was delegated the task of not being involved in activities and taking an overview of all Children in the room to quickly identify Children becoming sleepy or showing signs of medical distress. The procedures worked well, and all medical issues were quickly and effectively dealt with.

Wherever practical, research should go beyond doing no harm to having a level of benefit to the participants. The approach of this research had been to organise and fund an additional meeting of the Central Congenital Hypoventilation UK support group (CCHSUKSG). The group had requested an additional meeting that it did not have the funds or the capacity to organise itself. Since the WellChild Charity had funded the researcher, there was capacity to offer financial support. By funding the rooms and catering for the meeting and providing a play room staffed by play specialists and nurses from a local children's community nursing team, these resources were made available over the three subsequent years where the researcher aimed to facilitate the ongoing data collection and to give extra support to CCHSUKSG members. The Children overwhelmingly were positive about the playroom, which has been fed back on each occasion individually by Children, parents and the CCHSUKSG Chair.

The nature of the enquiry involved daily diary keeping but only of the positive aspects of care and support. There is evidence that keeping a positive diary enhances mental health (Lambert et al. 2009, Toly et al. 2014). However, any reflective process opens the possibility of buried or unresolved memories surfacing. Occasional contact with the researcher and at least two face-to-face meetings (together with the added pressure of keeping a diary as well as caring for a long-term ventilated Child) needed to be carefully considered and openly discussed in the research information leaflet and during the consent process.

The process of maintaining a reflective diary and regular supervision during the research was critical to ensuring the principle of no harm to participants. The research diary acted as a focus for reflection helping to identify issues where the researcher’s complex collection of roles and skills could have impacted on the participants themselves. The ability to consider these issues with a highly-experienced research team skilled in adults and children’s research helped reduce these risks.

4.5.3 Anonymity and Confidentiality

It is important to identify the difference between ensuring anonymity and maintaining confidentiality. In the terms of this research, confidentiality was possible, but anonymity was not, as the researcher met the research participants directly. Confidentiality was achieved through storage of data on secure NHS servers, and paper documents secured on alarmed NHS premises in a secure filing cabinet. Data was collected from participants on the final visit in two forms. The interviews were recorded on an encrypted recording device and paper or electronic diaries (collected on individual memory sticks) were put away before leaving the house in a locked bag. On reaching the car both were placed in a locked boot (locked in locked) and immediately upon return, uploaded onto secure NHS servers or received via a secure NHS.net account (secure email standard ISB 1596 NHS Digital May 2018). Paper diaries were kept in a locked filing cabinet in a locked NHS office in an alarmed NHS premises. The only exception in the transportation of data was a participant from Northern Ireland, where the encrypted recorder and paper diary were locked in a box in locked hand luggage which remained with the researcher on the plane. On reaching the car the established process described above was followed. The anonymising of data was achieved through each participant being given a numbered diary, scrapbook, memory box and memory stick. Secondary data anonymising was undertaken at transcription to remove identifiable data shared by participants. All participants at the CCHSUKSG event completed the local NHS trusts photographic permission forms at each event and the local NHS trust policies on the use of images was strictly adhered to at all times.

4.5.4 Deceiving Potential Participants

Deception is rarely deemed ethical in social research. To be acceptable, compelling scientific evidence for its use would have to be presented and agreed by the ethical committee. In this research, there was no such evidence or need, therefore an overriding approach was one of being open. This fully included the Children and as such, age appropriate explanations were provided by fully registered children's practitioners.

4.5.5 Analysis and Reporting (Justice)

An ongoing discourse with participants was used to inform participants of how the research was being disseminated and used. This occurred at two points in the research; at the steering group and also at research group meetings the researcher shared findings and reviewed the eventual findings prior to the write up. The researcher was prepared to be open and honest about unexpected or unwelcome findings. In the event (possibly due to the salutogenic theory focused approach) there were no difficult findings. The unexpected nature in the findings to the participants were typically positive, for example, how much they achieved as a family and how this differed sometimes to their reflection at a given point in time, rather than as what was picked up in the diaries over an extended timeframe.

4.5.6 Summary of Research Ethics

Ethics were carefully considered throughout this study. The initial approach had ethical approval.

When the Arts-based methods were expanded to include participatory play-based scenarios (see 4.8.3), ethical approval was sought by way of an amendment to the initial ethical approval and granted for this amended approach. All research has the possibility of causing harm to the participants if ethical standards are not strictly adhered to. In this study ethics were carefully considered at each stage and ethical approval was gained and amended when required. The overall approach taken was to use methods that were low risk and had the potential to be fun or helpful to the participants. No issues were raised with the researcher, CCHSUKSG or the university during this study. Evaluations by participants undertaken by the CCHSUKSG, noted the value to the attendees of the play rooms provided by the researcher. Some participants particularly valued the experience of keeping a diary and found the experience very positive, suggesting that, at least in part, this study met its aim of going beyond doing no harm and creating some benefit to participants.

4.6 Positionality in the Research

As a qualitative researcher, reflexivity is a vital component of the research process. This dynamic process of self-awareness was important in considering the researcher's position in the research (Flick 2009, Fowler 1988). Getting a balance between the role of researcher and nurse has been raised as an issue in the literature (Gerrish 1997, Spradley 1980). As a researcher, with a professional background in nursing, there was from the outset a discourse with the supervision team and through the research diary, kept throughout the development of this study around positionality. Positionality had some significant areas of challenge in this research. The first challenge was the choice of whether to inform participants that the researcher was also a nurse who had practised for many years as a children's community nurse (CCN) with a specialist knowledge of Children's LTV. A confirmation of this identity at the outset may have created some elements of being an insider which has been identified as valuable in nursing research. This trust can help achieve acceptance and make it easier to form open relationships with participants (Gerrish 1997, Spradley 1980). However, as a clinical expert in this area, there might have been a perceived power differential creating the potential of participants seeing the researcher as a source of knowledge or even authority undermining the researcher's ability to enquire and question about the day to day realities of participants' experience, without changing the natural patterns of interaction (Gerrish 1997, Spradley 1980). Participants, due to their wide experience of the health service, would have extensive experience of the nursing role but conversely their experience of the role of the researcher was likely to be significantly less (or none at all); therefore they were more likely to identify the researcher by the role they knew and understood that of the nurse (Fowler 1988). This risk was equally applicable to the researcher as there was also a risk that the researcher would revert to the extensive years of socialisation in his professional role, rather than his less developed role of being a researcher (Fowler 1988). It was essential therefore to have a clear stance on this issue. Without clarity, role confusion could ensue leading to important ethical and methodological issues. An example of this could have arisen in the process of consent. The risk was that if the roles were unclear to the participant or the researcher, there could be confusion about whether consent was being given or taken by a nurse or a researcher. This identity issue could then result in participants being confused as to whether they were consenting as participants or patients. There was an equal risk or a similar confusion arising in the researcher (Fowler 1988). A position was agreed in supervision that the researcher would identify himself as a researcher not a nurse, however, if questioned there would be an open and honest response. The approach chosen has been considered and questioned in other studies, where full disclosure of all a researcher's roles at the outset has been advocated (Fowler 1988). On balance, the risks to the participants and researcher of the possibility of the nurse role dominating that of the researcher (due to over thirty years' experience as a nurse and fourteen years' experience of looking after Children who require LTV) and the risk to participants with many years' experience of extensive Health Service use were considered the paramount risks.

In the study the consent process and the first four interviews were conducted before the question was asked whether the researcher was a nurse. On reflection of the research diary, the revelation of the researcher's background did not seem to change the relationships as much as expected and simple approaches identified in (Fowler 1988) such as directing questions raised around practice to the CCHSUKSG Chair or trusted information sources sufficed. In addition, on reflection of the research diary, it appeared that this was due to initial relationships being based on the role of the researcher which set expectations from the start. However, the issue of positionality is more complex than identification of profession. Thus, the wider aspects of positionality will now be considered.

The most significant issues in this study arose from the culture context and practice differences between the two roles. Moving from a culture of compassion to a culture of critical questioning has been identified in quantitative and qualitative research (Springwood and King 2001, cited in McGarry 2007:11). The innate nursing need to make a difference and intervene, has as a practitioner been an area of struggle for the researcher which culminated in the study of counselling techniques, later honed in the field of Children's nursing where a need for compassion and care whilst empowering a Child and family to take on care themselves, is the predominant nursing approach. As a nurse researcher, it has been argued that a nurse has a moral duty to maintain their integrity and identity as a nurse (Fowler 1988). This duty results in the nurse researcher needing to retain a clear focus on the needs of the Child and family. It has been observed that in research the process of seeking out and being interested in a participant's story is of itself enabling (Eide and Kahn 2008). In this study, the duty of a researcher, with a professional background in nursing was enacted by trying to achieve more than doing no harm. The researcher aimed to use approaches and methods that had the potential to be empowering, useful or fun to the Child or family, as such the ethical needs of both the research and the ethics of nursing were met by focusing on "human flourishing" (Fowler 1988:111).

As a researcher, the researcher needed a clear commitment to the advancement of knowledge. However, as a professional nurse, care and compassion are central to the nursing identity (Fowler 1988) as discussed, these approaches are not easily aligned. However, there are some pre-existing cross over points. As a Children's nurse, a questioning approach is essential to safeguard Children (RCN 2014). The duty to safeguard (RCN 2014) necessitates a questioning approach requiring a thinking of the unthinkable. This approach is a key skill for safeguarding

Children which requires a continual questioning of a family’s actions and motives (RCN 2014). However, the adaption to the questioning approach of a researcher is far more encompassing and challenging in research as the process of data collection and study of data becomes the central role. The researcher’s research diary notes the struggle that ensued within the researcher during this transition. Throughout this thesis, the researcher has addressed this concern through supervision and reflection.

Another area of challenge related to positionality, was that of being an observer when help is required. When the area studied is an area the researcher has a clinical expertise in, there is a professional ethical issue of whether to intervene as a nurse, or to maintain the observer status of the researcher. This concern is widely reported in nursing research (McGarry 2007). This issue was most common in data collection, when concerns or issues raised by the participants could potentially have been eased by knowledge the researcher had from practice. However, as a researcher moving into a role of adviser would have created a crossover from researcher to nurse (Ballie 1995).

One such challenge came through the Chair of CCHSUKSG who indicated the group wanted talks during their away days from the researcher on care packages. This request posed a potential dilemma. As a nurse, there was an internal struggle with the innate nursing need to give what may be of help (Fowler 1988). This was carefully handled and addressed by members of the researcher’s nursing team coming to speak as a separate entity to the research and the researcher. An additional challenge was a consideration of clinical risk to Children when organising a meeting of Children and families, where the child has a highly complex life-threatening condition. CCHS necessitates essential ventilation overnight and during sleep. As a nurse, the researcher could not separate himself from the safety and care of the Child and family (Fowler 1988). As a nurse it was considered prudent to ensure local ambulance services; local emergency admission units and regional children's ventilation teams were aware that a large number of Children with complex needs were in the area. (In addition, contingencies such as power cuts were addressed). The researcher considered this an ethical responsibility under *The Code* (NMC 2013). Consequently, the researcher worked with the Chair of the CCHSUKSG to update the group’s risk assessment and to ensure the above issues were addressed.

The role conflict between that of a researcher and nurse was most apparent during this time. The researcher’s research diary was used to consider these conflicting demands and to develop a strategy that maintained the researcher's role as the researcher, but met the responsibilities of having a professional background in nursing. This was largely achieved by working behind-the-scenes with the Chair of the CCHSUKSG to ensure the additional risks, that were apparent to a professional nurse, were risk assessed and addressed and that requests from the group for information on topics familiar to the researcher were addressed as a separate entirety by the researcher's nursing team. The researcher, in common with other nurse researchers, found a total separation of the role of nurse and researcher was impossible (Fowler 1988), however a clear differentiation at the start enabled a context to be set that maintained beyond the recognition by participants of the researcher's professional nursing background.

In summary, this section has identified these struggles and the process and mitigation taken, but more importantly it has attempted to reflect openly and honestly on the underpinning motivations and actions of the researcher, so the reader can evaluate for themselves the influence or not attributed to this study by the researcher's approach.

4.7 Development and Rationale of the Data Collection Methods

Following much reflection, a phased approach to data collection was felt to best fit the study in order to meet the aims. Using a phased approach enabled the researcher to gather data at timed intervals and allowed for reflection and further development in-between. The phased approach is set out firstly as a description, followed by application to then critically reflect on the reality of using the chosen methods (Flick 2009). To give an overview of the quality measures that were integral to each method, a table is presented in Appendix 13 (Flick 2009). Chosen methods included Arts-based diaries and participatory scenarios. None Arts-based approaches included interviews and focus groups. To help reflection, the data collection methods are diagrammatically represented as below (Figure 4.2).

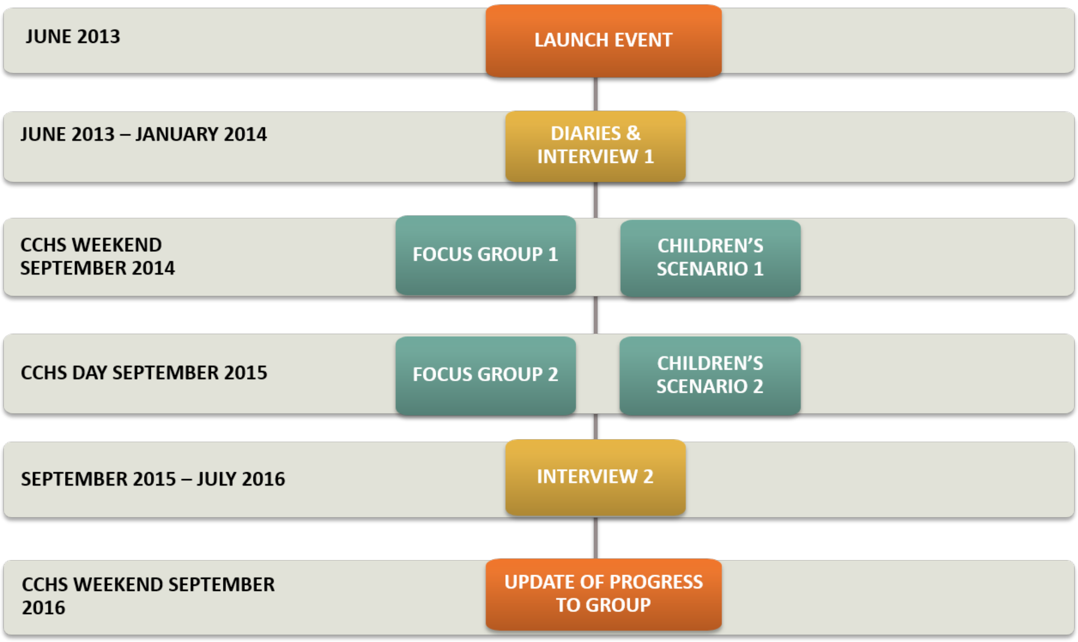


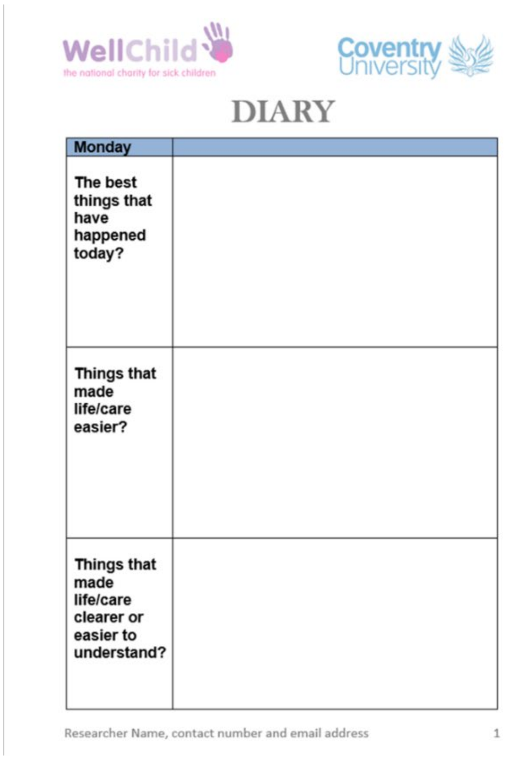
Figure 4.2 Process of Data Collection

4.7.1 Diaries

The first method of data collection for families for discussion was the use of a diary method as Figure 4.2 above demonstrates. Diaries have been a feature of human culture for a considerable length of time. Diary usage has been traced back to the 10th century Imperial Court of Japan and English monks (Whittemore and Knafl 2005). Diaries are a way of capturing the mood at a given point in time, making them a useful research technique and have been used in historical and current, qualitative, and quantitative research as described in the following quotation:

*“Diaries tell the truth, the partial truth, and a lot more beside the truth....... in them, you seek-and often find-an atmosphere, a sense of the mood of the moment, which could not be acquired in any other way. They should never, ever, be taken as the last word, but as the raw material for reconstruction of the past they are, invaluable “(*Pimlott 2002:24).

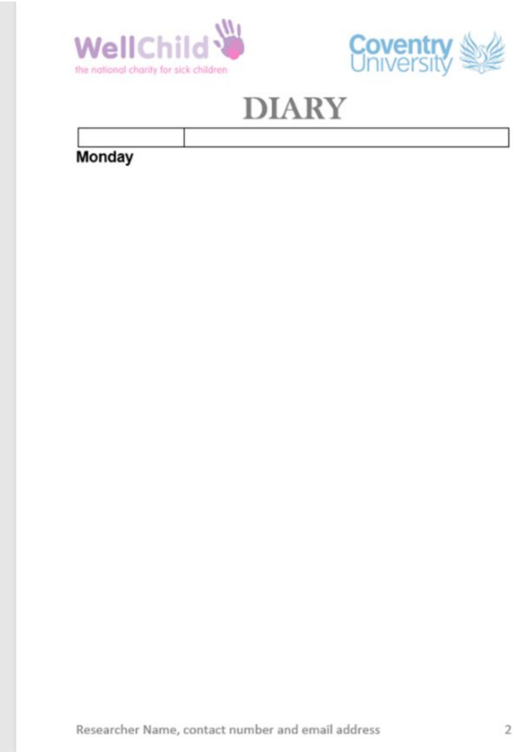
Diaries have frequently been cited as a narrative approach or even a mode of self-administered longitudinal questionnaire (Snowden 2015). However, in capturing the daily story of a family in both written and visual ways, this methodology was an Arts-based method in the wider view taken of Arts based methods (Eaves 2014). The diaries created for the study were designed to allow both written and drawn entries. For each day there was a written page and a page for drawings (Figure 4.3).



DIARY

Monday	
The best things that have happened today?	
Things that made life/care easier?	
Things that made life/care clearer or easier to understand?	

Researcher Name, contact number and email address 1



DIARY

Monday

Researcher Name, contact number and email address 2

Figure 4.3 Example of the Arts-Based Diary

This method was chosen as the families in this study all had a Child with complex care and long-term ventilation needs. As identified in the literature review, this meant they lived busy and stressful lives (see 2.4.2 and 2.4.3). It was envisaged that using a diary would allow them to undertake *real time recording* and in doing so record any issues arising as the day/weeks evolved. It was hoped that in this way memories could be captured that might have been lost in the day to day stresses of their lives.

The use of diaries with such a group was not entirely unique to this study. Diaries have been utilised successfully in families with Children with complex needs before (Nicholl 2010, Toly et al. 2014). Diaries have been used in resourcefulness training as a cognitive behavioural technique with parents of Children with complex care needs to enhance mental health (Toly et al. 2014). The principal advantages of using a diary are that they are contemporaneous, in that entries are made at the time)or close enough to the time) when events or activities occurred, so that the record is not distorted by problems of recall. Diaries are also particularly suited to gaining lived experiences as they facilitate the examination of reported events and experiences in their natural, spontaneous context (Bolger et al. 2003:36).

For this study, it was envisaged that the use of diaries would allow the researcher to gain a *real feel* of the day-to-day nature of the participants' lives. For example, it was felt that using this approach captured issues relating to the large and invasive care packages health staff deliver and it was also hoped that other features of day to day family life would be recorded. This method was also felt to be useful as participants would record items every day rather than only at the point of interview when thinking would be a response or a memory. This was similar to the findings of Nicholl (2010).

As with all data collection techniques there are challenges in using any method. Firstly, diary content is limited to small snapshots in time and for this reason could not reflect the whole of a "complex phenomenon" (Nicholl 2010:18). Clarkson (2003:56) also notes that diaries *"tell us what the participant wants us to know, which is not necessarily what the researcher is really interested in".* Diaries therefore, offer a version of events from the perspective of the narrator. However, in this study, this was offset by using the diaries as a platform with the one to one face to face interviews as a follow up. Further, the diaries were used as a platform to create the questions and structure of the interview. The follow up interviews also allowed entries to be examined in detail, after they had been written by participants, allowing any "gloss" to be reconsidered or challenged.

4.7.1.1 Diary Design

Prior to the start of the data collection, the decision was made to use both a paper and electronic version. There were many reasons for this decision; for example, the researcher had learned over years of clinical practice that many families already used paper diaries in their day to day lives. It was envisaged that a small paper diary could be easily carried around and used in any situation without the need for any kind of technology or technological skill. When using a paper diary however, the user may have concerns about their writing and spelling and for some, they would prefer electronic means, so they can smarten and correct diary entries. For the researcher, it can also be hard to transcribe and understand handwriting in paper diaries (Snowden 2015) Therefore, for this study, the researcher chose to also provide an electronic diary format.

Electronic diaries overcome some of the issues identified for paper diaries but have unique issues of their own such as evidenced in Clarkson (2003) and Nicholl (2010). When using an electronic diary, it is important to ensure that any system used is as accessible as possible, for participants to become involved in the study. Word and email at the time of this study were far the best-known electronic data capture and transfer methods. It was anticipated that by families entering data on their own devices, data entry would be swift and familiar to them, thus enabling timely recording of diary entries. However, the opportunity for more modern and engaging data capture (such as a custom-made electronic diary) was initially missed. However, after discussion with the parent representative, the Chair of the CCHSUKSG and a fellow PhD student (who was specialising in electronic data capture), who had raised concerns about a bespoke electronic diary approach (without access to training and technical support), a further search of the literature was undertaken around electronic diaries. From this search it was found that the majority of studies had considered or used bespoke electronic methods. Only one study was found that had studied the use of email. The findings identified that no training was required by participants when using an email-based approach and there was a high level of acceptance from participants as the technology was familiar to them (Whittemore and Knafl 2005). Therefore, to further develop the data collection methods, the low threshold of word and email were chosen as electronic data capture methods, as they were familiar to most computer users allowing accessibility and speed of access, which were the primary requirements in this study.

However, data security is an issue with all data but in particular electronic data. This was a consideration in the inclusion of electronic diaries. The data remained within the families' own data systems until sent to an NHS email address. NHS e-mail is secure (secure email standard ISB 1596) and the data can then be securely transferred to NHS servers. Participants also had the choice of saving the diary on a USB stick, which was physically picked up by the researcher in a locked case, within in a locked car boot and then transferred directly to NHS systems. These procedures ensured that data security was maintained at all times.

The format of the diary required much consideration. Diaries can be potentially more than just words and for this study the researcher wanted to allow all the family members to be participants. In the families studied however, many of the children and siblings were too young to add written or drawn entries to the structured format of their family diary; to attempt to address this issue scrapbooks were developed as per Section 4.8.

4.7.1.2 Formulation of Diary Questions

As discussed, to achieve the research aim of capturing what works for Children and families living with LTV at home, required a new approach. The decision to include theory in the formulation of the research questions was decided upon after extensive reflection, supervision, critical reading and quality review (Appendix 13). This was done to enable a salutogenic (Antonovsky 1987) focus to the data collection. A decision was therefore made to focus data collection via the diaries on the 3 theoretical concepts of comprehensibility, manageability, and meaningfulness (Antonovsky 1987) which had been used previously by Einberg et al. (2015); Jakobsson (2014) and Daniel and Mathias (2012). However, as described elsewhere, using salutogenic theory in Children’s LTV research study was unique and setting the diary questions around these three concepts was also novel.

To formulate questions on these domains was more difficult than had originally been envisaged. The words used needed to both reflect the concepts as described above and be understandable to the participants. Therefore, the researcher used the extensive literature review to develop a table of different descriptions used in the literature (Appendix 8). This table was then utilised to develop and draft three initial questions for the diary as below:

1. Can you tell me about what or who made today meaningful or motivating, or something that gave you satisfaction?
2. Can you tell me about things, people or thoughts that made today more manageable for you or your family?
3. Can you tell me about thoughts or events today that have helped to make things more understandable, clear, or more predictable for you or your family?

Once formulated the questions were submitted to a parent representative who offered to appraise them. She replied that she did not understand the questions and therefore had not been able to record any data. Whilst initially this felt like a set-back, it demonstrated how important it was to ask verification of data collection tools with users directly. The questions were therefore re-defined and re-developed as short prompts as follows:

1. Special things that have happened.
2. Things that make life/care easier.
3. Things that make life/care clearer or easier to understand.

Further consultation was undertaken with the same parent representative and the Chair of the CCHSUKSG. As a result, the first question was changed to:

1. What are the best things that have happened today?
2. Things that make life/care easier?
3. Things that make life/care clearer or easier to understand.

The researcher felt confident in the refinement process and that the final agreed questions are based on salutogenic approach, but adapted in a pragmatic way. Families still had to report what was going well and what helped them in day to day life, whilst being easy to understand.

Each diary had a guide at the start to explain how it was to be used using a Diary System Users Guide (See Appendix Nine) and every family was asked to complete the diary for one month and, if practical, the families were asked to try and make an entry to the diary every day. It was also important to gain the input of all family members regardless of age, gender or cognitive ability. The combined participative Arts-based approach aimed to achieve a blend between a narrative account and a creative Arts-based diary. This was achieved in the diary method by having a lined daily page for narrative data and a blank daily page for more creative non-written input (see Figure 4.3). This was strengthened by the use of scrap books and memento boxes as described below to form a creative participative Arts-based approach.

4.7.2 Interviews

Underpinning the use of diaries and throughout the data collection methods the researcher used interviews as a method (See Figure 4.2 above).

Interviews are one of the most frequently used methods in qualitative research and in particular, research in health-related subjects (Miczo 2003, Silverman 2011, cited in Saks and Alsop 2013). As a method, they gather data and provide a way of investigating that data through conversation. Interviews can be structured, semi structured or unstructured (Edwards and Holland 2013:12). Highly structured interviews deliver focused information around areas of interest to the researcher. This focused approach also allows for a more straightforward comparison of the data gathered. Unstructured interviews conversely allow for greater exploration of a subject following leads that participants offer (Green and Thorogood 2014, Saks and Alsop 2013).

In this study, the purpose of the interviews was to gather a wider understanding of the participant's responses to the process of keeping a salutogenicaly focused diary and the questions within the diary. During the interview, the researcher also planned to ask participants what the contents of the scrapbook and the memory box meant to them and therefore open questions were asked about each item's personal significance, meaning or memory links. There were risks that in this interpretive process that the Child's (or adult's) sense of meaning would be lost (Beauman 1997; Bendelow 1995). Therefore, careful consideration was taken of research in this area (Coad 2007) and advice taken from the researcher's Director of Studies, who was an acknowledged expert in this area. Finally, the interviews were concluded by asking the AI inspired question of what an 'ideal, dream service' would look like to the Children and their families.

A semi-structured approach was chosen to allow easier comparison across the participants. The diary methodology, used in the primary data collection, was then used to inform the structure of the semi-structured interview. The participants were contacted a week prior to the researcher visiting their home and asked to highlight what entries or items were most interesting or important to them. Focusing the questions around participant's choice of diary entries or "scraps" or mementoes strengthened the areas important to participants in the interview and in the overall data by allowing them to explain and reflect on what they had written (Kenten 2010 cited in Edwards and Holland 2013:59). This approach also gave respondents more power over what was discussed (Johnson 2002 cited in Saks and Alsop 2013:89). As this study was for a thesis rather than a large research grant, the researcher undertook the interviews**.** This had advantages in terms of the interviewer's knowledge of the study and understanding of the ethical issues and risks to the study's trustworthiness. However, this approach also had risks in terms of the researcher's investment in the study, which if not carefully monitored could pose a risk to the positionality of the researcher and the study itself. The researcher mitigated these risks using a research diary and close supervisory support.

4.7.3 Focus Groups

Focus groups were used within two events (see 4.2). Focus groups as a data collection method provide a compromise between the two most popular qualitative methods, one the process of interaction, gained by participant observation and two, the content of interaction gained by in-depth interviewing. As such the focus group methodology works in a different way, as it acknowledges the influence of the group. (Krueger 1994). Focus groups have been argued to gain a deeper understanding of issues by harnessing the group to explore the issues raised (Kitszinger 1997). There are many descriptions of focus groups, but the one that best describes the intent of this research is a "focus group is designed to obtain perceptions on a defined area of interest in a permissive non-threatening environment"; "the discussion is comfortable and often enjoyable for participants, as they share their ideas and perceptions" (Krueger 1994). This also fulfils an aim of being useful and enjoyable to the participants, although care must be taken to remember the purpose of these groups. The CCHSUKSG is primarily a support group and the risk of such groups being difficult to move from support to data collection has been identified (Straw and Smith 1995).

There are risks to the data with focus groups. Some researchers question the value of focus group data (Agar and MacDonald 1995). Focus group data is often misused with strongly verbalised views being attributed to the whole group (Sim 1998). Highly vocal individuals may monopolise the session or minority viewpoints may be silenced by the group. However, with good facilitation these quieter views can be captured, members of a focus group are constantly evaluating information and a member who says little may make a comment, which pinpoints the issue precisely (Sim 1998). One of the advantages of focus groups however, are that they do not discriminate against those with poor literacy skills. Sim goes on to suggest that a focus group methodology is more useful for gaining an overall feel for a subject than objective findings (Sim 1998).

The primary intention of focus groups within this study was a method of checking for data saturation, feeding back and checking out early themes and final findings. In this study focus groups were used at two points in the data collection process for checking out findings, which has been identified as a particular strength of this method of data collection. It offers a form of triangulation between individual views and a group consensus (Fusch and Ness 2015).

4.8 Arts-Based Methods

Arts-based methods were used as part of the phased approach with the Children with CCHS and their families to encourage engagement and included scrapbooks, mementos, and child focused Arts-based events (see 4.2) Each are set out in this section.

Arts-based methods remain an emergent field of research at the time of this study. Arts-based methods have been described as non linguistic methods and using non-language-based methods of expression and are commonly associated with narrative inquiry, poetry, music, performance, dance, movement and visual art (Coad 2007, Bagnoli 2009, Leavy 2009 cited in Eaves 2014:149). There has been a debate as to whether there should be a distinction between research, where the Arts-based is the primary research approach, and art-informed research, where art informs the approach (Springgay et al. 2005). Some writers feel the importance is not exclusively in the methods used, but rather the influence of art or Arts on methodology (Eaves 2014). This approach to the definition enables art-based methodology to be seen in its widest context, but draws on a specific approach, whilst Arts-based approaches is a wider understanding (Eaves 2014). This study used Arts-based in the widest meaning to support the use of scrapbooks, mementos and child-focused scenario-based play events. Each of these creative and inventive Arts-based methods will now be considered from both a methodological viewpoint and then in terms of their application in this study.

4.8.1 Scrapbooks

It was always envisaged that this study would actively include children in the study methods and that these would be engaging and would interest children across a wide age spectrum of ages, needs and cognitive ability (Carter 2005). The method of scrapbooking has been described as effective with children (Carter 2005, McCarthy, Sebaugh 2011). As an approach, it has a low barrier of entry in terms of the complexity of the activity, so is open to children from a wide range of cognitive abilities. Scrapbooking, as an activity, has wide social acceptance and its growth in online use suggests this is a medium that remains popular across generations. After much consideration using key literature (Carter 2005, McCarthy, Sebaugh 2011) a scrapbook was chosen as part of the suite of Arts-based methods.

Scrapbooks have over time constantly evolved; changing in popularity and usage (Good 2012). They have indeed been used in paper form since the early renaissance for the purpose of preserving and cataloguing mementos, drawings text, ideas and more recently photographs have appeared as part of the mix of objects displayed. Indeed, scrapbooks paved the way for the photograph album (Goodsell and Seiter 2011) . Scrapbooks have been described as “an everyday grappling with multiple realities” (Chalfen 1987 cited in Goodsell and Seiter 2011:318). The internet was viewed by many to be the demise of scrapbooks (Good 2012) but this was not so. In fact, an on-line scrapbook app ‘Pinterest’ had grown to over 70 million users by 2015 (Omnicoreagency 2018) which may indicate how popular and adaptable scrapbooking is. Some have argued all social media is a type of scrapbooking, where snippets of interest are pulled together to share with family and friends (Good 2012). Worldwide, there are over 2.2 billion monthly active ‘Facebook’ users (Zephoria 2018). One reflection of this wider view of scrap booking was that scrapbooks were reaching far more people than ever before (Good 2012).

Overall scrapbooks can be set apart from other collections of information by the assessment of the areas represented here. Firstly, they contain assembled personal media, to document friendship and a person’s social circle. A precursor to the modern scrap book was the “album amicorum” (friendship book), which was originally used mostly by students, to list and identify their contacts on their travels (Good 2012). As emigration increased, the idea of the friendship book evolved into the scrapbook. Interestingly, since their inception scrap books have been used to pull together artefacts that can illustrate a family history and as such can be used to “define family identity” (Goodsell and Seiter 2011:320) or a subject of interest to the person who created it. Some researchers have argued that scrapbooks have a higher level of significance and meaning and are a way of creating a family narrative. which aligns with the perceived dominant cultural family model. As such it can create “family capital” by showing alignment with this dominant cultural family model giving evidence of this link (Goodsell and Seiter 2011). Secondly, scrapbooks are a way of pulling together and indexing the “scraps” that are of interest. Often in the form of flat physical objects such as a baby’s hair, a child’s first drawing or a leaf pressed for collection. As forms of printed material expanded, people increasingly wanted ways of indexing things that were of interest to them, as the sources of such objects increased with mechanical printing (Good 2012). These scraps could be any flat, small, printed item that linked to key events in the life course of the collector’s family and friends, alternatively the scraps could relate to an interest or hobby.

Thirdly scrapbooks are a medium for “expressing taste”, where the user keeps links to the music or culture that they identify with, collecting tokens that link to that culture and demonstrate personal linkage with that culture such as tickets or photographs (Good 2012:566). The use of “selfies” where a person takes a photograph of themselves with a person or place that demonstrates their direct presence and link with their chosen person, band, place or culture is a modern extension of this phenomena.

In terms of research, scrapbooks have been identified as a data collection method which enable people to express feelings and views (Karns 2002). They have been used particularly in studies involving children and young people. Scrapbooks have been used to gather the views of children and young people in a novel way (Bragg and Buckingham 2008). One example is Carter (2005) who used a scrap book activity to evaluate a children’s community nursing service from a child’s perspective. In this study, scrapbooks were one of a suite of methods which were used to gain the views of children. The children’s scrapbooks contained “stories”, cameos of events and diary style entries (Carter 2005:53).

Scrapbooks have also been used in therapeutic services, such as bereavement support research, including being used as a therapeutic tool in paediatric oncology, where scrapbooks were used as a method of empowering families with children going through cancer treatment to increase positivity and motivation (McCarthy, Sebaugh 2011). Scrapbooks were therefore chosen as an Arts-based method, which would be particularly interesting to Children who may want to contribute to the data and study by providing “scraps” in their scrap book. This choice was supported by research, which found this approach to be effective for children with complex medical needs (Carter 2005) and with adolescents (McCarthy, Sebaugh 2011). Furthermore, the idea of the scrapbook fitted well with the concept of the diary. Diaries are a record of thoughts and feelings and scrapbooks are a collection of artefacts that are individual and connect memories.

4.8.1.1 Scrapbook Design

One consideration in design was whether to use an electronic version of the scrapbook, paper or offer both formats. An electronic online version was initially considered. 'Facebook' a popular social media tool was also considered in recognition of its wide existing use in the CCHSUKSG (who use a closed group Facebook page for social support and contact). However, Facebook had a clear usage as a social forum for the group, which was different to scrap booking, so was therefore discounted. An existing online app (Pinterest) which had a much closer format and connection to the concept of scrapbooking was identified for consideration. Pinterest has an electronic format closely aligned to the paper scrapbook concept allowing a user to pull together "scraps". The way they are pulled together within the app closely resembles a paper scrapbook. The two principle differences are that these "scraps can also include digital information in the form of pictures, word, videos and the like. The second is that this electronic scrap book can easily be shared with family and friends or the entire Pinterest community. At the time of the study Pinterest was not as well known in the UK and it was felt it may have a limited interest to children as its users are mostly adult. In addition, all social media type approaches have risks for Children in terms of unsolicited adult contact, so must be used with great care (Livingstone et al. 2011). A further consideration is that data protection issues are less in the researcher's control, widening the risk to participants (Townsend and Wallace 2016). Therefore, deliberation was given to the use of electronic data capture off line. Microsoft Word or PowerPoint were considered due to their accessibility; however research with scrap bookers has identified the need for a system that allows for natural interaction, such as touch and voice (Petrelli, Whittaker, Brockmeier 2008:2). After careful deliberation, reflection, and supervision, a decision was made not to consider an electronic scrapbook but rather solely a paper version (see Figure 4.4).

4.8.1.2 Scrapbook Format

Consideration was given to the possibility of adding the questions to the scrapbook, but a particular strength of scrapbooks is their individuality and creativity so after reflection and discussion at supervision a decision was made to keep the scrapbooks blank. Great care was taken to select colourful but non-age-related scrapbooks to ensure older children and siblings were not put off by a childish design, whilst engaging younger children, as per previous literature in this field.

McCarthy, Sebaugh (2011) and Carter (2005) identified the concept of a “scrapbook kit” which included pencils, glue, stick on eyes and other aids, but the decision was made after discussion with the researcher’s teams play specialist and supervision team, that the tools for scrapbook creation were widely available in family homes (see Figure 4.4).

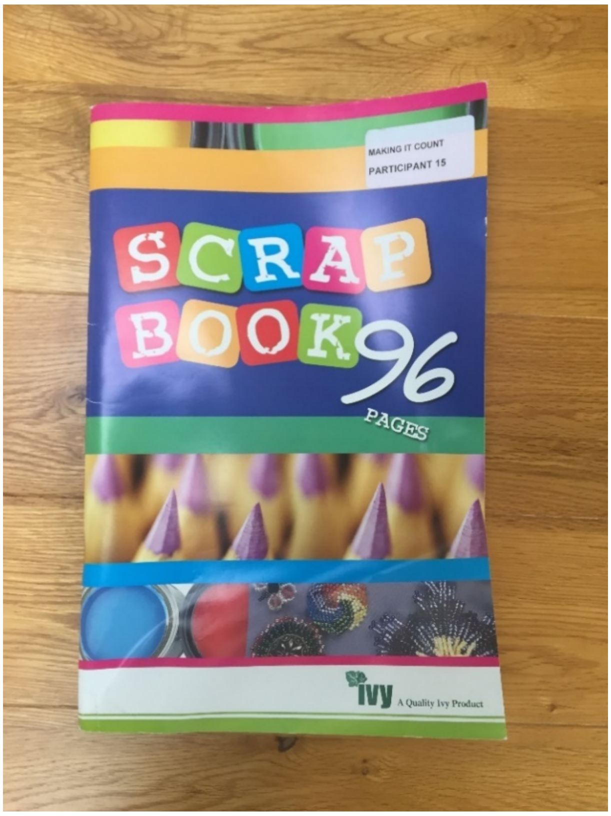


Figure 4.4 Example of the Scrapbook Provided to Participants

4.8.1.3 Scrapbook Data Collection

It was envisaged that the scrapbooks would be used as part of the family interview, to gain an understanding from the Child and other family members what the meaning of each entry was for them. The data in scrapbooks can be important to a Child and family, so each scrap book was photographed page by page and then left with the family. Carter (2005) used a process of electronic scanning but noted the issue of data loss in the scanning process Carter (2005). In this study scrapbooks were photographed, which avoided data loss through careful framing of the pictures, however Carter (2005) also noted the flattening of potential three-dimensional images by two-dimensional photography and the loss of texture of objects. This issue was impossible to avoid without taking scrapbooks off the Children, so the decision was made to photograph them in the family home.

4.8.2 Mementos

To widen the approach still further a Memento Box was added. A memento is described as "an object given or deliberately kept as a reminder of a person, place or event" (Petrelli, Whittaker, Brockmeier 2008:1). Mementos are important objects to their owner; they are heterogeneous and can be used as a discussion point to gain an understanding of the meaning and narrative behind the object from an owner's standpoint (Petrelli and Whittaker 2010). The meaning that people ascribe to a particular item of value to them has cross overs with scrapbooking, but has the advantage of being easier to manage. Mementos like items in scrapbooks are pictorial links to memories of value to the owner. Mementos are often not valuable or rare. A mass-produced article can have a particular value due to the link it provides to memories of the previous owner or user of the object. A memento equally can act as a link to an event or special time for its owner. The study of mementos is linked to narrative research, where the owner of the object describes how and why the object is important to them and the story behind that object (Petrelli and Whittaker 2010).

Memento researchers have a number of techniques to understand the narrative behind an object in context. A key approach in memento research is described as the "memory tour" (Petrelli, Whittaker, Brockmeier 2008:3). A memory tour is where the researcher is taken on a tour of the participant's home by the participant. The participant then describes where they store and place mementos and the value they ascribe to them. In memento research the placing of the object is of equal importance to the object itself. Therefore, an object's placement forms part of the story of the object. An object may be rarefied and placed in a central and visible position or hidden away in a dusty corner, thus the owner's story behind this placement is a key part of the narrative, that is attached to that memento (Petrelli and Whittaker 2010).

There was very little research at the time of this study into mementos in children, young people and their families. One family study found parents described as feeling they have a duty to collect mementos that chart the progress and success in children's lives. Mementos can be extremely varied and can range from a lock of a baby's hair to awards from activities undertaken by a child or young person. Mementos are often collected with an intention to pass them on to the child once they have grown up and have children of their own (Petrelli and Whittaker 2010).

Boxed collections in one study were found in 80 percent of the families' homes (Petrelli and Whittaker 2010). Boxed collections were rarely on display, but often were of particular value to respondents (Petrelli and Whittaker 2010). These boxes acted as a store of memories which when opened and described, started a wide and emotional discussion about their contents (Petrelli and Whittaker 2010). It is this link with memories and emotions which is linked with the other significant use of boxes of mementos with Children and families. Memory boxes have also been used extensively in grief counselling in Children. In the memory box approach, colourful boxes are provided to store items that link to the baby or child or relative who has died. This process of memorialising is now increasingly common in palliative care and often includes taking hand and foot impressions shortly after death (Tan et al. 2012). This powerful and emotionally charged use of memory boxes is different to memento research in terms of its aims but the usage is like memento boxes in the linkage with past memories and emotions.

4.8.2.1 Memento Box Design

In considering the use of this method, the first consideration was how it would be used and the applicability of this approach to the group this study aimed to recruit. Memory mapping was considered, but it was decided that this approach was too invasive to the family's privacy. In addition, the research supporting this approach was focused predominately on older adults. In this study this method was considered and chosen as a method to engage predominately children in the study. Thus, this method would need to be aimed primarily but not exclusively at engaging Children.

Electronic methods have been considered by researchers using mementos in two ways. The first, has been to identify how much digital data was selected for inclusion in the memory box. The research suggested that, despite the importance of digital data in daily life, digital material rarely is considered by participants as a memento, with only one digital memento being identified out of 169 mementos in a study of families (Petrelli and Whittaker 2010). The second, is the use of digital photographs as a form of memento box, by electronically capturing the image of a memento and recording its significance. In this approach, a digital photograph is taken of an object and the participant can tag the object as to its significance. However, these methods were excluded, due to the low level of evidence to support them and the strong association of the technique to older adults (Petrelli, Whittaker, Brockmeier 2008).

Research into grief and memory boxes had an interesting cross over and was supportive for the use of this approach in Children. The limited number of studies into memory boxes found strongly linked the collecting of mementos to the creation of memories in Children and their parents (Petrelli and Whittaker 2010).

In this study, the focus was different than for the memory boxes. The memento boxes used were to store objects that had been collected over the month of data collection. A box (see Figure 4.5) was selected which had a vibrant colour, but was not age specific and could be used by a child or adult.

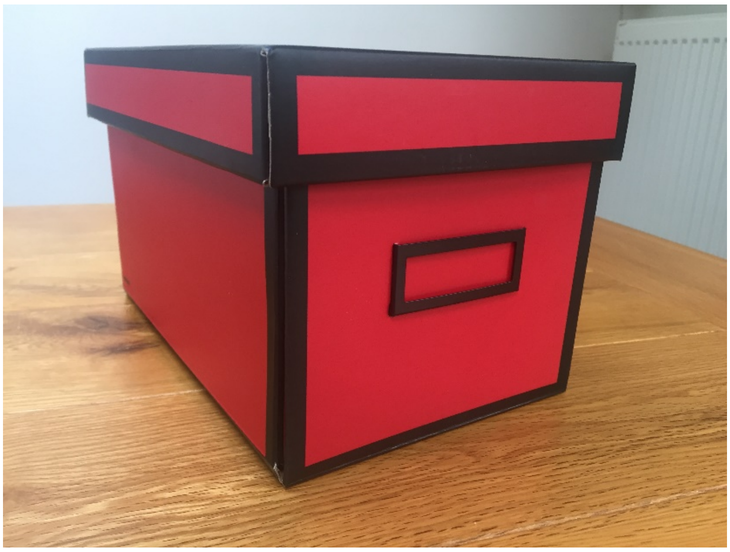


Figure 4.5 Example of the Memento Box Provided to Participants

Each family was provided with a memento box, with simple instructions for its use, as part of the participant pack of Arts-based methods. This tool was explained during the launch (Appendix 4).

4.8.3 Child Focused Arts-Based Events

The researcher felt that the Child's voice including the affected Children and unaffected siblings was an essential component of the families' lives and without their voice it would not be possible to gain an adequate or accurate account of what works in care packages. The lack of use of the scrapbooks and memento boxes by Children led to not only a rethink of the data collection tools used with Children, but also the very nature of Children's research.

Researchers who study Children can have differing views of Children in terms of Children's input to design. One view is that Children are social actors in their own right and" there is no automatic assumption that methodologies need to be adapted" (Robinson and Kellott 2004:86 cited in Fraser et al. 2004). However, more contemporary views are that Children are integral to the data collection and so use focused, sensitive and meaningful approaches to elicit Children's views (Coad 2007). Arts-based methods, in particular, have been identified as being effective in eliciting Children's views (Coad 2007). On reflection and through supervision, the researcher concluded that the creation and application of any child-focused participatory Arts-based methods had not considered sufficiently the specialist nature of actual Children in the research. A significant redesign therefore took place, which led to the creation of the participatory based play sessions explained below. The nature of the changes required revised ethical approval which was sought and received.

4.8.3.1 Participatory Play-Based Scenarios

Scenarios using imaginary situations were subsequently chosen as a fun, developmentally appropriate, approach for the children given that the children anticipated to attend were in the age range of four to ten years of age. This approach used was an adaption of the vignette method, but fitted well to AI and salutogenic frameworks in view of the positive stance.

In such events, vignettes are often used in social research with stories, scenarios or examples used to generate discussion amongst the participants to elicit perceptions, beliefs and attitudes (Hughes 1998:381). Traditionally, vignettes are a way of participants looking in on and reflecting on the stories or examples they are shown or told (Barter and Renold 1999). In this method, questions or focus group discussion follow on from the participants being shown or reading the vignette, however, some researchers have questioned this as giving participants selected information about an event by only giving them fixed questions, is not understanding the importance of human interaction (Barter and Renold 1999). The use of vignettes therefore, in qualitative research is focused on trying to gain participant's views of what was behind the actions described. However, there is also some evidence that vignettes do not always elicit responses to situations that are replicated in real-life (Hughes 1998). This significant critique contends that to try and understand what participants would do in a real event by questioning them about an imaginary scenario will not translate into real life (Barter and Renold 1999). Carlson (1996), Rahman (1996), Hughes (1998) Barter and Renold's (1999) literature suggested that there is a correlation between participant's responses to vignettes and what participants do in real situations. The lack of involvement of participants in the scenario has led to claims that this process is artificial as participants cannot change and respond to the vignettes as they progress (Barter and Renold 1999). Some researchers also suggest, that vignettes need to be close to real-life and mundane experiences to capture meaningful data (Finch 1987, Hughes 1998).

Other researchers, using vignette-based research in children, have suggested the more unfitting or contentious the vignette the more the children engaged (Hazel 1995). Therefore, it is important to consider not just the vignette approach, but the use of this approach specifically with children. For young children, it is particularly important to make vignettes easy to understand and to consider the flow of the story (Barter and Renold 1999). In this study, the researcher wanted to harness the capacity of children's imagination by moving them from outside to inside the story. Thus, a participatory scenario was used within play sessions to enable the children to become actors within the story. Although this approach is similar to a vignette, this is a novel development of the vignette method. By enabling the children to become participants in the scenario, the children could shape the story as actors within it. The children were making their views and comments as participants and not observers. The researcher made the stories very simple and was particularly observant to the flow of the stories.

A principle advantage of participatory research, like the scenario play events, is that it helps to address the issue of the power differential between adults and children (Robinson and Kellett 2004). A key issue in research, linked to this power differential, is that research with children should be undertaken by professionals skilled in working with Children. There has been a strong recognition of the need for different services for Children provided by Children's trained staff. As cited elsewhere, the researcher is a Consultant nurse and Registered Sick Children's Nurse. The team who supported the events were all Community Children's nurses who were Registered Sick Children's Nurses. The team also had qualified play specialists. This aided the team in their recognition of the power differential and in how they worked with and approached the children. Therefore, by using play, the Children became the principle actors in the scenarios and it was envisaged that there would be a reduction in the power differential between adults and Children. Play is the way Children make sense of the world around them and is a key component of Child development (Coad 2007). After the scenarios, free play sessions were put into the program with the research props, such as a tent, placed in the play room, enabling the Children through the medium of self/group play to express themselves freely.

In developing the scenarios, a link needed to be established to the area the researcher wished to study. Making a link between the three diary questions based on Antonovsky (1987) concepts of comprehensibility, manageability and meaningfulness was achieved by using the river analogy with the children (see Figure 4.6) described elsewhere (3.12),(Antonovsky 1987).

Figure 4.6 River Analogy Interpreted for Participatory Play Based Scenario Antonovsky 1987

Using this analogy meant the researcher was enabled to make a fun story out of a complex theoretical construct. In this way, it was hoped that it would be understandable to the young children attending when turned into a play scenario. Antonovsky's analogy was developed into two different scenarios over 2 time periods of 2014 and 2015. The first, was a direct interpretation of the river concept described in detail in 4.11., 4.3 and the second, was linked to the journey concept, but added in some breathing apparatus to make a link for the Children with their need for ventilation (discussed in detail below 4.11.4.3). An overview of each is presented in 4.8.3.1 and 4.8.3.2 prior to further discussion of the event.

4.8.3.2 Scenario One: 2014

Scenario one was used in the first children's participatory research session (4.6) and developed from the analogy of the river (see 3.12) (Antonovsky 1987) which was developed into going on a camping trip. This consisted of a trip on a boat up a river to a deserted island, with a tent and an imaginary fire. The researcher developed props for the scenario, including an eight-metre-long river made of tarpaulin. An inflatable dingy mounted on wheels, and a pop-up tent and an artificial fire for the camp at the end of the river were also used. These props were set up in the corridor of the conference centre, whilst the children were being entertained in the playroom (Figure 4.6). The conference centre corridor was a secure part of the hotel's conference suites and the CCHS group were the only group in the suites that weekend, making the area secure for children's activities. This is described in detail in 4.11.4.3.

4.8.3.3 Scenario Two: 2015

Scenario two was used in 2015 and had a similar start with the paper suitcases being filled with drawings by the children of what they would take on a camping trip. To link the water base and the river concept, outlined by Antonovsky, a diver comes and leads an interactive story created jointly with the children. The use of a figure in the scenario who is dependent on breathing equipment, was used to prompt a discussion about using breathing equipment when asleep (the diver informs the children that he is sleeping under the river, described in detail in 4.11.4.3. The link with the river scenario remained clear, but this was developed upon into a more extreme interactive scenario to test the contention that more unusual vignettes gain a better response from children (Hazel 1995 cited in Barter and Renold 1999). This was then envisaged to be a lead in to a discussion with the children relating to what they had put in their suitcases (Described in detail in 4.11.4.3).

4.8.3.4 Conclusion of Arts-Based Methods

In conclusion, the first group of participatory Arts-based tools were therefore an innovative mix of scrapbooks, memento boxes and child-focused events using scenarios and play. The scrapbooks and memento boxes were used simultaneously to underpin the diaries and for participants to allow them to represent, in any way they wished, their answers to three questions posed, ether as text, pictures or objects. The researcher wanted to use a participatory Arts-based approach in its widest terms, including objects, photos, pictures, and text, all used together to "support active remembering with multiple types of objects that can be appropriate in highly flexible ways" (Petrelli, Whittaker, Brockmeier 2008:10). Separately, the child-focused Arts-based events were used to ensure the Children's voices were heard. Whilst these seem like eclectic approaches to data collection, this was important to explore the participants' views and widen the potential for data collection. It can increase the *burden* on participants, as well as adding to the complexity of the analysis (Thurmond 2001) but whilst this was considered by the researcher it remained very important to do this mix of participatory Arts-based methods. These issues are addressed in 4.9 - 4.11 and the analysis 4.12.3 - 4.12.6.

4.8.4 Summary of Data Collection Methods

Overall the data collection methodology consisted of using diaries, participatory creative Arts-based tools (scrapbooks, visual Arts-based and memento) and were underpinned by 'conventional' research methods such as interviews and focus groups (Figure 4.7). These phased methods aimed to collate a wide data set, that located views of the children and families in their day to day experience. Each method has been considered in depth as a key component of the quality process, in reference to the quality framework described by Flick (2009) in Appendix 13. Whilst the multiple methods employed could be seen as complex, they were needed to capture the whole family's views in a fun and engaging way.

Figure 4.7 Arts-Based Participant Pack

4.9 Recruitment and Sampling

As a national group, the CCHSUKSG has members across the four countries of the UK. The group runs family weekends on a bi-annual basis. Prior to this study, the last weekend had been in August 2012, a year before the data collection and no new meeting was planned. However, the group was interested in the suggestion of having a one-day meeting in 2013, but the Chair of the group did not have the capacity to organise this additional meeting, so offered the opportunity to the researcher to organise this instead. This enabled the researcher to present the study to members, with prior permission for this approach, from the group Chair. The researcher worked with the Chair of the group to balance the needs of the attendees and the research. A modern hotel in the Midlands was selected, as it balanced the needs of the group for good value, accessible hotel rooms and nearby leisure facilities with the needs of the research to have conference rooms and facilities.

The researcher arranged provision of audio-visual equipment (which was not available at this hotel). Time was purchased for play therapists and nurses working within a local NHS Trust to set up and run a playroom. The staff were selected for their skills with working with children, but also because they had training in CCHS and therefore could respond appropriately and alert parents if there were any issues within the playroom**.** The playroom was open to all the children, young people and siblings who attended the day. No research was undertaken with the children or young people during the launch event. The researcher also worked, with the Chair of the group, to approach charities and ventilator companies, to access additional funding to provide free tickets for the families to a nearby theme park. The theme park tickets were provided to the Chair of the CCHSUKSG, and all families (whether involved in the research or not) were given a free theme park ticket.

4.10 Conduct of the Research Launch

In this section the conduct of the research is described in full to enable an in depth understanding of the approach taken and as a key component of the Quality review (Flick 2009).

The day was organised and opened by the Chair of the group. There were a variety of speakers and events arranged by the Chair throughout the day. The Agenda was a mix of professional talks and social activities. This was in keeping with the established format for CCHSUKSG days and had been followed for many years.

The researcher was allocated a 45-minute session before lunch. The Chair introduced the researcher as a research student from Coventry University. The researcher introduced the parent representative who was acting as a research assistant for the day. The parent representative explained that her interest in this study was from her personal experience gained from having a LTV child of her own. The introduction was achieved with presentation (jointly undertaken with the parent representative) which gave an overview of the research project (Appendix 4). The presentation very simply outlined a salutogenic theoretical approach (see 4.11.4.3). This was considered important as the five steps of using AI had shown the importance of “choosing the positive as the focus of inquiry” (see 4.4). This clarified to the researcher the importance of preparing potential participants to think in a different way, when using methodologies such as salutogenic theory and AI. The presentation then outlined current research in LTV and the gaps in the current evidence base, which this study sought to address. The data collection tools were described, and the process of data collection was outlined.

Members of the group were asked if they had any questions around the research. Practical and process questions were answered. The researcher was, however, concerned that having one to one conversations about the diaries, would influence the content of the diaries and in turn the findings. Therefore, a decision was made to conduct a general, open group discussion to allow the group themselves to clarify the questions raised by other group members. This approach was very effective, as the group knew each other well and were comfortable discussing issues together.

The researcher thanked the group and advised them that, if anyone was interested in taking part in the research, both himself and the research assistant would be around at lunch and for the rest of the day. The Chair then thanked the researcher and the research assistant and outlined the program for the rest of the day and broke for lunch.

Despite the process described above, being integral to the day and a play room being provided, the researcher was still concerned about a perceived pressure on group members to take part in the study. Recruitment was therefore undertaken in a gentle, unobtrusive way. Group members were left to approach the research team and express an interest. Once an interest had been expressed the research was explained in more detail and if the group member remained interested in taking part in the study, information leaflets were given out (Appendix 5). The interested group member was advised to take some time to look at information leaflets and to either come back later in the day, or if they had further questions to come back to the research team for a further discussion. If a group member came back the freedom not to participate, or to leave the study at any point, was re-explained and if still content to continue the group member signed the consent form and became a study participant. At this point the data collection tools were given to the participant and the next steps explained. The participant was thanked and reassured again that they could leave the study at any point.

Additional recruitment strategies were also employed. Following recruitment during the diary data collection period, the researcher attempted to balance chasing responses with not pestering participants (and thus jeopardising the concept of free will). In all cases at the halfway point through the diary, scrapbook and memento box data collection period phone calls were placed to families. Most families responded but as some had not started the data collection, they were reassured that the start date did not matter, but also offered the opportunity to not take part in the study. Four families subsequently did not complete the diary and decided to leave the study. The remaining eight completed all part of the study.

One week prior to the final family interview, the researcher phoned, texted and emailed each family to confirm the time of the visit and to ask them to highlight in their diaries, scrapbooks and memento box, the entries most significant to them. The researcher considered it important to conduct the interviews face-to-face. This did pose some logistical issues however the families all indicated that they preferred this approach, and all were generous in their time during interviews.

In qualitative research, there is debate about what size a sample should be. Some researchers have attempted to put a number on the appropriate sample size for a qualitative study (Adler and Adler 2012). Others make the point that it is not the number of participants in a qualitative study it is the concept you are trying to study (Corbin and Strauss 1990). Glaser and Strauss (1967) describe thematic saturation as a process, where recruiting to the study ceases, when no new themes are emerging. The numbers of participants in this study are summarised in the table below.

Arts-Based Sample Size.
Diary and Interview numbers

- 12 families signed up at the launch.
- 4 families dropped out.
- 8 families competed the diary, interviews and focus groups.

Box 4.1 Summary of Participants Who Took Part in the Diary Data Collection Process

Issues of sample size and a consideration of whether data saturation was achieved will be returned to in the analysis section of this chapter (4.13.3).

In summary, the recruitment to this study was greatly aided by the support of the CCHSUKSG. The similarities between Children with CCHS enabled a group with more similarity than a cross section of LTV Children, but the rareness of the condition would have made recruitment without the CCHSUKSG highly problematic. The infrastructure provided by the group made launching simpler and less intrusive. The overall recruitment and retention of the participants was excellent.

4.11 Conduct of the Data Collection

Having explained the methods in detail, along with the ethics and recruitment strategies, the methods will now be critically reflected upon in terms of conduct and using them in reality. Further, this review of the conduct of data collection provides in depth description to expand the review of the methods in terms of quality outlined above and summarised in Appendix 13.

4.11.1 Using the Diaries

The diaries worked well for the parents, however the drawing pages were not used by the young children. No siblings or older children wrote in the diary except the oldest participant in the study (participant seven) who completed their own diary. The written data collected was extensive with over eleven thousand words contributed in the diaries. This data was rich and contextual, which was a key aim of this approach. By moving away from a one-off interview, to a reflection over a month, the daily reality of participant's lives was vividly illustrated. The first two questions below were widely considered by participants as understandable and contributed the majority of the diary data.

What are the best things that have happened today?
Things that make life/care easier?

However, the third question, (*things that make life/care clearer or easier to understand?*) was found by many participants as either confusing or something that did not happen every day. One participant termed it the "eureka question" (Participant Five) which in many ways encapsulates the meaning of comprehensibility. Those families who had a child who had been diagnosed more recently found this question easier, as how they comprehended their lives was under significant adaptation. Conversely those who had a Child who had been diagnosed for many years and were established in their understanding and adaptation to life with CCHS, (indeed the young person in the study and his family struggled to remember life without CCHS), struggled to answer this question. There were no significant issues with reading entries and the diaries collected in paper form were easily converted into electronic data. An interesting aspect of this data collection method was the personal value some respondents placed upon keeping a diary of what was going well. Half the families reviewed their perception of the month during the process of reviewing the diary in preparation for the interview. This fitted well with the researcher's approach of aiming to do more than "do no harm".

In summary, as a researcher with a practical background in nursing, the researcher had committed to an approach of trying to choose methods that would reflect "human flourishing" (Fowler 1988:111). As a method, the diary tool collected useful data in the written form. The diary methodology was a good fit for this study and achieved the aims of gaining a sense of what day to day life was like for participants. It also got underneath the stressful events that dominated their recollection at interview (to gain a real sense of the day to day reality of family life). The diaries, however, failed to evolve into the wider participatory art-based approach, which had been envisaged with no children participating. The daily page for submission of drawings were not used and the diary tool became far more word based than envisaged. The researcher reflected on this and was pleased that additional methods for collecting non-word-based data had been used (See 4.11.4).

4.11.2 Using the Interviews

In reality, the researcher used two (known here as Round One and Two) interviews to support data collection (Figure 4.2). In the interviews a common approach was taken, in order to enhance choice and engagement, where each participant chose the time of their interview and location. All the interviews took place at the participants' homes, except for one first phase interview, where the participant chose a local children's hospice and for one phase two interview, where the participant chose the CCHSUKSG day. However, it was really in the participant's home where the researcher felt most comfortable and the participants had enhanced control over the interview process. In this way, it was felt that "the hierarchy which can be experienced, created and enacted in places" was reduced (Edwards and Holland 2013:44). Additionally, by visiting families at home, a real sense of their lives was apparent, not just from their words, but from their home environments. The journey to their homes also made the context of their lives real, from the isolation of the Scottish Highlands and the east coast, to the busy city life of cities in the United Kingdom. For those who lived far away, the act of travelling to them was an acknowledgement of them and was valued by the families.

Whilst being in the home, other family members were often present and two interviews had both parents participating. This choice allowed more control for participants and a joint space to reflect. However, there are methodological differences with this approach as it has additional social interaction (Edwards and Holland 2013:48) which was acknowledged by the researcher, as a different dimension in these interviews. The environment itself added to the discussion, with cues of what was important to each family and links to the Child's condition, such as adaptations being a prominent part of each respondent's home environment (allowing an additional dimension of “the material culture of their chosen space”, (Edwards and Holland 2013:55). In interviewing participants face to face, there was the advantage of being able to respond to non-verbal as well as verbal responses. Interestingly, there were also disadvantages from home-based interviews, with distractions from young siblings or family pets being a feature of some of the interviews. The recordings of the interviews however show that families quickly refocused on the interview, regardless of these distractions. As parents of children or young people with complex needs, there seemed a sense that interruption was a constant feature of their lives, which they had adapted to and could quickly refocus.

4.11.3.1 Round One Interviews 2013-2014

Prior to commencing the first interview, consent for tape-recording the interview was confirmed. Initially the families were asked about their experience of keeping the diary. Following on, the researcher went through their diaries (scrapbooks, memento box entries if used) with particular reference to those that they had highlighted as significant, seeking clarification, and further exploring the issues raised. Each family were asked since diagnosis, what would be the most important entries that they would have made in the diary in each of the three question areas. Finally, they were asked to describe the dream approach/package (Cooperrider, Whitney, Stavros 2009) that would enable them to have the most positive entries in their diaries, scrapbooks, and memento boxes. The interviews ranged from 35 to 55 minutes in length.

4.11.3.2 Round Two of Interviews 2015-2016

At round two, prior to commencing the interview, consent for tape-recording the interview was reconfirmed. The same format was used within each of the three conceptual areas and the findings related to each concept considered in turn. Participants were asked if they felt if the findings were still felt to be a true reflection of their experiences and were given time to discuss each conceptual area or give further examples. Audio review was undertaken of the interview, rather than full transcription (this was in keeping with the focus group approach discussed below).

In summary using the interviews at two rounds, there may have been a risk that by using a familiar and well-known technique, such as interviews, that this approach would reduce the value of the participatory Arts-based methods envisaged in this methodology. However, by basing the interview questions on the participants’ highlighted responses from the diaries or descriptions of scrapbook entries or mementos, it was envisaged that, the interviews would become the platform for exploring the Arts-based derived data. However, the round one Arts-based methods failed to produce enough data for this to be tested. The approach taken of getting participants to highlight their most important diary entries helped to avoid the interview questions exploring researcher assumptions, rather than participants' experience derived from their data. Questions that arose from the participants' accounts were important as their words were individual and centred on their own worlds.

Another positive, but unexpected, result of this approach was that it felt like the participants went through a process of reflection based on their diary entries. Some of them made this comment to the researcher and that the reflective process in itself became an important event. Furthermore, the interviews enabled a one to one in-depth discussion to occur, allowing a relationship to be created between the researcher and participant. These relationships lasted throughout the research process.

4.11.4 Using Focus Groups

The primary use of focus groups was a method of checking for data saturation, feeding back and checking out early themes and final findings. In the study, focus groups were used at two points (Figure 4.2) and will be known here as Focus Group One and Two.

Focus Group One 2014

This first focus group was held to test out the initial themes and whether data saturation had been achieved. Five of the eight families were present. Although this was not the whole group, the researcher considered this in supervision and did not go wider across the group as this was only the first of two-member checks. Each theme was set out on a poster with the codes and sub themes related to it and were hung on the walls of a room at the 2014 CCHSUKSG Conference. Each participant was invited to look at the thematic areas and supporting codes and then the focus group was run with each of the thematic areas and each of the codes within them being considered and discussed. There was a significant number of themes and codes to be discussed limiting the depth of the analysis which was a weakness of this approach. Participants, however, recognised the themes and codes described and gave extra examples and depth to the codes and the researcher's understanding of them. There was, however, a discussion of how there would have been other themes, if the research had included problems and issues. The issues, noted by families, were a lack of cover in care packages. Lack of consistency of staff and commissioning a service that lacked personalisation were, interestingly, both areas that were covered in the findings, but in terms of their possible solution rather than underlying issues. The synthesis of dream approaches, collected in response to the AI inspired question in the phase one interviews, was welcomed as an accurate and helpful future vision for LTV services.

Focus Group Two 2015

A second focus group was conducted at the 2015 CCHSUKSG family day, attended by five families, after the initial results had been exposed to an in-depth analytical and theoretical review of the Focus Group One. The purpose of this focus group was to act as a member check, to ensure analysis had not moved away from the family's experience during thematic review. Each theme was considered and reviewed. The researcher read and explained each theme in turn and the participants discussed each theme in turn. The participants were happy with the new themes and felt they still represented what they had shared in diaries and interviews.

In summary, the experience of focus groups in this study was that they were useful for member checking data. The group dynamic did achieve a discussion; however, this discussion soon became dominated by problems and issues. This was interesting, as when reflecting on the data collected in the diaries and interviews, the participants were surprised at the contrast between their positive diary entries and the problem orientated focus they adopted when together. It is possible that by running the focus groups at CCHSUKSG days, that this enhanced the group dynamic that had built up through the weekend. Another possibility was that the researcher failed to steer the focus group onto the research agenda sufficiently. As a researcher there was a dilemma, as the researcher was reluctant to oversteer conversations, as that would be to subvert the freedom of the group to shape the data themselves, therefore such an approach would have been incompatible with the overall approach taken in the research. On reflection, if time and practicalities had allowed, one to one interviews at all stages rather than focus groups interviews would have been used. The 2015 focus group was particularly interesting in this regard, as it could be compared with the participants who did not attend and were therefore interviewed at home. The one to one home interviews were significantly more focused with less prompting. This appeared to be to be due to the one to one nature of home interviews, versus the group dynamic of the focus group. It is also a reflection of the researcher, that the group dynamic of the focus group did not offer the depth, context or richness of the diaries and home interviews and this validated the decision to limit focus group use to member checking data. The experience of the focus groups supported the approach taken in this study of using them to consider findings, rather than as a data collection tool.

4.11.5 Using Arts-Based Methods

A reflection of the scrapbooks and mementos will be firstly undertaken followed by the additional child focused Arts-based methods.

4.11.5.1 Using Scrap Books

Scrap books had been devised as a key component of the initial Arts-based approach, as discussed elsewhere (4.8.1 - 4.8.1.3). However, in reality, the scrapbooks were only used by one family in this study and then only for the first two days. Therefore, the potential of scrapbooks may not have been fully realised in this study. As described elsewhere, scrapbooks have been effectively used before in research (McCarthy, Sebaugh 2011) so this was disappointing. On reflection, this was either the applicability or application of the method which was ineffective or unsuitable. In this study, they may just have been too much for busy Children and families to complete alongside the diaries. There was already a space on every diary page for drawings (however never used). In terms of application there was nothing printed in the scrapbooks in terms of questions or activities, which could have either confused participants, or led them to believe it was an unimportant part of the data collection. Carter (2005) specifically mentioned the need for a scrapbook tool kit to get users started on reflection and omitting this could have been an oversight. The opportunity for using these tools in the playroom to gain an interest in them from the children was missed and when reflected upon, this approach was used to very good effect later in the study in the children's scenarios.

4.11.5.2 Using Memento Boxes

This method was even less effective with no mementos being collected. The researcher has no regret in attempting to use this approach, but on critical reflection, now feels that most of the critique outlined above around scrapbooks would also apply to using mementos. The main difference in memento research is its unfamiliarity to participants. Reflecting back, it may have been that the researcher did not spend enough time explaining this method and the opportunity for using the play sessions to introduce it was also missed. Most families showed the researcher the Child's adapted room on the home visits, so there was real potential for adding this as data in the form of a memory tour, however, there was no ethical approval for this approach sought or given, so the opportunity was not pursued.

4.11.5.3 Undertaking Participatory Play-Based Scenarios

As described elsewhere, two child-focused participatory play-based scenarios were used at two points (Figure 4.2) and will be known here as Event One and Two.

Conducting the 1st Children's Event (2014)

The Children's research was based in a room next to the main meeting room booked by the CCHSUKSG. The researcher briefed the team from a local NHS trust of children's nurses and play specialists on their role in the event. The team were briefed on the scenarios and their role in recording written notes on the event and supporting the children. The team also had a health risk assessment and the skills to deal with medical emergencies (as in if a child with CCHS falls asleep or becomes unconscious, they cease breathing and need ventilating. The children with tracheostomies could require suctioning or an emergency tube change). This was a key area, where the professional responsibilities of a nurse changed the support provided, but these skills being present in the playroom also allowed the children to enjoy the session free from parental oversight. This in turn had significant advantages to the research, in terms of gaining the child rather than parental voice.

The researcher gave a presentation to the CCHSUKSG about the children's participatory scenarios. At the 2014 and 2015 CCHSUKSG conferences (outlined in Appendix 10), the researcher explained that all children and young people were free to come to the playroom and take part in the participatory scenarios, and that data would only be collected from children and young people who had agreed to take part and whose parents had consented. 17 Children attended the event, six from the families in the diary group and 14 others took part in the research which was felt to be a good cohort to inform the study.

On arrival at the playroom, Children were asked, in an age appropriate way if they would like to take part in the study. If they chose not to, they went straight in, if they were happy with taking part, the parents were asked if they were happy for their child or young person to take part and again if not, they proceeded into the playroom, without taking part in the study. If the parents expressed an interest, they were taken through what was happening and given the study information form to read (Appendix 6.) If they were content for their Child to take part, they were advised that the Child or themselves could withdraw the Child from the study at any time and still carry on with any activities if they wished. They were then given the form to read and sign and then once consent was gained the Child joined the playroom as a participant, otherwise they simply joined the activities. No Children refused to participate and equally there was no evidence of parents applying peer pressure on each other. Two of the older children, who were ten and 11, chose later in the day not to join in the scenario.

Figure 4.8 One of the Children Creating their Suitcase (reproduced with permission)

The children in the morning had free playtime, in a playroom, staffed by the research team. Each child, during the morning, at a time of their choosing, was asked to draw on a paper suitcase, what they would take with them on a camping trip. One of the research team then discussed the contents of the suitcase with the child and took field notes. The children also made Arts-based materials during the morning to go in the river, which included fish, crabs and shells.

Figure 4.9 Children with their Suitcases at Start of Scenario One (reproduced with permission)

In the afternoon, in groups of four, they were taken on a trip up the imaginary river. The children then put on high visibility jackets as imaginary life jackets and then with their suitcase, got into the boat. The trip they were going to take was explained. The children became very involved and excited about the trip. The river was filled with themed pictures, that they had drawn in the morning of fish and crabs and anything else they had wished to draw.

Half way along the river, they encountered a terrible storm, the boat was rocked around, and each child was asked what they would save from their suitcase. This question aimed to determine what the children valued most in their suitcase, allowing an interpretation of priority, of what they had put in their suitcase (rather than direct questioning). In practice this form of enquiry worked well. The next part of the scenario explained that fortunately, the storm abates, and the children reach the island. After some play, they sit around the campfire, where they discuss with the children from the boat and the researcher, what they have brought with them in their suitcase. After some more play, they are asked what or who they are looking forward to getting back to. The children then took a boat trip back.

After the scenario was finished, the tents were put in the playroom for the children to have a free play session, which was observed, and key points recorded by the research team.

Figure 4.10 Children Enjoying Free Play with Research Props after Scenario One (reproduced with permission)

Overall, the children were highly engaged in all aspects of the session. The material they produced was extensive. Their comments and participation produced rich and plentiful data. The skill of the play specialists and children’s nurses in the research team was key to the conduct, timing, safety and effectiveness of the session. At the end of the session the research assistants were all debriefed by the researcher. The debrief consisted of a general discussion, then a specific discussion around the creation of the suitcases, the boat trip and the free play session.

Conducting the 2nd Children's Event (2015)

The conduct of the setup of the second event followed the preparation plan for scenario one. The main change implemented was that the team had a much more detailed research briefing, as after the first event the researcher's reflections had noted, that on occasions, it appeared some of the research assistants had prompted children in a desire to help them. The researcher asked the research assistants to avoid this and provided ways of supporting children without steering their responses (in the event the researcher managed to directly work with each child and young person).

14 Children attended; six from families who took part in the diaries and interviews and in total 11 took part in the research. The event began with a repeat of the consent process and then opened with the paper suitcases being filled with drawings by the children of what they would take on a camping trip. The researcher sat with each Child, during the creation of their suitcase to take notes and to maintain a consistent approach to this key data collection point.

The Children in the second scenario, instead of a boat trip, met the researcher who had a suitcase with him. The Children sat in a circle and the researcher set the scene for the participatory scenario, at first, he brought out all sorts of strange things that got the Children laughing and interacting with the researcher and adding to the story. The researcher then brought in the outlandish element, to test the assertion that outlandish scenarios work better with Children (described in 4.8.3). The researcher told the Children that he is planning on going to sleep at the bottom of the lake overnight, as he likes sleeping with the fish. This approach worked surprisingly well, with the Children quickly responding, joining in and laughing, asking how he was going to breath underwater.

Figure 4.11 Children and Researcher in Discussion Scenario Two (reproduced with permission)

The researcher then put on a full set of diving equipment and described how it worked. There was then a discussion about whether the Children needed anything special to breathe at night. The Children, spontaneously, and in great detail discussed CCHS and what they needed when they were asleep at night. The Children's theoretical understanding of what they needed was excellent. The Children were then asked what they had in their suitcases and whether they felt they had everything they needed for an overnight camping trip. None of the Children had brought any ventilation equipment. When asked why they hadn't brought any equipment, just as in scenario one, they explained they had their mummies so didn't need to bring it. After the discussion, the Children had an opportunity to dress up as divers, which was very well received and allowed ongoing roleplay and discussion in a free play environment.

Figure 4.12 Children Play with Research Props Scenario Two (reproduced with permission)

No Child or their parents declined being part of the study and none of the Children indicated at any point that they wished to leave. The scenario used in 2015 seemed of interest to a wider age range than scenario one and the group discussion it facilitated was particularly helpful. The reasons for this might be the researcher's growing confidence, improved briefing, or the outrageous participatory scenario. It may have also helped that the Children were more familiar with the researcher/team and vice versa.

4.11.6 Final Reflections of using Participatory Arts-Based Methods

Overall, on reflection, the participatory Arts-based methods had both positive and challenging issues. The vision of the scrapbooks and mementos working together to underpin the diary were felt to be less successful. The researcher had hoped the Children would be involved in the diaries but in reality, this was not the case. None of the Children or families used the daily drawing page in the diary and only one Child entry was submitted from all the participants, in the form of a scrapbook with two drawings. None of the participants chose to use the memento box. Therefore, the scrapbooks and memento boxes were limited in effectiveness, in enhancing the diaries to collate a wider participatory Arts-based approach which involved the whole family.

This led to a significant period of reflection and the researcher began to do this from a Child's perspective, including going back to the underpinning beliefs of the research process. One issue that the researcher considered was that, in approaching Children and adults together at the initial launch, they had become seen as one large group. The researcher did not offer the opportunity to their Children for discussion and had instead been focused on their parents. This was carefully considered by the researcher as significant and hence the Children's participatory Arts-based scenario methods strengthened in development.

On reflection, once the researcher began rethinking, the two Arts-based participatory scenario-based play sessions worked so much better than could have been anticipated. The creative approaches taken were effective in taking the concept of a vignette, where participants comment on a scenario from the outside, to participatory scenarios where the Children became actors in the scenario and contributed from within the scenario. The inclusion of free play allowed this expression to develop in any way the Children wished. This innovative approach, although carefully designed to be fun and safe for the Children, had risks as a data collection approach. To the researcher's knowledge, this approach has never been used in a research study before, so this was novel learning.

The scenarios exceeded initial expectation and have identified a new potential approach to using vignettes with younger children. Having the suitcases as a common data collection tool in both scenarios, allowed a good correlation between the two. The importance of recruiting Children's trained nursing staff was critical, in terms of safety, with medical events occurring during both Children's days and being swiftly dealt with on both occasions. The importance of play specialists to the timing and conduct of the research activity was invaluable. The main learning to come out of reflection on the Arts- based participatory play scenarios, was the importance of extensive preparation of the playroom staff as research support staff. Data collection in Scenario One, in terms of the suitcases, appeared to have at times included too much direct questioning. During Scenario Two, a much clearer process of training and briefing was undertaken, and the researcher was the primary data collector. Overall, both sessions were enjoyed by all the Children with everyone joining in and having fun. The data gathered was significant and will be considered in the results and analysis section. The more outlandish approach taken in the second scenario, with the introduction of a diver, was even more successful than the first (supporting Hazel's (1995) assertion that inappropriate or controversial approaches worked better in Children).

Overall, the initial mix of participatory Arts-based methods gathered a wide and useful data set from participating adults and the failure of the scrapbooks and memento boxes, although disappointing on review, led to a far stronger art-based approach being introduced. These methods were instrumental in achieving quality through the research process (Flick 2009), and a strong family data set.

4.12 Overall Summary of the Conduct of the Data Collection

The methods, eventually, worked well together and were a good fit with the underpinning theoretical base of salutogenic theory and AI. The researcher felt that the aims of the study were met, by using the eclectic approaches chosen, developed, and used. A Quality review approach (Flick 2009), based upon a reflective approach was important throughout (see Appendix 13), and especially during any adaptations, which were vital to get the right methods for this study.

The diary was highly effective for engaging adults, as was the scenario approach for Children. This novel mix of methods was enabled by robust supervision, reflection and reflexivity. The diary was also the most successful method in gaining data from parents. This approach helped the parents to explore their contextual situations. As a positive reflective framework, it was based upon the salutogenic theory and AI and was effective in collecting what worked in participants' daily lives. The diary enabled the researcher to get a picture of what day to day life was like and help facilitate recollection at interview. The close link to the underpinning theoretical approach worked well and enabled new and novel approach and findings. The theoretical link was also helpful in analysis.

Diaries proved highly effective at getting parental insights of life with a Child dependant on LTV. However, the wider vision of an Arts based eclectic diary was not realised, as the scrapbooking and memento box methods were unsuccessful in this study. However, on reflection, it is likely that the lack of use of these approaches was linked to insufficient attention being paid to their use in the launch. Diaries worked well in obtaining family derived questions to explore in Interview. Interviews were effective for probing deeper into the diaries and checking into the findings in depth. They also were used to ask the AI inspired question around what a dream service should look like. They were time consuming, but vital for the inspection of this data and in recognising the importance of the participants and established the relationships that were so important for a study that crossed three years. Focus groups were disappointing, as the participants easily slipped into the problem orientated discourse, as such their use only to check out data was validated. The new approach to scenario research developed for this study of participatory play-based scenarios, was a risk as this was a new method, however, they were highly effective and enabled the Children to become fully involved in the research and by doing so enabled the overall vision of a Child and family dataset.

Quality review was integral throughout the study (Flick 2009), enabling a firm base for each stage of development. Without a thorough process of reflection, review, member checking and ongoing development of the approach, there was a real risk that the children's voice would have been absent from the data. The on-going quality review cycle (Flick 2009) provided evidence to review the methods and enable a child focused approach enacted through the participatory play-based scenarios, which was instrumental in gaining the rich family data set that was subsequently achieved.

The final mix of methods worked well at getting a Child and family viewpoint and was highly effective at gaining a Child's family-based perspective of life at home with LTV and its management. Importantly the approaches utilised, realised the aim of gaining a rich salutogenic solution focused data set rather than a problem-based discourse.

4.13 Data Analysis

Data analysis is an essential component of the trustworthiness and quality of the research. Health studies, in particular, have been criticised for not being open and explicit about data analysis (Flick 2009, Saks and Alsop 2013). Fundamental to the openness and quality framework within this study, is a clear linkage and critique of the study's ontological orientation. As discussed in 4.2 this study has been based within the qualitative paradigm.

Qualitative studies as well as identifying the story of the participants, aim to identify the deeper meanings behind their narrative. It is therefore important to demonstrate not only comprehension of the phenomena being researched, but also insight into how the participant constructs within their lives each phenomenon identified (Green and Thorogood 2014). Qualitative research analysis has been identified both as a science and an art (Whittlemore, Chase and Mandle 2001). The creativity of the approach allows innovation and new understanding, rather than following a pre-set fixed route. Therefore, whilst rigour is vital, so too is creativity and innovation in the qualitative analysis process. The qualitative researcher, in common with the quantitative researcher, focuses on answering the research question. Qualitative research questions however, are often open and exploratory, further complicating the qualitative researcher's endeavour. The analysis of qualitative data is therefore a broad and complex task.

Each data set, as outlined in this chapter, will be discussed separately and then pulled together as one account of the conduct of the analysis. At each stage, the process followed in analysis and quality management will be outlined with supportive literature applied.

4.13.1 Philosophical Considerations in the Design of the Data Analysis Approach

There are many data analysis methods associated with the qualitative tradition, which include different underpinning methodological approaches (Miles et al. 2014). Grounded theory is closely linked to coding approaches, whereas Phenomenology is more closely linked to thematically extracting data. Narrative research is linked to presenting the participant's narrative in creative and expressive ways. It is important, when considering approaches to analysis, to consider the underpinning assumptions that are behind the choices being made (Flick 2009). However, it is of note that many qualitative researchers would describe their approach as eclectic rather than fixed on one approach (Miles et al. 2014:11). The following section, as well as describing the approaches and methods chosen for this study will also consider their underpinning assumptions.

Content analysis was chosen in this study as an overall approach to the analysis. Content analysis is a very broad approach (Edwards and Talbot 1999) and has been described in both the quantitative and qualitative paradigms. In qualitative research, content analysis tends to be derived from the naturalistic paradigm and is commonly underpinned by a belief that analysis of text can reveal the underpinning connotations and significance, and by doing so offers understanding of how participants conceive the world around them (Edwards and Talbot 1999). Qualitative content analysis is a process of looking in detail at discourse, whilst focusing on the contextual meaning in the data (Hsieh and Shannon 2005:1278). Qualitative content analysis has been described as having three separate approaches outlined below.

- Conventional - codes are derived directly from the text.
- Directed - analysis start with theory or research derived codes which are used in analysis.
- Summative - counting and comparisons of keywords linked to the context of their usage

(Hsieh and Shannon 2005).

In this study two approaches of content analysis were utilised. Initially the data was looked at utilising a conventional content analysis approach as described in 4.13.3. After this sweep of the data was undertaken, as described in 4.13 - 4.13.3, the philosophical considerations in the design of the data analysis approach were considered further. Issues of sample size were also reconsidered, which then evolved into a decision to undertake a second round of content analysis; this time utilising a directed analytical approach. Philosophical considerations and then data saturation issues underpinning this approach and decision will now be outlined as part of the overall quality framework underpinning this study (Flick 2009).

In quantitative studies, care is taken to look at the data in isolation from theory, to minimise influence and potential bias. In qualitative research, there has also been a focus on divorcing theory and findings to strengthen trustworthiness. However, this theory / analysis split has elicited different views within the qualitative approach. The critique of this approach contends that divorcing qualitative analysis from the theoretical approach taken, is treating data as unquestionable truth that needs representing (Pierre and Jackson 2014) and that the study of participant's words without the constructive critique of theory is too accepting of the objective truth of such data. This approach argues that "words can never retain their presence" (Pierre and Jackson 2014:716) once written. Due to this critique, this approach questions the assumption that only the written description of participant's experience should count as data. In this way of working the data is considered alongside or through the theoretical lens (Pierre and Jackson 2014). However, this approach is not without its critics in the field of qualitative research. Most thematic approaches take the path of only counting participants' input as data and leaving theoretical consideration until the discussion. However, to answer the research questions, the researcher determined a need for theoretical review and after much reflection and supervision, it was agreed to include theoretical analysis as a separate analytical approach, after the initial analysis had been undertaken.

4.13.2 Using Salutogenic Theory in Data Analysis

Salutogenic theory (Antonovsky1987) was integral to answering the research questions in this study. Salutogenic theory was adapted and used to formulate the diary questions. In the study, and launch it was used to describe the approach taken to health within this study. Salutogenic theory has been described as an approach which is "good to think with" (Jakobsson 2014:242) and the aim was thus, to look at the data differently, to achieve new understandings of the phenomena studied.

Whilst the initial approach to analysis in this study was focused on keeping data separate from theory, by a process of conventional content analysis (Hsieh and Shannon 2005), it was felt that more depth was needed in the analysis process. As such, after much debate around the risk of becoming theory driven (debated in 4.3, 4.4, 4.13.8) in the second phase of analysis this study moved the theoretical lens of salutogenesis into the forefront. This type of theoretical analysis in qualitative content analysis has been described as taking a directed approach (Hsieh and

Shannon 2005). In directed content analysis, the theoretical base of a study or research in that area is used to derive codes to test the data.

In this study, the three underpinning conceptual areas of the salutogenic theory - comprehensibility, manageability, and meaningfulness (Antonovsky 1987) - were used to develop new categories to further test the codes and categories, which had been established in phase one of the analysis. In this manner, the analysis was conducted without and with the theoretical lens of the salutogenic theory. The similarities and differences between these two data sets, in themselves, became areas for analytical consideration. Mixing approaches, between isolating data from theory and adapting theory to use in analysis, brings additional complexity into the analytical process and with that complexity comes the risk of a muddled or unfocused approach. In this study, qualitative content analysis was the consistent underpinning approach, with two different methods within this technique being used at different points in the analytical process. Using qualitative content analysis in this manner gave a consistency to the overall approach. In depth work was undertaken to consider the findings generated by these two different approaches to unpick their effects on the data, quality (Flick 2009) and the trustworthiness of their use. This is considered in 4.13.7, 4.13.7.4.

4.13.3 Representativeness and the Data Sets

As an overall data set although the number of participants in this study was small (see 4.10), the whole family approach, enabled by the eventual success in gaining the Children's data, achieved significant breadth and a significantly higher overall number of participants (see 4.11.4.3). With the diary data, no new themes emerged after diary five (although there was further strengthening of the existing concepts). The Children's data identified several new concepts in session one and new dimensions to existing concepts. The second Children's session, whilst not producing new Children's concepts, did strengthen the Children's perspective in the overall data set. As CCHS is a rare condition, the scope for further expansion in sample size was limited (see 1.1). The decision to undertake a two-stage analysis process was made to strengthen confidence in the data gathered, in terms of representativeness, trustworthiness, and overall quality by increasing the depth of analysis into the data gathered.

4.13.4 Development and Rationale of the Analytical Methods

Phase 1 conventional content analysis (Hsieh and Shannon 2005).

As previously described in this chapter, this study had two primary areas of data collection: Arts-based, and non-Arts-based. Each area will be discussed in terms of the process of analysis undertaken to give the reader an in-depth understanding of the quality of the approaches used.

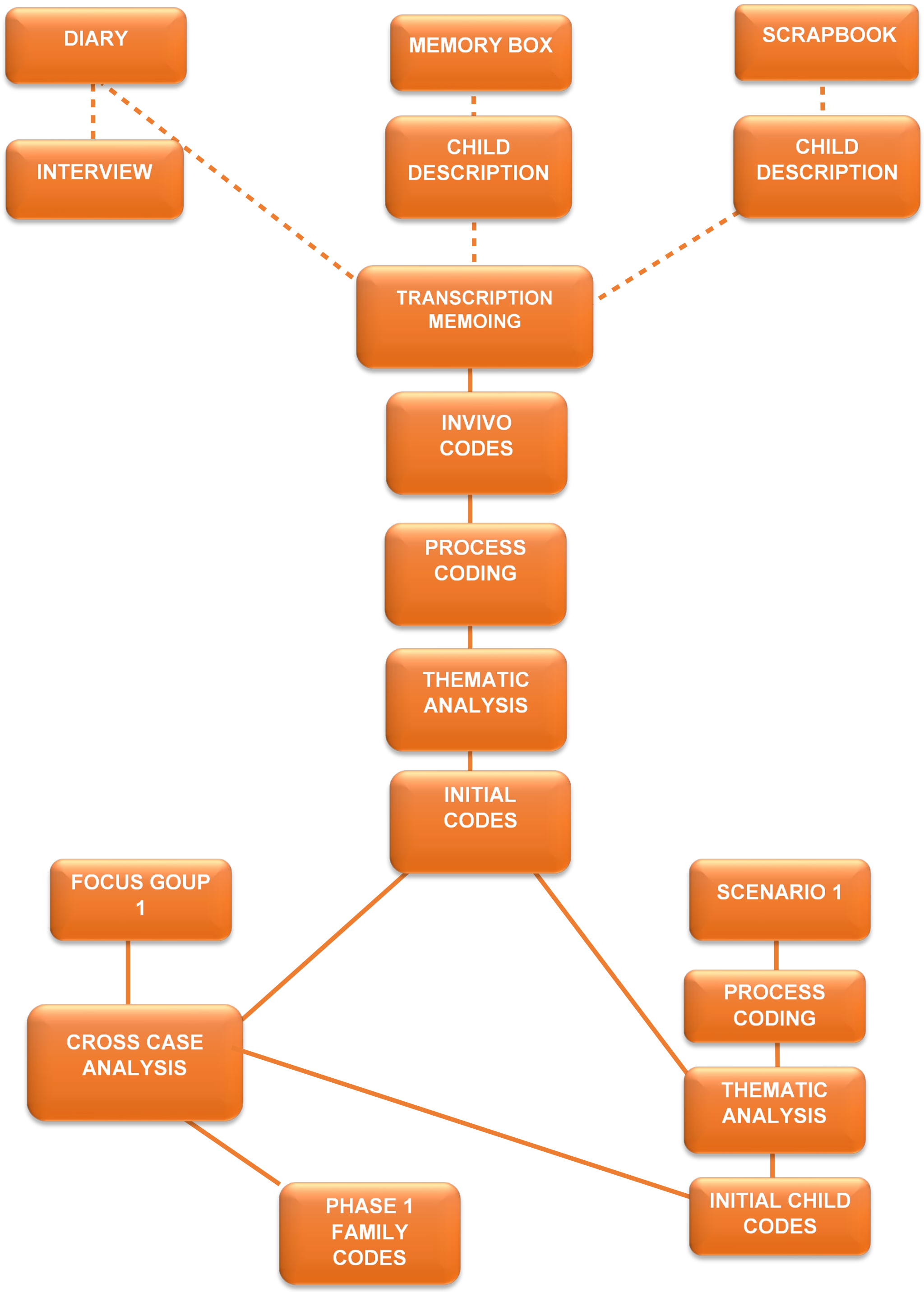


Figure 4.13 Phase One Analysis Process

4.13.4.1 Analysis Approaches for Arts-Based Data Phase One

The plan for analysis of the Arts-based data in phase one was to take the data contributed in diaries, scrapbooks, and memento boxes whether drawn, or collected and to take photographs of the pictures or items and display them alongside the participant's descriptions of the item or picture, which was to be then recorded and transcribed. Each picture or item would then have had the participant's description attached to it. By this process it was envisaged that analysis would then have been based on the participant's description, which would have been considered alongside the picture of the chosen object. However, as described earlier in this chapter, a lack of data was collected in the scrap books and memento boxes (only two pages of scrap book entries were available for analysis and nothing was collected in the memento boxes). It was therefore decided to not analyse these two pages and to rationalise the analysis to reflect the text-based data that had been collected. The lack of Children's data necessitated new methods of data collection to achieve the study's objective of gaining a family data set.

As a reminder, at the 2014 CCHSUKSG away day, a new Arts-based Children's data collection method was launched in an attempt to capture the Child's voice. The revised Arts-based method was that of interactive play-based scenarios based on the vignette technique described further in 4.8.3. These scenarios were used to attempt to engage Children through an Arts-based methodology described in 4.8. The two play-based scenarios that were created to enact the research intention of achieving a Child, young person and family Arts-based data set, took different approaches than had been originally envisaged. Therefore, the method of play based participatory scenarios required adapted analytical approaches. The suitcase method described in 4.11.4.3 was common to both scenarios and will be considered first.

The Children's suitcases were designed as the first activity in scenario one and two. The picture below is an example from scenario one.

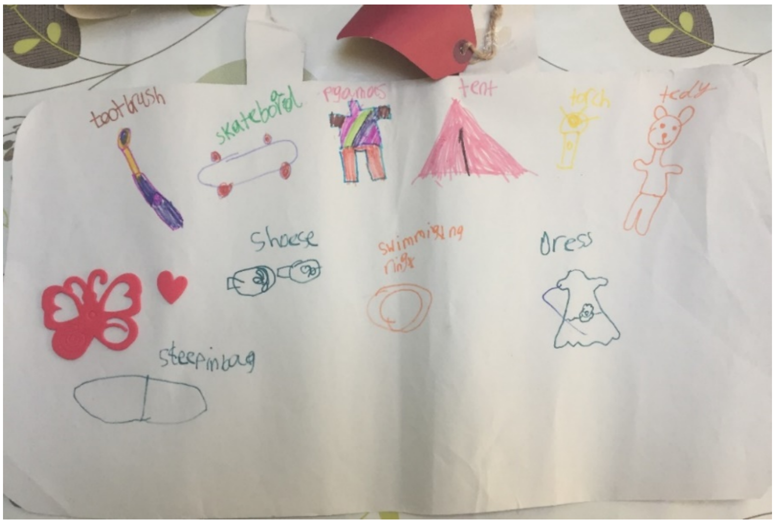


Figure 4.14 A Child's Suitcase from Scenario One (reproduced with permission)

The Children's drawings were not interpreted, but the Children's descriptions of what was in their suitcases were used for analysis. This approach was chosen, as the Children's interpretation of their suitcase contents was of primary importance, rather than a researcher led interpretation. Coad (2007:495) suggests “there is a need to ensure there is mutual comprehension between the adult researcher and the Child’s meaning in the artwork produced”. Therefore, the Children’s descriptions were collected for analysis and were placed with the primary data for analysis. Scenario one also had specific data collection points built in. After the scenario, free play with the scenario props allowed further observations to be made. At each of these data points research assistants were trained to take detailed notes of the Children’s words and actions. The notes collected from the creation of the suitcases and the observations recorded by the observers and researcher of the Children’s interactions during the scenario were recorded, coded and grouped into categories. The Children’s data was then considered against the diary, interview and focus group data derived categories.

4.13.4.2 Analysis Approaches for Non-Arts-Based Data Phase One

The primary approach taken in the analysis of the data was that of coding. The qualitative content analysis approach is often associated with the technique of coding. Coding is a process of applying different types of codes to sections of transcript or other data sources. Coding in itself is a key component of the analysis process, as the activity of coding requires in depth consideration and analysis of the data (Miles et al. 2014:72). Coding as an approach implies a belief that social singularities exist in the mind and experience of participants (Miles et al. 2014). However, this approach is not from the positivist deductive approach, as it accepts that human actions are loaded with meaning. Coding is used to identify a framework that underpins human interactions. As an approach, coding allows clarity and openness about the research process. By taking this clear and formalised approach, coding aims to deliver findings that are “credible, dependable and replicable but in “qualitative terms” (Miles et al. 2014:5). As such it forms a key part of the quality process (Flick 2009). The approach taken in the creation of codes in phase one of this study was based on the conventional content analysis approach (Hsieh and Shannon 2005).

However, coding as an approach is not without its critics. In the Interpretivist ontology coding has been criticised for following a “quasi-statistical analytic practice” (Pierre and Jackson 2014:715). This critique contends that participant’s words are treated as data waiting to be labelled and even counted. By taking this approach, it has been argued qualitative research is betraying its core beliefs (Pierre and Jackson 2014).

Coding, despite these critiques, had become widely used in qualitative research, as a method of applying rigor to analysis (Miles et al. 2014). This approach was used in this study as a key step in this analysis process. By maintaining a continual link to the overview of the text, the researcher aimed to reduce the risk of coding becoming the dominant approach. Coding, as an approach is also underpinned by many different methods. These methods were carefully considered as a part of the quality review, and a number of techniques were chosen for this study. Each of the chosen techniques is now critically discussed in detail.

Memos are a coding technique, which are defined as “the arising write-up of ideas about events and their relationships, as they strike the analyst while coding” (Glaser 1978:72). Memos go beyond reporting data. They are conceptual and tie data together into recognisable clusters. Memos can allude to interpretation of meaning, contain researcher’s reflections, include feelings, methodological reflections, possible further research avenues, cross reference or contain elaboration from research notes (Miles et al. 2014).

In vivo codes were used to keep a focus on the participant’s own words. In this approach, each code is named with participant’s own descriptions (Miles et al. 2014:74). These descriptions are often linked with the language of the group or culture being studied. Therefore, in the field of health research, in vivo codes may use professional language or context specific descriptions. In vivo codes, by using participant’s own words as codes, remain close to the participant’s words. The intent of this approach is that by using the participant’s own words, the poignancy of their descriptions is not lost in the analytical process.

The technique of process coding was utilised to deepen the analysis. In the process coding approach codes are looked at to see what the action is in the words. In this approach to coding all codes finish with verbs to capture an insight into what is happening. This technique is designed to move beyond description to get to the process and implication behind the words being analysed.

The non-coding method of cross case analysis was used to reduce the number of codes, by considering the processes and outcomes across many cases, to understand how they were qualified by local conditions and thus develop more sophisticated descriptions and more powerful explanations (Miles and Huberman 1994:172). In this study, this method was used in the process of data reduction to reduce codes into fewer more powerful explanatory codes. This took place after the initial coding, as the Children’s codes were integrated into the data set and then again after directed content analysis.

Member checking was a key approach used during analysis to maintain credibility, trustworthiness and quality assurance in this study (Flick 2009). The approach of member checking is utilised to check that participants believe that the data produced after analysis is credible and matches what they believe they have contributed to the study. When undertaken at different stages, member checking is a technique to ensure underpinning meanings that remain identifiable to participants are retained through analysis (Carlson 2010). Some researchers advocate that member checking is a continual process (Carlson 2010) however, the time pressures that parents looking after a Child with complex medical needs are subject to, was considered an ethical barrier to a continual member checking approach in this study.

Therefore, a process of periodic member checking was considered more manageable for the participants and was therefore utilised within this study.

4.13.5 Critique of the Conduct of Conventional Content Analysis Phase One

4.13.5.1 Critique of the Conduct of Phase One Non-Arts-Based Analysis

The diaries were successful in capturing parental view and some young people's views presented in text form, but due to the unsuccessful nature of the initial Arts-based methods the diaries by default became a non-Arts-based tool. The Children's data collection was redesigned and was collected after the initial data collection, therefore in discussing the conduct of the analysis component of the research it is pertinent to consider interview data first. The interviews enabled an in-depth questioning of the participants and in some interviews, Children joined in around the data they felt was most important to them.

The researcher became familiar with and immersed in the data. This immersion started with consideration of the participant diaries, interviews and summary written after each data collection activity. The immersion then continued with listening to the recordings of the interviews, and re-reading the diaries in their original format. All the diaries and interviews were transcribed. The text was participant and line coded to make subsequent links with the full text clear and to identify what each of the participants contributed to each category or theme. The analysis then progressed onto coding. Coding in itself is a key component of the analysis process, as the activity of coding requires in depth consideration and analysis of the data (Miles et al. 2014:72). First wave coding was undertaken through a memo approach. Each memo was dated, entitled with the key concept noted and when moved was linked to the line numbers of the section of the transcript. Each transcript was worked through, whilst listening to the recording (in the case of the interviews) to ensure over or under emphasis was not made to the participant's statements and to maintain a clear data derived approach to the analysis. Each memo was recorded alongside the text. The use of memoing in this study had the advantage of capturing understanding gained, as the data was read in context soon after data capture. In a study, conducted part time over a seven-year period, this approach was particularly valuable in maintaining this knowledge across a protracted analytical process. There was also a wider benefit of identifying possible links from the whole text analysis, which also aided in formulation of concepts later in the process.

In vivo codes were then used to keep true to participant's words and honour their voice in the research (Miles et al. 2014). In this research, the context was the family home and the wider group culture (which was that of the CCHSUKSG.) An advantage of the in vivo coding approach is that during coding the researcher is drawn to focus on words used by the families themselves. An example was the "random carer" code, described in 5.2.4 which was a directly quoted name given to carers, who arrive to cover shifts, who have been previously unknown to the Child or family. This in vivo code was a commonly used phrase by a small group of respondents. This code identified the trust required to handover care of a participant's Child to a carer. The participants, through this code, also described the fear that they believed was caused to their Children by this group of "random" carers.

The text was then considered again, and process coded. This simple, but effective technique focused the researcher on what was going on in the data. An example of this was the risk-taking code, which identified an important difference between family and professional perspectives, by going beyond the description and into the action it triggered or motivated. These three separate sweeps of the data worked well with different nuances of the data being captured by each different coding technique.

The coded text was then considered as whole in depth (codes had been applied to the whole data set in context), whilst taking reference to the codes from the coding process and the overview of the data the initial categories were formulated. The data was then cut and pasted into these categories along with any codes assigned to each segment. Where an aspect of text fitted in more than one category, it was put in all the categories it was linked to and its interconnections to other categories were noted. The categories were then summarised. Categories that were given, where practical, in vivo names. The strength of this approach was that it maintained the participant's story through the analysis process, but also had the rigor and in-depth questioning created by three sweeps of coding. An important part of the Quality approach of this research was to keep reviewing the data together, whilst also achieving the rigor and close questioning of coding. At the same time the researcher aimed to do this without running the risk of exclusively focusing simply on the codes which in turn then potentially dominates the analysis and thus loses the wider context (Miles, Huberman and Saldana 2014).

In considering the representativeness of the data collected, there were a number of processes adopted in the analytical processes to strengthen the representativeness of the data. When formulating codes and concepts it is possible to become over focused on the highly articulate few, who with clarity present and express their argument rather than unravelling the descriptions of less articulate but potentially insightful individuals (Miles et al. 2014). Each code was therefore considered for balance, by looking at which participants had contributed to each section of content that went on to underpin codes and concepts. If there appeared to be a 'domination' by a few participants, the researcher re-considered the data set and ensured that alternative data was sought from the wider data set. One key manual technique used to strengthen the overall understanding of the data was cross case analysis. This approach was utilised to look across the cases to increase generalisability and reduce the codes and strengthen the content of the codes. The researcher undertook cross case analysis by a process the researcher called table scanning. This approach involved each category being printed out and put on a three-metre table. The researcher then scanned across the table by walking up and down its length noting interconnections across categories. The categories were then reconsidered in the same manner, but focusing on the underpinning data. This resulted in a questioning, connecting, and rationalising of the categories.

Overall memoing proved, in the researcher's view, a good technique for gaining first impressions of the text. However, the depth of analysis came from In vivo codes and process codes. These two techniques were (in the researcher's view) very effective analysis techniques that worked well together. In vivo codes maintained a participant focus, whilst process coding developed a clear interpretive approach to the analysis by continually asking of the data "what is happening here". The approach of table scanning was used at each point of transition and reduction in the data. In this manner, the eventual codes had undergone coding, reduction and review, whilst remaining true to the data set as a whole.

A process of in-depth consideration of each of these preliminary categories was then undertaken with a member of the supervision team. These initial categories and codes were then reviewed and grouped, preliminary thematic titles were assigned to the initial categories. The use of a second researcher at this point allowed questioning of the researcher's analytical approach and initial conclusions. The researcher also wanted to gain an initial view of representativeness from a non-participant who had experience of LTV. The themes complete with their underpinning categories and codes were taken to the parent participant, who undertook a review of the themes, categories, and codes. This analysis was insightful and gave the researcher areas to recheck review and reconsider. However, caution was required as the parent, who undertook this role, although having a ventilated Child of their own, had a Child with a significantly more disabling condition. Therefore, some of the reflections around the codes could have been influenced by a different experience of disability.

To consider the findings and to provide further assurance of quality, member checking was conducted through a focus group to check the initial themes. The decision was taken to share only the categories and themes, rather than the full data sets, to reduce the volume of data that participants were asked to consider. This is an approach advocated in Carlson (2010). Each theme and concept were considered, and the focus group was recorded, and researcher notes taken. Collection of data was undertaken by audio taping. Each tape was listened to twice. The first-time broad issues and context were noted. On the second listening of the tape, each of these broad issues was contextualised and documented in detail noting agreement, issues and further context around each concept and theme. These descriptions were then considered against the research notes recorded at the time. The themes and concepts were then reviewed and adapted after careful consideration and reflection on this data. This process also allowed data saturation to be tested by seeing if a group discussion raised any new codes, categories or potential themes. The following section will consider this aspect further.

4.13.5.2 Critique of the Conduct of Arts-Based Methods Phase One

The initial intention of the first phase data collection to create an art-based approach which captured the whole family's views on living with CCHS was, as discussed, not successful. The information produced by the families was all text apart from two scrap book entries. The initial Arts-based methods and analysis approaches were therefore abandoned, and a new approach of participatory play-based scenarios was developed described in 4.8.3. This new approach to data collection also required a modified analytical process. As a reminder, the method which effectively elicited the views of Children was that of participatory play-based 4.8.3.3.

The data analysis process started with immersion. This included typing the data up with the contents of each Child's suitcase listed with the Child's description. These descriptions were then considered again in reference to the Child's suitcase drawing. The data collected and each data point in the scenario was also transcribed into a written document coded to the Child and the data collection event. All the data was then coded using memos, in vivo codes and finally process coded (as described above in the diary and interview data collection). All of these codes were applied to the whole text to maintain the context up to the final steps of the analysis. The data was put into initial codes with data pertaining to a number of codes being cross referenced to all. The initial ten Children's codes were reduced using a process of inspection. Inspection was carried out by scanning the typed data, situated within the initial codes looking for a crossover or repetition. This was undertaken as in the diary and interview data analysis by table scanning (described 4.13.5.1). The final Children's codes were considered against the adult diary and interview codes in another process of reduction. This reduction was carried out again by table scanning of both the Children's and adult's codes. Each code was then reconsidered in-depth going back to the data to look for alternative or deeper meanings as an important quality check (Flick 2009, Miles et al. 2014).

The Children's codes of 'I brought my mummy' and 'living in the moment' had unique characteristics to the Children's data and were added as new codes. An example of this was in the, 'I brought mummy' code. There was a reference of the Child's perceptions that their CCHS was owned by their parent. Whilst taken on face value, this could raise safety concerns for the Children, as they were not accepting any ownership of their condition; however, on in-depth analysis of the underpinning data the reality appeared to be that for this group of young Children, this transfer of responsibility initially liberated the Children from the responsibilities of the condition to experience being a Child unencumbered by LTV, at a time in their lives where close adult supervision is normally available.

The other Children's codes were merged to form family codes (like any Child, taking the next big leap, siblings, family support, friend support self-care and school) and the categories were refined and re-described to take account of the Children's data. There was a danger to quality within this process of the Child's voice being lost. Therefore, once merged, the data was reviewed again to ensure that the Children's voice had retained its presence in the data (which had a significantly larger adult data set); codes were reconsidered and where necessary the Children's data was strengthened where the codes were considered to be dominated by adult perspectives.

4.13.6 Conclusion of Phase One Analysis

The phase one data collection met its aim of establishing a family data base (after adaption of the Arts-based approach). The analysis of diaries, interviews and the participatory play-based scenarios enabled a number of different approaches to collect the family data. This triangulating of the data created a good quality control technique for comparing and contrasting data. The Arts-based methods utilised in this study, also created an opportunity for creating a much wider dataset, thereby dissipating the risks of coding by having a mixed methods approach. The initial failure of the Children's data collection, whilst resulting in a novel and productive methodology of Children's play-based scenarios, meant that the Children's data was analysed after the adult data and initial findings. The initial inclusion of the Children's data into the findings didn't fully reflect the Children's codes, leading to a significant rethink of the quality achieved in process of moving from adult and Children's data sets into an integrated family data set. The issue of power differentials between Child participants, adult participants and researchers are well documented (Robinson and Kellett 2004) questioning this approach. To address this concern, a significant review of the analysis was undertaken, to strengthen the Children's voice in the findings and an additional play-based scenario was undertaken in phase two of the study. The research questions had required a family-based data set to answer the questions and safeguards were enacted, but some reservation remained with this approach.

The different coding approaches worked well together, challenging initial conclusions and getting behind the data. The critique that coding data is a betrayal of the qualitative approach (St. Pierre and Jackson 2014) was to some degree offset by applying codes, whilst maintaining an ongoing link with the whole data set. On review of the quality framework (Flick 2009), the use of coding was seen by the researcher as a significant strength in the analysis process. The slight concern, that had led to a reminder to research assistants not to prompt the Children during the creation of the suitcases for scenario one, left some doubts about this aspect of the phase one Arts-based data set.

4.13.7 Development and Rationale - Analytical Approach Phase Two Directed Analysis (Hsieh and Shannon 2005)

The qualitative researcher in setting out to discover new patterns and relationships (Kelle 2005) must make fundamental decisions about knowledge. The quantitative hypothetical deductive approach based upon a hypothesis was ruled out in this study, based upon the rationale set out in section 4.2. However, the link between theory and data analysis remains an area of significant debate in qualitative research. The risks of theory dominating, and distorting analysis was behind the creation of grounded theory (Kelle 2005). In any qualitative analysis process the researcher's preconceptions are impossible to completely remove and therefore must be externalised and reflected upon in the research process. Salutogenic theory (Antonovsky 1987) was adapted to be utilised at every stage of the data collection from the launch to the diary questions. Grounded theory offers the caution of data becoming subsumed by the great theoretical approaches. However, theory also can provide a lens for looking differently and often deeper at the data and challenging conventional analysis. MacLure (2008:174) describes a process of analysis that accepts all these approaches, as ways of looking at data in different ways and from different perspectives to make analysis more rigorous.

The researcher, through a process of reflection on the findings of the literature review and the gaps identified in the current literature base, after a quality review process, concluded that a new approach was required. Directed content analysis was decided upon to undertake a theoretical inspection of the data. This type of theoretical analysis in qualitative content analysis has been described as taking a directed approach (Hsieh and Shannon 2005). In directed content analysis, the theoretical base of a study or research in that area is used to derive codes to test the data. In this study, the three underpinning conceptual areas of the salutogenic theory comprehensibility, manageability, and meaningfulness (Antonovsky 1987) were adapted to develop new categories to further test the codes, which had been established in phase one of the analysis. In this manner, the analysis was conducted without and with the theoretical lens of the salutogenic theory. The similarities and differences between these two data sets, then in themselves, became areas for analytical consideration. Mixing approaches between isolating data from theory and using theory in analysis brings additional complexity into the analytical and quality assurance process and with that complexity comes the risk of a muddled or unfocused approach. In this study qualitative content analysis was used to be the consistent underpinning approach, with two different methods within this technique being used at different points in the analytical process. It was envisaged that using qualitative content analysis in this manner would give a consistency to the overall approach. In depth quality review was also planned to consider the findings generated by these two different approaches to unpick their effects on the data and the trustworthiness of their use. This is considered in 4.16.6.2 and 4.16.6.4.

The approach of directed content analysis starts with theory or research derived codes which are used in analysis. A method was required to apply a theoretical approach to the data "inspection" (Denzin 1978:74) and directed content analysis was chosen. Inspection is the process of taking an idea that appears to have merit and looking at it through the literature and in different contexts to test the idea. In this study, the approach taken was to adapt this approach and use Antonovsky's (1987) three concepts of comprehensibility, manageability, and meaningfulness as theoretical areas to reconsider the data. This process included an in-depth consideration of the codes collected in phase one and a regrouping of these codes into Antonovsky's three conceptual areas.

In summary, when considering the approaches developed to undertake qualitative analysis, complexity in itself can create risks to the quality and trustworthiness of a data set. Diverse forms of analysis were integral to the overall quality approach that was taken in this study. Data was mixed in terms of approaches and methods to test the data by different collection and analysis techniques. In this manner, the researcher endeavoured to seek depth rather than quantity in the data collected and used a mixture of methods to deepen the analysis. However, due to the theoretical nature of the research questions, the initial approaches to analysis needed to be expanded to include directed content analysis. An in-depth critique and quality review of the effectiveness of this decision will be offered after the conduct of the analysis has been outlined. As this study used a mix of Arts-based methods the process of data analysis varied according to the method that had been used to collect the data for each part of the data set. For clarity, the methods were grouped into phase one conventional content analysis and phase two directed content analysis.

4.13.7.1 Conduct of Directed Content Analysis Phase Two

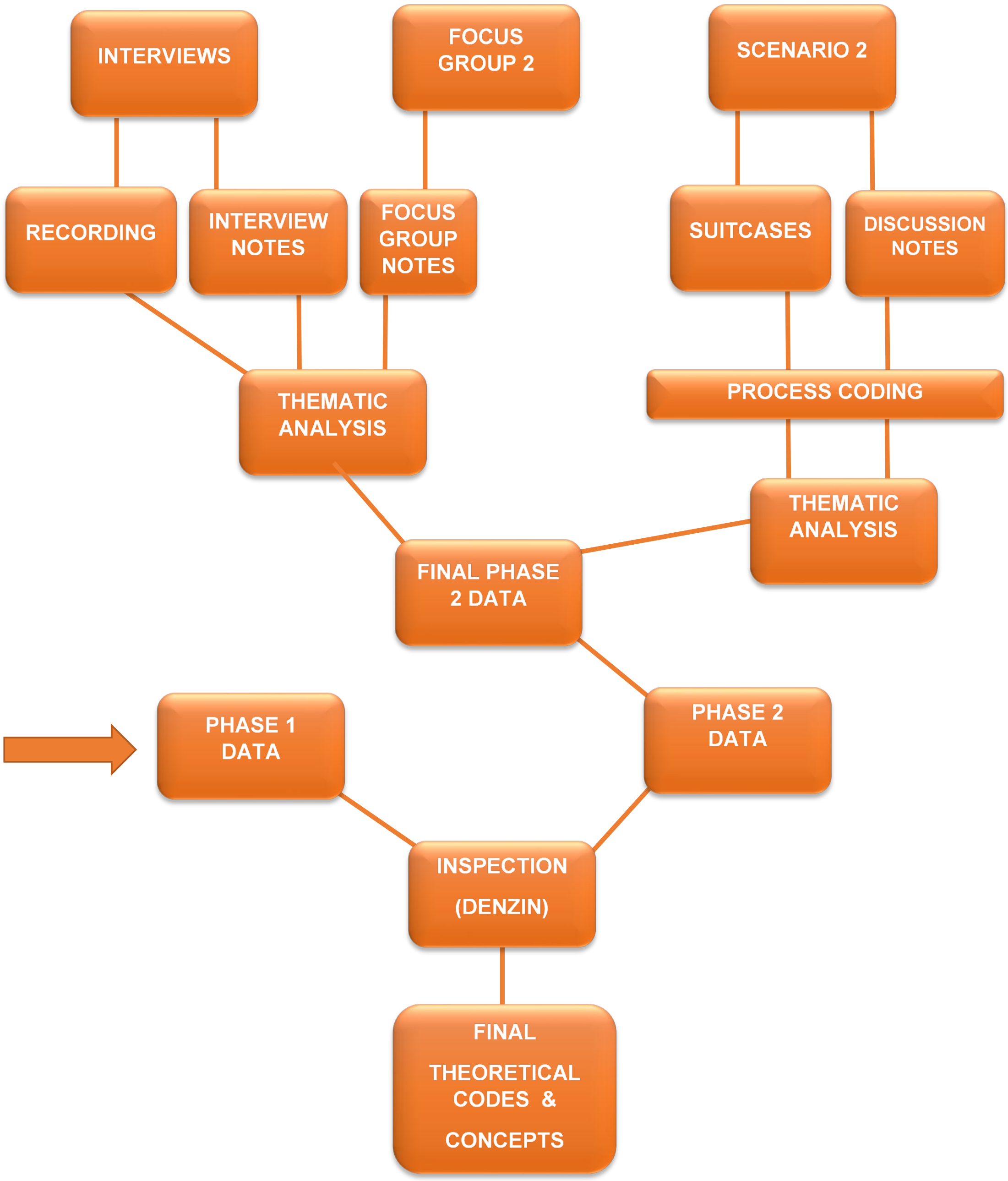


Figure 4.15 Phase Two Analysis Process

To undertake directed content analysis required a method for reviewing all the data through the theoretical lens of the salutogenic theory (Antonovsky 1987). To enact this approach the phase one codes were considered through adapted use of each of Antonovsky's (1987) three conceptual areas to add depth to the analysis by looking theoretically at the text.

Codes and categories were developed through a process of directed analysis from the data. The final analytical process was to compare the new theoretical codes and concepts with the phase one codes. From the 29 original categories, 18 new categories were created in the three Conceptual areas of comprehensibility, manageability, and meaningfulness (Antonovsky 1987).

The relationships between the original and final categories is shown in the table below. The transition of the categories resulted in a reduction in the number of categories from 29 to under 20. From the figure 4.16 below, it can be seen that the transition was a complex process.

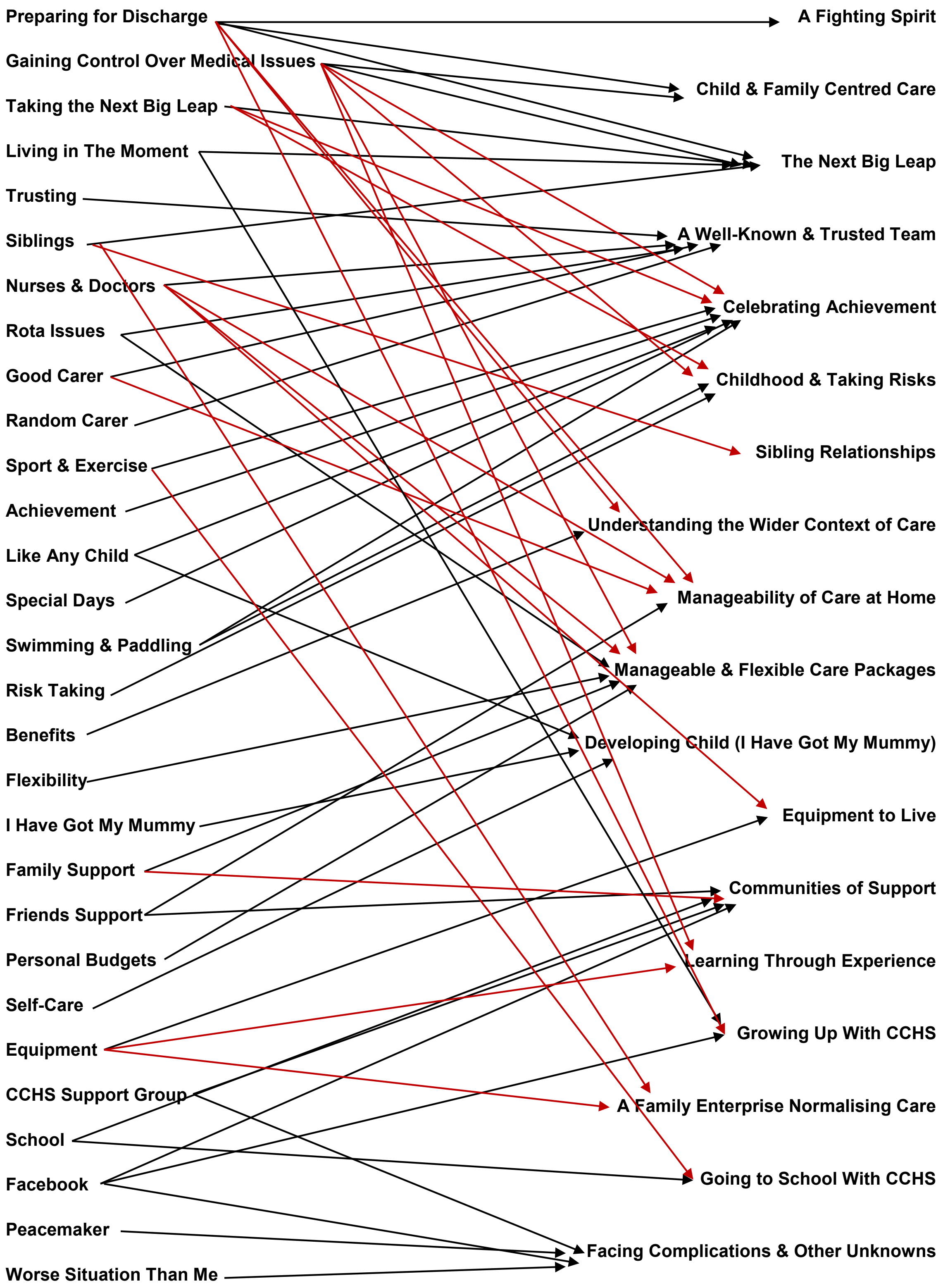

Preparing for Discharge
Gaining Control Over Medical Issues
Taking the Next Big Leap
Living in The Moment
Trusting
Siblings
Nurses & Doctors
Rota Issues
Good Carer
Random Carer
Sport & Exercise
Achievement
Like Any Child
Special Days
Swimming & Paddling
Risk Taking
Benefits
Flexibility
I Have Got My Mummy
Family Support
Friends Support
Personal Budgets
Self-Care
Equipment
CCHS Support Group
School
Facebook
Peacemaker
Worse Situation Than Me
A Fighting Spirit
Child & Family Centred Care
The Next Big Leap
A Well-Known & Trusted Team
Celebrating Achievement
Childhood & Taking Risks
Sibling Relationships
Understanding the Wider Context of Care
Manageability of Care at Home
Manageable & Flexible Care Packages
Developing Child (I Have Got My Mummy)
Equipment to Live
Communities of Support
Learning Through Experience
Growing Up With CCHS
A Family Enterprise Normalising Care
Going to School With CCHS
Facing Complications & Other Unknowns

Figure 4.16 The Relationships Between the Original and Final Categories

An example of this change was the removal of the ‘preparing for discharge’ category. It can be seen from the figure 4.16 that preparing for discharge disappeared as a category, but became linked to six of the new categories. However, the total number of categories reduced as the interconnections became clear once new more potent and powerful categories emerged. This change was not to reduce the impact of the defining time of the pre-discharge period of time, but to more truly reflect the breath of impact the participants’ experiences of the time in hospital had across their familys’ lives, by moving from a historical time period focus “preparing for discharge” to looking how the event impacted in an ongoing manner across the day to day lives of the participants.

When considered against the original data set, the new categories were felt by the researcher to reflect the families’ voices in a more trustworthy way, while arranging it in a way that enabled theoretical analysis to be undertaken, to further deepen the understanding of the data. This process enabled the cross-data links to be considered, which were not apparent in the coding approaches that had been used to that point. The comparison of these two approaches led to another in-depth review of the data with original diary, interview and Arts base data being relooked at as whole.

4.13.7.2 Critique of the Conduct of Non-Arts-Based Methods Phase Two

The researcher considered this to be a significant point of change that required more than open written analysis and discussion to assure the quality criteria was achieved. After formulating the codes and categories the overall themes were developed. The categories and themes were taken back to the group to member check both the new categories and the new overall themes.

The process of member checking was undertaken as part of the quality review (Flick 2009) through the medium of a focus group. The event was undertaken at the 2015 CCHSUKSG weekend due to the importance of this transitional stage to the data. The researcher, through supervision, concluded that the four families who were not at the focus group would be interviewed at home.

The focus group was presented with the findings in each of Antonovsky’s (1987) three conceptual areas. Each code in each conceptual area was discussed in turn. Finally, an overall vision identified through the family’s answers to the appreciative enquiry (Cooperrider et al. 2009) question was presented (see 4.11.3.1). The families were asked to comment on each area and finally on the researcher’s summary of the future vision. The focus group was recorded, and the researcher wrote detailed field notes after the event.

The tape was listened to twice with careful notes being taken on each review. The notes were cross referenced with the field notes and final summaries were produced for each conceptual area and the overall vision. The four families who were seen at home were presented with the same information and were asked to comment at the same points as the focus group. The interviews were recorded. The tapes were listened to twice and considered alongside the research notes. The families at the focus group and during the home interviews recognised the data and as well as adding more examples, felt the new categories and codes were representative of their experience. In addition, they considered the new concepts offered a deeper level of insight, rather than the original in vivo derived description, which they concluded were accurate and interesting. One of the original in vivo names was referred to more than once in the discussion, so was utilised in the final naming to recognise the descriptive value it represented (The next big leap). The focus group while adding debate, consensus and additional context, were not as detailed as the subsequent home interviews. However, the four interviews reached the same overall conclusions, but had many more examples and explored the data in more depth. The focus group and interview final summaries were pulled together and considered against the concepts as established through the review. The outcomes of this process of member checking were then integrated into the findings.

4.13.7.3 Critique of the Conduct of Arts-Based Data - Phase Two

A focus group to consider the findings was not practical to quality review the Children's group due to the age range of participants. The decision was therefore made to strengthen the Children's data with a second participatory play-based scenario. This strengthened data set would then be used to critique the outcomes of the directed content analysis's conduct of this scenario is outlined in section 4.13.5.1.

The suitcase tool was common to both scenario one and two. In scenario one the research assistants conducted this activity. In scenario two the researcher undertook this activity, as there had been some concern that the assistants may have not had sufficient instruction on not prompting the Children and could have influenced them. Therefore, a direct researcher to Child approach was considered more trustworthy and was used in scenario two in the suitcase exercise. The second participatory play-based scenario took a different approach, with the scenario used to engage the Children in a final interactive discussion described in 4.11.4.3. This discussion then was used as the primary data collection point. Free play was then encouraged with the research props. The researcher took field notes and then wrote up notes after the scenario. The increased depth of the data obtained from this second event suggested that the primary event had not achieved data saturation.

The researcher became immersed in the notes collected from the creation of the suitcases and the observations recorded by the observers and researcher of the Children's interactions during the scenario. This continued on into transcription of the data. Coding using memos, in vivo and process codes were carried out as in the first wave analysis. The data was then tested against the original Children's data and the new thematic codes (created by directed content analysis). Scenario two didn't provide evidence to change the original Children's codes created, but did strengthen them significantly. This data was used to challenge and develop the new concepts and codes created by the thematic review from a Children's perspective, leading to a strengthened Child voice in the new concepts and codes.

4.13.7.4 Critique of the Phase Two Analytical Process

The process of qualitative research accepts the researcher influence on analysis and addresses this effect by open analysis and reflection (Hughes n.d). In this study, this risk was addressed by extensive discussion around this issue in supervision. On review self-analysis was also apparent throughout this period in the research diary. However, the researcher considered a member check essential to check the quality and trustworthiness of this transition. The second focus group, interviews and second Children's scenario supported the trustworthiness of the analytical process, which had been enacted by moving from conventional content analysis on to directed analysis conducted through Antonovsky's (1987) three conceptual areas. Focus groups, interviews and participant play-based scenarios are a key quality check (Flick 2009). These activities are also an affirmation of the importance of participants, as was traveling many miles to interviewee's homes, but this affirmation in itself may influence participants to be positive around the researcher's endeavours. By using a mix of methods, the potential pressure to affirm which could have arisen through the group culture of the focus group was, in the researchers view, largely offset by some interviewing at home where parents were in control and relaxed and comfortable.

4.13.8 Conclusion of Analysis Process

Analysis is the keystone of a research study. Without strength in this key area the trustworthiness and the quality of the whole study could be put in doubt. In this study, there were some significant challenges around analysis, both around the creation and merging of the Children's and adult's data to produce a family data set and the use of theory during the analysis process.

A close link with theory was important to answer the research questions. However, this underpinning theoretical base was a potential strength and weakness in analysis. The use of a two-phase analysis process was developed as part of the quality management process (Flick 2009) to reduce this risk, by analysing the data with and without theory. The use of a modified qualitative content analysis approach enabled the two-phase analytical approach decided upon to undertake the theoretical review to be enacted. This then enabled the clear context of an established analytical framework, reducing the possibility of a confused and unstructured approach. This study's approach was to split the phase one process of theory free data collection from the second phase, where salutogenic theory (Antonovsky 1979, 1987) was used adapted to use as a lens to consider the data. This enabled the data to be considered through a clearly identified theoretical viewpoint, which was then considered against codes, which had been gained without such theoretical influence in analysis. However, both approaches had questions originating from a theoretical model. In the researcher's view, this enabled increased trustworthiness and importantly a way of analysis informing the evidence around question one, but openness about possible theoretical influence is required.

Overall the researcher concluded that theoretical analysis was essential to answer the research questions. By looking at the data, with and without a theoretical lens, the difference between the two methods are apparent. By challenging this transition through reflection, supervision, a focus group, interviews, and a further participatory play-based scenario, a robust quality framework (Flick 2009) was used and clear process of challenge, was conducted.

The mix of approaches and methods led to a significant extension of the data collection process. The main changes enacted were the creation of the play based participatory scenarios. The Child focused approach used in this technique created a strong Children's voice. As described, the process of analysis was carefully developed to ensure that the Child's voice was captured and fully represented in the family data. This process has been carefully outlined to allow consideration of the researchers' approach. By being open and critical about this process and debate, the researcher hopes to have enabled the reader to be able to consider those decisions and the process of analysis undertaken in this study.

4.14 Conclusion of Methodology and Methods

This chapter provides an in depth rational and critique of the methodological approaches taken in this study. The chapter is extensive and detailed to present an open reflection of the quality measures enacted the Researcher has included all methods used including those that proved unsuccessful. Traditional and novel approaches have been utilised throughout to gain new and novel insights into the research questions. As the study progressed, the depth of methods and analysis required to answer the questions became clear and led to extensive additional study. The findings identified by these approaches will be outlined in the following chapter.

Chapter 5. Findings

5.1. Introduction

In the previous chapter the analytical methodology, analysis and quality framework was critically explored with key points related to the data analysis process considered. From this process, a decision was made to undertake two sweeps of content analysis. The first sweep was undertaken utilising a conventional content analysis approach and the second utilising a directed approach. The directed approach was based on Antonovsky's (1987) three constructs of comprehensibility, manageability and meaningfulness. This approach has been continued into the findings, with the findings grouped in each of Antonovsky's (1987) three conceptual areas. This approach came out of the data, as certain events had a significant and ongoing effect on their whole family life. However, when this occurs, the data is considered from a different conceptual angle. It is therefore hoped that this approach, rather than resulting in repetition, achieves a clear and interpretive focus on the data gathered from this group of highly complex Children and their families.

The findings are presented below within Antonovsky's (1987) three conceptual areas in the order of meaningfulness, comprehensibility and manageability which represented the priority that became apparent from the findings (discussed in detail in 6.5.1). Key codes are used in each category to explore the in-depth findings. Underpinning codes (including direct quotes from participants) are demonstrated to enable the reader to have a clear link between the data and findings presented. To protect the children's identities their names have been replaced with the letter N to represent their first name.

5.2. Meaningfulness

As an aide-mémoire, meaningfulness is the motivational component of the framework, which is the extent to which a person has areas of their life that are important to them, that they very much care about and that make sense to them. This collection of codes considers this construct for the families within this study.

5.2.1. A Fighting Spirit

The initial time on a Paediatric Intensive Care Unit (PICU) prior to actual diagnosis or early post diagnosis appeared to be a time of extreme instability for families. These were described as terrifying times where on some occasions the Child had life threatening complications. For some families, insensitive communication featured in the families' accounts exemplified in this quotation:

“and he says (DR) then all seems to be okay, there seems to be a bit of a spark about him, he seems viable, my Child was two” (2 470-471)

In the following passage one parent identifies how the medical focus on a diagnosis or problems affected how hospital staff described medical issues. The parent then describes how on reflection, she would have liked to hear the message in a way that would have helped her re-establish hope for the future*:*

“I had to focus on what was positive in the fact that he didn’t have extra complications, he might not have, there’s all these percentages, there’s 40% that have developmental delays, you know, 30% of them have other problems of Hirschsprung’s and things like that, but I wanted to hear 60% of them don’t have this” (11 284-288).

What was interesting, was that it was these early events and communication exchanges that appeared to form the basis of future relationships. If communication exchanges were positive experiences, trust appeared to be built. Conversely, the opposite was true, where early communication was perceived as poor, negative relationships with professional staff were developed; trust issues also became a significant feature. One implication of this was that families felt they had to be fight for everything for their Child as described below:

“You fight so hard to get your Child home from hospital…. it’s all you think about” (2 524-525).

This perceived fight helped families to cope and in effect became a personal mission. However, facing each challenge *as a battle* had consequences, in that in some cases this resulted in families finding it hard to trust professionals planning the long-term homecare packages. This had profound effects which commonly became an issue, either within days of discharge or over the following months:

“I did bring him out early and umm. and I had to do three nights a week myself for three weeks and it nearly brought me to my knees and um I just think I was just so impatient umm and I recognise in hindsight I shouldn’t have done that but I did it and I knew it wasn’t for ever but it was hard I think because there was three nights together every week for three weeks was hard, umm but yeah I think you quickly realise you can’t do that on your own” (11 143-147).

5.2.2. Child and Family Centred Care

The impact of hospitalisation on the family and their Child was clear in the data, with this time being described emotionally and vividly. Most participants had been patients in PICU (many miles from home which added to the pressures of living or splitting their time between hospital and home, especially when there were other siblings at home. This is illustrated by the participant below:

"I was torn between my son in hospital and my five-year-old daughter at home and it killed me" (11 142).

As the relationship between the baby and the parents/family grew, there was some evidence that this became even more difficult with long-term hospitalisation. One mother below describes this process:

"it gets harder to leave your baby as they get older it gets even harder, because the bond is building every day" (11 169-170)

The data also highlighted that the PICU environment could be very frightening places for young affected Children and siblings due to the intensity of machines. Emotive events were constantly around them with them witnessing significant medical setbacks or even the death of other Children. This left parents' feeling concerned about the possible impact on the Child and siblings, as described vividly below:

"I know how much she saw when she was a Child, she was only five when N was born and because she sat with us in intensive care ...you know... every day for almost six months, and a kid that age to sit with all those doctors and nurses and machines running" (1 69-71).

Sibling relationships and bonding appeared for some, to be affected by this environment with the abnormality of the environment or long distances between them and their sibling becoming a block, whilst in others the opposite occurred. This is reflected upon below:

"Um there was an immediate bond between him and N the minute he saw him in the incubator the day after he was born, and he was just desperate to have him home," (10 46-48).

Meaningfulness within the family was found to be far more complex when there were siblings, than when it was a lone Child. For example: for mothers and fathers who were often split between meeting the needs of their hospitalised Child and their Child or Children at home. This split affected their family sense of *meaningfulness* as they were being pulled in widely different directions.

A vital part of meaningfulness to the families was a feeling that life is emotionally meaningful, which appeared to be closely associated with hope and emotional well-being. The multi-disciplinary team that supported the family had a central role in helping the family establish hope to start to plan for a future. In the early days on PICU parents reported that they could quickly lose hope without a consistent person to help them to get through the initial confusion, despair and in the planning for home. This is depicted below:

"*I just thought this was life in a hospital bed attached to a ventilator which was your first image, and you know she ……. (named nurse) was an amazing support in that transition from intensive care on to the ward"* (10 56-62)

Furthermore, all but one of the infants with CCHS in this study remained in hospital for a significant time after their birth. For this group of families, it became clear in the first hours of life that there were problems with the Child's breathing. Therefore, the first day at home, with the joy and fear of bringing home a Child dependent on LTV was mixed and amplified with the equally strong joy and apprehension, normally experienced by any parent taking home their new born baby. This interview quote sums up this complex mix of emotions:

"once we got him home and he was all ours ……. it was...one of the best days of our lives I think" (8 480).

Not feeling on their own once home, was also important to families. Data was collected, that highlighted it was important to have easy access to skilled specialist advice from people who knew the Child, family and Child's condition well. The following quotation illustrates just how important this could be to families:

"*quickly realising that you have got the hospital to back you up was really, really good, mmm they were the biggest thing for me."* (11 182-184)

Having a life that was meaningful was important to the families. '*Normality*' was perceived as something worth striving for, with getting home, being the primary goal. This focus on normality started in PICU. Initially focusing on smaller, more achievable, outcomes appeared to help create a sense of meaningfulness for parents. As time went by and the affected Child stabilised training and most importantly discharge preparation became central. However, this process took a considerable length of time. Anniversaries and birthdays became times of extreme divided loyalties and particularly important milestones in the passage of time. An illustration of this is offered in the subsequent passage:

"Sibling [named] was desperate to have him home and we were able to have him home the night before sibling's birthday so it was his fifth birthday so he was here the next day and it was so special cause he so wanted N to be part of it and it was kind of the first birthday celebration we had as a family since N had been born and it was just lovely that he was at home and he wasn't in hospital." (10 50-54)

Focusing discharge on family outcomes, such as a birthday or anniversary appeared to help put control back into the hands of the family.

Once home, the day to day management of CCHS became a normal part of family life. The diaries and interviews included extensive evidence of day to day family life. The play room scenarios in the focus group data highlighted how engaged the Children were living in the present and the activity they were enjoying. CCHS was focused upon when it became a barrier to whatever activity the parents, siblings or Child were engaged in. This focus on normality and achievement seemed to become a motivational drive in itself.

5.2.3 The Next Big Leap

Having personal outcomes to aim for appeared first in the data from the discharge home from hospital, which could be a stressful and difficult step. However, it appeared to be motivational, in that the families then had outcomes to aim for and build upon. These outcomes were closely related to Children and parents construct of a "normal" family life. Once home there was an ongoing pattern of families or individual family members focusing on their next goal. Within the diaries and interviews this was often characterised as *taking the next big leap*. The diary data collection period covered a school holiday period for most of the participants. Taking a Child away for the first time, was the next big leap for two of the families. One parent's feelings about taking her Child with CCHS away for the first time are presented here:

"School holiday and decided to have my first trip to my parent's caravan without carers or his father. My mum was happy to come with me and help look after everyone! The sun shone and had a great day – let's hope the night goes smoothly!" (26/10/B)

"Great day. My mum got up early with all the boys and let me get some much-needed sleep. So, we did it a night away, really pleased, just love doing stuff that I would have done if we didn't need to include a ventilator! Fantastic sunny day." (27/10/B)

The Children were even more focused on what they wanted to achieve. In the participatory play-based scenarios, the Children were highly engaged and motivated and were completely focused in the activity they were undertaking. This was clear in their suitcases (described in 4.13.4.1) in which camping equipment torches, and even a camp fire, were drawn. An example is illustrated in the picture presented below:

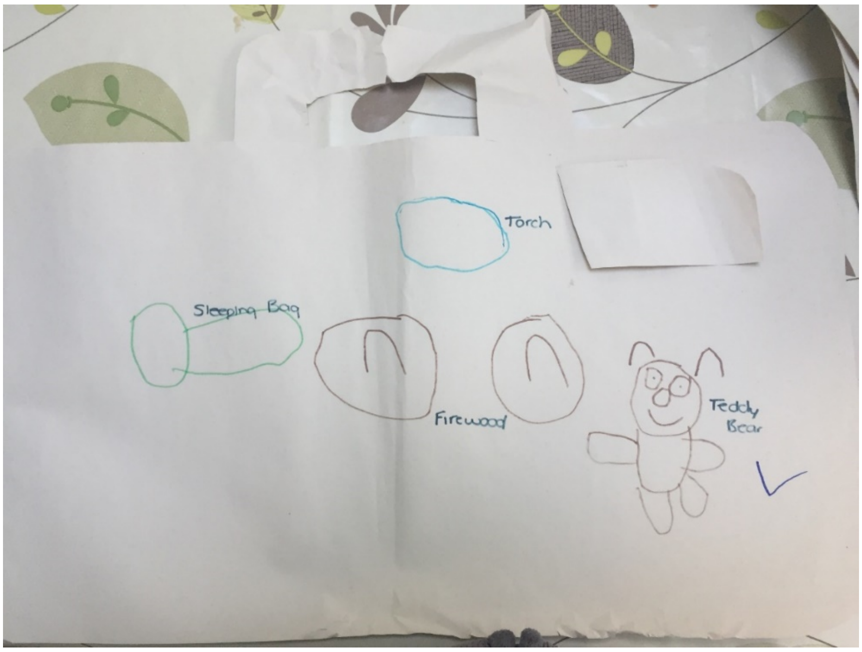


Figure 5.1 A Child's Suitcase Drawing (reproduced with permission)

This motivation became connected to CCHS, once it stood between them and what they wanted to achieve, becoming a driver to master the CCHS. Most Children diagnosed with CCHS initially have a tracheostomy due to initial intensity of support. A tracheostomy has significant effects in terms of speech, self-care, body image, equipment required and the undertaking of certain activities, due to the risk of a foreign body entering the airway. Therefore, most Children with a tracheostomy are encouraged to transition to mask ventilation.

The change from tracheostomy to mask ventilation was a big leap for Children and families. It also marked a significant step towards a more normal life unencumbered by medical equipment, invasive care routines and visual signs of medical support such as the tracheostomy tube. The motivational drive to make this change was identified in the data as the start of the process in three of the Children (participants 2,5,8) who made this important transition during the data collection phase. To start the process is a decision which needs to come from the Child. In the extract below, the decision was made by the Child to change from tracheostomy to mask ventilation to enable her to be like everyone else. The quick acceptance of the mask in contrast to a previous attempt, which had been based on a parental decision, demonstrates the importance of this change being based on the Child's motivation to change, to achieve to an outcome that was important to them:

"*she wanted it, it was her own decision, we were a bit unsure, aye we were a bit nervous about it actually, but she wanted it off herself she's getting to that age where she doesn't want any Children questioning her and she wants to go to school without the trachy. We tried the mask on her when she was about 5 ½ but she wouldn't tolerate it but this time she went down she just took to it like a duck to water they were actually quite shocked that as well at how quickly she transferred over to it" (5 416-439).*

5.2.4. A Well-Known and Trusted Team

The affected and non-affected Children of the parent participants in the study were one of the, if not the most important thing in their lives. The Children with CCHS, who participated in this study, had in all but one case gone through a period, where there had been doubt about their very survival. Once ventilation has been established and the Child stabilised there was due to the underlying effects of their CCHS, daily life-threatening risks. These risks occurred if the Child's condition was not closely monitored and ventilation commenced at any signs of them starting to go to sleep or becoming unwell.

To hand over the care of a young Child to another person is difficult for most parents, but for the parents in this study, having been close to the possibility of the loss of their Child, their Child having ongoing care needs essential to their survival made this process of handing over the care of their Child incredibly hard. Both practically (because of the knowledge and skill needed to manage the condition) and emotionally.

There was a need for a bond of trust between the professional team and the family, which was based on honesty and openness. This bond required time and commitment to establish. Moreover, giving an open and honest opinion and being open to discussion were particularly valued by families. Participants reported positively about carers, who were skilled carers, carers who interacted well with the families, but even more significantly they described carers as being ‘special people’, who focused on the Child and that for some it was not just a job, as described in this entry:

“you know when you have really good cover. Their immediate concern is for the Child - we as parents can always tell within the first few minutes of meeting a carer if they are special” (5/1/U).

There was a sense in the diaries and interviews that parents felt there were some carers who approached their role as vocation, exemplified by this extract:

“it’s not just a job they care about him as well, so those sorts of things stuck out to me as being really important people” (5/1/U).

Parents referred to being able to relax, when they could entrust the care to the carer, though noted that carers needed to understand how precious their Child was to them as this quote outlines:

“you don’t want people to just think of him as just as just a job you want people to, to know that he’s really special and they’ve got to look after him” (11 115-116).

The importance of some carers to the Children and the level of trust they had in them were exemplified in scenario one of the participatory activities when prompted about the need for medical equipment to be able to stay on camp one Child suggested that:

“A male carer in his team would have come with him and then everything would have been OK” (S1 63).

However, parents indicated that this was not always the case. Some carers were described much less positively, sometimes in terms of the individual but particularly carers from care providers, who didn’t undertake Child specific introductions and training or quickly rotated staff. Parents described the way that carers were managed and organised by the organisation that they worked for, as being a significant factor in allowing carers to become familiar and accepted in the family. Some families felt that professionalisation of care was going too far and taking away the special carers, and the ability to build up meaningful relationships. Families at times described this professionalisation as a boundary which, whilst they recognised the need for such “boundaries”, they felt they had gone too far, causing the loss of meaningfulness in the care package. There was also a sense that these “boundaries” were one sided in their implementation, as exemplified below:

“that’s hard to relinquish cause you want control of your son, umm and his health and wellbeing umm and its hard and you have to abide by their rules in your own home” (11 366-368).

For the Children and families, it was particularly hard when long-standing carers who were meaningful to them and their Child were changed as described in the following extract:

“I don’t like it when PCTs make change for the sake of making change, if someone is settled in a package, the carer’s happy here and the family are happy why move them if it doesn’t serve any purpose other than they think they need to recycle their staff, because this is people’s lives it’s just not just moving them from department to department, its people’s lives and the Children get attached umm so I would just I think they should get more credit, the carers” (11 399-405).

A regular team of carers, rather than a transient workforce, was also considered essential by parents, as they felt this was important to their Children’s emotional wellbeing. It was reported that Children could become scared and not sleep, if they did not know who was going to be in their bedroom watching over them at night. This became more of an issue as the Child became older and more aware. The growing significance of this to a Child is described below, first in the diary entry, then in more depth during the interview:

“tonight’s shift was covered by someone who was last here about two years ago. N was really upset going to bed tonight, the first time he has done this. Up until now ‘random’ carers annoyed me but now looks like it will be a big issue for N - something which I will need to address with the care team” (22/10/U).

“he’s a kid he doesn’t know any different than waking up and there being an adult sitting in his room, it’s not always the same person so you know that was a new thing. Umm him being just so upset because he didn’t know who it was who was coming, I didn’t know what the girl looked like even to give him a description of what she looked like and you just think god it’s no wonder he doesn’t want to go to sleep tonight” (10 523-529).

One-off or occasional carers who did not know the Child well enough to understand how to enable the Child to communicate their needs and wishes, were reported to scare the Child and in turn, take their voice away. Conversely, an established carer who knew the Child well was felt to be able to reassure the Child and give them a voice, as outlined in the following extracts:

*“and then with the carers coming in if they don’t know him as well it is very hard for him to tell them, (*referring to a Child with a tracheostomy who struggles to communicate*)” (2 266-267)*

“she knows his every need you know, and he will tell her, and no matter how upset he gets she will understand him because she could understand him when he had no speech” (2 278-280)

"she talked, and he signed and made gestures, heard him, talked and I looked, and I thought wow I don't think I have ever talked to him like that" (2 284-285).

5.2.5. Celebrating Achievements

Achievements appeared to be emotionally important to the Child. This meant in turn each achievement was deeply meaningful to the parents and wider family. One young person completed his own diary, which was full of sporting references and achievements, showing just how important sport was to him and his personal identity exemplified bellow:

"*Went to ………. college to play tennis I was playing doubles and won three matches out of four*" (26/7/B).

In the data, there were three main types of achievement - sport and exercise, special occasions, development and milestones of Childhood. It was in relation to sport and exercise, where the physical manifestations and risks of the condition were felt to be most pronounced. Parents were aware of this and were concerned about the risks and possible limitations around exercise. However, affected children and siblings knew there was extra vigilance but often exceeded expectations as below:

"N ran around and around the garden all day today and didn't show any signs of needing to sit down and have a rest - he was like any other Child" (7/11/B).

Achieving in sport related activity was also seen as development, as this was felt to be exceeding expectations and often achieving what was seen as impossible. Most parents had been through very difficult times in hospital, where survival and future prospects were very much in doubt, often leading to low initial expectations and real meaningfulness and pride on achievement, the following diary extract exemplifies this feeling:

"I mean he actually won a ...he won a race at his sports day the other day which ... when you think two years ago we probably didn't even think he would compete in a race" (8 78-79).

Overall, each developmental milestone was viewed as important as many families had initial low expectations of the affected Child. Risks from early hypoxia, (an inadequacy in the oxygen reaching the body's tissues) prolonged hospitalisation and supportive technology, such as tracheostomy tubes, had slowed development. The following quotation describes just how meaningful the progress could be to a parent:

"*I know it's really special whenever a Child says its first word or takes his first steps, but it was magnified beyond all kind of belief"* (10 309-311).

There were also significant condition management related setbacks in achievements. For example, in the following passage a tracheostomy tube was upsized - whilst this tighter fit improved the effectiveness of his ventilation, it also stopped the air bypassing the tube and going up through the larynx (vocal cords) improving ventilation, but with significant consequences for speech, the Children and family, as described below:

*"and then obviously when he had just started speaking, he got upsized in tubes so then went silent for a year" (*8 87-88).

Conversely, special occasions were significant markers of progress and often markers of the affected Child's social achievement, as revealed in the following passage:

"actually, her first big thing for us was ...erm...we got her home at 16 weeks just for the weekend, that was her christening" 5 569-580.

However, there was a fear and a risk element of Children being excluded from special occasions, such as birthday celebrations, due to fear of their condition. The meaningfulness of this type of event when it was achieved both to the Child and parent is typified in the following diary entry:

"N went to a friend's house after school today. N's house. He was so excited bless him as he never gets asked to anyone's house as everyone seems scared of his condition. He had a wonderful time it went better than I'd hoped, and I had a great time too chatting with another mum so happy day." (25/11/B)

5.2.6. Sibling Relationships

Diaries contained numerous references to siblings especially when asked 'the best thing that happened today' question. Siblings appeared to be a central part of both the parent's individual and the family's construct of meaningfulness. Siblings were highly significant both in their own right and in the lives of family members as captured in the following journal entry:

"Sibling [named] wrote an essay for A-level English called 'my hero' about her little brother it brought tears to our eyes" (12/1/U).

Sibling success in exams and at school was a real inspiration to parents as they worried that having to spend so much time supporting the affected sibling could disadvantage them. This is summed up in the following quotation:

"Sibling [named] got a really good report at Parents night – very proud of him. I often worry about the impact of N's medical needs and the attention and time that it takes and how this impacts on his older brothers, so it's nice to get feedback that they are doing well and that hopefully it's just a positive experience for them" (18/10/B).

Siblings were observed in the playroom watching over their brother or sister with CCHS. Some siblings when being checked into the playroom were specifically identified as the person who could sort out any problems with their brother or sister (S1 ON 10:1-3)

5.2.7. Childhood and Taking Risks

Families once confident in their Child's care progressed to consider the balance between restrictive safety focused care, to considering what they wanted to achieve as a family and the balance between quality and quantity of life. This is summed up in the following quotation:

*"let's do it he's very much quality over quantity always he always has been, and you know in the early days we didn't know you know what the life span was" (*7 502-504)

It became clear from the data that parents were balancing quality of life and risk. This was most apparent in the data around the importance of certain activities to ensure that the Child had a normal enjoyable Childhood. In this diary quote a level of risk is indicated to achieve a Childhood experience:

"went to the beach (ssh don't tell anyone) N loved jumping the waves in my arms." (23/9/B)

Interestingly, swimming and paddling were felt to be risky, because when affected Children hold their breath, they have none of the normal physiological prompts to surface and take a breath. Those with tracheostomies have, in addition, a high risk of aspiration of water into the lungs while swimming or paddling. However, despite these risks and complexities, swimming and paddling were perceived as positive activities, that were worth doing as they were felt to be meaningful, motivating and mood enhancing. It was an activity linked to perceptions of normality and family life as noted in this diary entry:

"*Swimming together as a family was lovely. It made us feel "normal".* (18/8/E)

The adults took calculated risks to enable their Children to achieve what they saw as important milestones of Childhood. They were motivated to overcome CCHS to enable their Child and family to have as normal a life as possible. For the Children in an exciting activity, their focus was on the activity with CCHS being a responsibility of their parents. The management and potential risks of CCHS were forgotten, as the focus on the desired activity became central. When risk became a block to Children undertaking something they really wanted to do they became highly motivated. Swimming for one Child was the next big leap that motivated them to start the transition from tracheotomy to mask ventilation. How Children and their parents comprehend and contextualise CCHS will be considered in the following section.

5.3. Comprehensibility - the Cognitive Component

As a recap, comprehensibility is when a person is able to make sense and comprehend, thus confronting stimuli in making sense of both ordered or explicable events within the context of their daily lives.

5.3.1. Learning Through Experience

Time in a PICU or on a ward was key in building confidence and competence. Early contact with other families in the CCHSUKSG helped parents use this time to make sense of their Children's condition, build their skills and develop their confidence:

"*one of the other families told me when we were in PICU...to learn to do it in the hospital on your own first because in an emergency situation you are going to be on your own so it's best to learn to do it in a non-emergency situation, so you feel more confident and don't panic*" (9-107-110).

Helping parents to take part in the clinical care of their Child's complex care needs early on in the PICU or wards, appeared to help parents to start to take control of the condition and was a very important milestone in taking on ownership of the condition. This was often seen as highly significant to parents as described in this quote:

"*The best things, the best thing ... was ... being trained and being able to take N off the unit*" (9 291).

Once the family was at home there were possible complications of the condition, which they may not yet have experienced. For example, CCHS Children do not increase their respiratory rate when unwell, requiring extra vigilance, so often these complications do not become apparent until the Child is in the sole care of the family. This could cause concern for parents as described below:

"*went to the GP – N had a chest infection (slight) next time require increased in ventilation will take to GP to get checked out. Now on AB timed ventilation back to normal tonight*" (26/9/U).

The combination of a complex condition, with highly complex equipment and management, appeared to result in the need for considerable skill and knowledge to care for a Child with CCHS. Further, the necessity to maintain a Child's airway could raise these risks further still. Hence, taking control of the Child's medical condition was a significant undertaking for parents. However, some parents such as the one quoted below, showed significant adaption in very early stages of their Child's condition, using difficult events as a learning experience:

"*N pulled his trachy out for the first time so we now know what it looks and sounds like*" (4/9/U).

Due to family and work commitments data highlighted that one partner, frequently mothers in this group, often spent more time on PICU or wards so were more advanced with the skills and confidence needed to provide care at home. Once home the other partner had to make sense of the same skills and thus catch up. This is described in the following diary entries:

"*Father done a trachy change*" (4/5/B).

"*Knowing that Father is able to help in case of an emergency*" (4/5/E).

Due to the immense amount of complex equipment and the need to provide continual observation and care, overcoming the environmental issues were an important component of care, that featured in the diaries such as the quote below:

"*Moved all the furniture around in the cottage so we could fit N's bed in with us. Set up all his equipment ready for bedtime. Lovely to sleep in the same bed as hubby when caring for N. After half a night, we decided to move the sats machine as it was in the wrong place and we had to keep getting up to silence it.*" (15/8/E)

As the parent's confidence grew, data highlighted that they started to develop the ability to debate and challenge decisions made by the care team, where they felt decisions were not always in the best interest of their Child. In the following passage a parent convinces the nursing team to train up her son's new nursery team in ventilation:

*"… the outcome was that they are now going to ventilator train the staff at the nursery, I feel much relief about that because I was very nervous ……." (*10 104-108)

5.3.2 Growing up with Congenital Central Hypoventilation Syndrome

Children with CCHS are continually growing and developing as any 'normal Child' would be expected to do, but at the same time they gradually takeover the care of their own condition. Within the data self-care was viewed by Children and parents as a precursor to safety, independence and normality. Each step towards self-care therefore, appeared very important to the affected Child and their family. This developmental process required the family to support the Child to go through the stages of understanding and learning about their condition and how to manage it. However, for the parents, as their Child took steps towards self-management, they also had to reconceptualise, during each one of these stages, how they cared for and supported their Child to safely to manage their CCHS. This process of selfcare therefore, had two key elements for parents: the first supporting their Child through the stages of self-care and the second, how they adapted the management of their Child's CCHS as they progressed towards being self-caring. This process therefore resulted in learning and adaption being a constant feature of their lives

"It's all a learning curve, it has been a learning curve from the day he was born" (2. 415).

Initial understanding of CCHS for the Child was focused on ensuring that they would be able to verbalise to an adult that their tracheostomy tube was blocking, or indicate they needed suction to clear some secretions that had built up in the tracheostomy tube making breathing difficult. The Child also knew that when they were on the ventilator to go to sleep the risks dropped dramatically. This reduced parental concern significantly and is described below:

"Going to bed tonight N said that he couldn't breathe and that he needed a Tube change. We changed it and it was clear so possibly just a bedtime delay tactic but love that he is so aware and able to vocalise his needs. It makes leaving him in the care of others so much easier, we only ever leave him with trained people but knowing that he can express his own needs makes it that bit easier..." (14/10/E)

The next significant stage of understanding for the Child about CCHS was that it was their CCHS. Children's early understanding of CCHS was to see CCHS as a parental issue rather than their own. This was vividly portrayed when the Children undertook the river exercise (Scenario One see 4.1.4.3). During this exercise Children drew onto paper suitcases what they would take on an imaginary camping trip (which was then mocked up for them to experience). Interestingly, during the boat trip in the event, of the older Children only two took any ventilation equipment (both discarded it in favour of other items from their suitcase when the storm hit (Scenario One). The younger Children did not pack any medical equipment, but drew pictures of their parents in their case (literally as a picture of them in the suitcase Figure 5.2). When asked on arrival to their camp what they needed to stay the night after showing the researcher all the contents of their cases, they were asked if they needed any equipment to stay such as ventilators. They simply replied "I've got my mummy" since they had put their mummies in their cases or just assumed, they were there. It seemed as though they had transferred their CCHS to their parents and proceeded to get on with life unencumbered by CCHS.

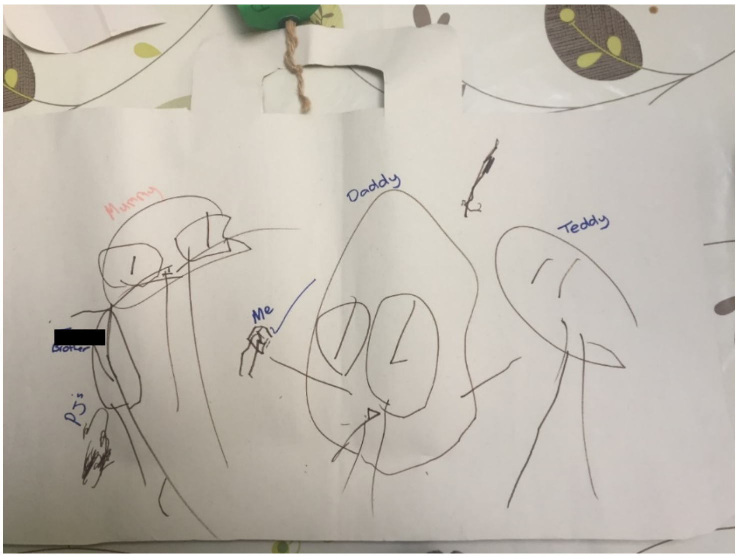


Figure 5.2 One of the Younger Children's Suitcase Drawings Containing the Whole Family (reproduced with permission)

Children at this stage were in the preschool and primary school age. When discussing equipment required to breathe when asleep (in scenario two see 4.1.4.3), the Children could describe in great detail what they required and why. However, they either saw this as their parents' issue and again took their parent along with them or did not connect the need for the equipment on an overnight camping trip until the diver (used in scenario two) explained what equipment he needed to breathe. When asked about their breathing equipment it was clear they understood its purpose and importance as exemplified below:

"I have my machine to breath at night" (S2P12).

There appeared to be a strong link between their understanding and the context they found themselves in. The understanding they described in a planned routine situation was not apparent in the scenarios where they were outside of their normal context.

The subsequent step, was then perceived by parents to be, when a Child showed wider understanding outside routine care. Parents described when their Child started to take over ownership of their CCHS and began to make sense of it. They began to appraise situations and to take make decisions, by assessing their own knowledge of the medical implications of their condition in new situations. In the next quote, parents recall the point at which their daughter started to consider the implications of her condition in a new situation:

"*And we know that she worries about it because remember the time we went over the ferry and we didn't have her ventilator with us, the ……… ferry, you'll have passed it at the wee bend. We went over that, and we were just on a trip and we didn't have the vent with us and she started panicking, what if I fall asleep ……" (5 256-261).*

For Children, who are tracheostomy ventilated, the move to mask ventilation is a key step towards self-care, which like most developmental steps evolves over time. For Children with CCHS moving from a tracheostomy to a mask is a major move towards independence. For this transition to occur successfully, it has to be a decision led by the Child, as it is the Child who has to learn to cope with this new interface. Often the trigger for the Child is a desire to be able to undertake an activity, which would be impossible without a change in their current interface for ventilation.

As described in 4.10 the research was launched at a CCHSUKSG Family Day where the researcher had organised a play room for the Children. This was not provided as part of the research in the first phase, but as a thank you for allowing the research to be launched on the day. However, the diaries and interviews later showed how important the play room was. For three Children, it started the process that lead to the removal of their tracheostomies and a transfer to a mask ventilation interface. The following diary and interview quotations tracks this process from initial idea through to decision and change over to mask ventilation:

"Yes, because actually our family group after that conference that's where I think that is where the initial spark was" (2 127-128).

"The kids were just discussing it, yeah these kids were just discussing it between themselves and then J just made the comment to A that umm girls wear masks, which I thought was a bad comment, oh crap he is never going to wear a mask but umm so I think that was all going on in the background," (2 164-168).

"because it was that day that he said that mum I'm not wearing my trachy tonight, I'll wear the mask, I want to wear the mask tonight and he hasn't wore it hasn't gone through the trachy since." (2 180-182).

With regard to the transition of using the mask, the CCHSUKSG and their Facebook page were perceived as significant resources for families to use to help their Children become independent. This was through discussions, sharing stories and even sharing videos of care, as described below:

"I asked the group on the Facebook page if anyone could post a video of their masked Child getting ready for bed, so N could see the mask etc. 3 people did it and N loved it, extremely positive!" (25/8/U).

Once a Child is mask ventilated there was the potential for big steps in self-care, as illustrated in the diary extract below:

"N put himself to bed! Well almost. After he'd brushed his teeth he went straight into his bedroom and put Vaseline all round his face and then put his mask on! He is so clever. I put his sats probe on round his toe and then he was all set. He was so proud of himself and I am so proud of him" (21/11/B).

This important process was the beginning of transferring responsibility for the management of the Child's CCHS from the parent to the Child. Whilst these steps were of great importance to the Child, they were also extremely challenging and frightening for parents, however they were motivated and excited by their Child's developmental progress.

5.3.3 Going to School with Congenital Central Hypoventilation Syndrome

Education is a key part of all Children's physical, emotional and social development. Selecting a school was felt to be difficult for families including situations where a Child has no voice due to a tracheostomy tube or developmental delay. Often a difficult decision had to be made about whether specialist or mainstream education should be chosen. This is highlighted in the following interview quotation:

*“weren’t sure that he was gonna be okay for mainstream nursery as he had no language, so we had a big push with speech and language” (*10 93-95).

The letting go involved the affected Child starting preschool or school and was seen as a significant step for most parents, as outlined by the parent quoted below:

“.. it’s leaving her alone is school, that was a big thing” (5 617).

This complexity and risk were also a major issue for preschool, schools and medical teams, often leading to long delays in getting into preschool or school, as in the experience of this participant:

“*she was taken out of nursery for the first half of year until they got trained*” (5 1149-1150).

The training of staff to enable Children to attend preschool or school was time-consuming. Few of the parents received support from their nursing teams in this frightening and complex transition. This often resulted in them training the school staff, with no support from the clinical team. The following series of diary entries follow a mother’s journey from back up to finally leaving the preschool staff in charge:

“*Making progress with nursery training, N settling really well, will be good for him for me to not be in the cloakroom and I’m looking forward to some time to myself – to go to the supermarket or do the housework!”* (1/10/E).

“The nursery staff completed their trachy and resus training yesterday, so I can finally leave N at nursery and escape the cloakroom. I don’t feel I can go far as his ventilator isn’t at nursery, they would need to hand bag him if the need arose, so I don’t feel comfortable being far away, but great that I can go home, and N can enjoy a normal nursery experience without his mum in the cloakroom.” (11/10/B).

Some parents enjoyed this experience and time with their Child at school whilst others felt it was an invasion into their Child’s independence. Special education needs coordinators (SENCOs) were viewed as a key to enabling access to school and as key workers. Children with tracheostomies had particular issues with engaging with school and peers (due to communication issues caused by the tracheostomy) however schools were praised for their efforts in engaging them, as in the following quotation,

“I’ve no criticism of them because they brought on his speech and even in school they were doing sign language for the school poem and they had the whole class doing sign language and ... they really, really helped N,” (2 95-97).

Exercise and complications of sedentary activities were particularly important in CCHS in the more sedentary school environment. Children, who have this condition, are unable to adjust their respiratory rate in response to exercise. This meant that some were liable to become unwell when exercising or undertaking long periods of sedentary activity, such as in school lessons. This complex balance and the skills needed to care for Children with CCHS in a school environment is outlined in this entry:

"Today at school N had PE. He was extremely tired afterwards and went very pale but fortunately N's TA knew exactly what to do and made him sit down and rest and have some water. It's very difficult as he does not show signs of respiratory distress like other people. Am so grateful that half the school is trained up on N's vent and know what to look out for" (24/11/E).

School staff were seen as particularly caring and supportive parents derived support from involvement in school life, as demonstrated in this diary entry:

"both his mainstream school and special needs school are fantastic and make life a lot less stressful when we know he is in good hands" (10/1/E).

Parents reported that they grew to have the normal expectations for their Child at school, such as succeeding, learning and achieving. These stories had a particular poignancy, particularly, as many did not expect them to be able to participate in this way.

The responsibility for a young person with CCHS continues beyond normal limits of parental responsibility and on into adulthood, as exemplified below:

*"went to brother's stag do in Leeds and mum came and picked us up when we were ready to go back to the cottage mum and dad looking after me during the night" (27/7/*B)

5.3.4. A Family Enterprise Normalising Care

A highly complex medical condition such as CCHS can be a major barrier to normal Childhood in terms of family care and support. Developing comprehension across each of a family's core members enabled a change or increase in family support.

The siblings were perceived as key core family members in making sense of the condition in most family structures. Depending on their age at the affected sibling's diagnosis, their understanding developed over a short or long period of time. Siblings initially think their brother or sister will get better. Understanding that CCHS will not go away is part of the process of understanding, acceptance and normalisation. Helping Children to gain the understanding that CCHS is permanent was, however, viewed as problematic. In the following extract, a parent discusses the moment the siblings started to understand this concept:

"*We were discussing CCHS at dinner tonight and it appears that sibling one and sibling two really got a lot from the time spent with the other kids at the family meeting at the weekend. We have always said that N will always need a ventilator and that one day he will use a mask, but I* *think after meeting some of the older kids they better appreciate that it is a ‘forever thing’ and a bit more about what the mask might be like for N*” (2/10/U).

Siblings who understood their brother or sister’s condition were felt to be in a unique position to help watch over the affected Child without adult intrusion. Older siblings who were trained, tended to spend significant amounts of time in the same place as the affected Child, so were in a unique position to help out in an emergency passing equipment or seeking adult assistance. These siblings, both boys and girls, became fully trained enabling them to go out unsupported or even to provide direct care. The proceeding diary entry captures this process:

“N sibling done a trachy change, knowing that Sibling is able to help out in an emergency” (8/5/E).

A sibling who was trained thus, enabled an affected Child to discover increasing independence with other Children, without the intrusion of adult oversight. The following interview quotations outline this:

“The middle sibling she’s…. we trained her as well and she’ll like maybe take her for a walk round the block, with the dog, things like that which is nice for her to be able to do these things (5 1093-1097Participant. 1).

“Older brother has actually taken unpaid leave when umm when my mum was really poorly, and I moved in with her” (7 328-330).

Siblings were noted in the play room prior to scenario one and two, as watching their sibling with CCHS and taking them back and forth to their parents (S1 S2 RN1).

Siblings were therefore viewed as essential in the family unit’s way of conceptualising, managing and normalising of CCHS. Both the diaries and interviews demonstrated a real balance between the Child with CCHS and the unaffected siblings. There was little evidence in this group of any sibling being resentful or negative, which in the wider literature on disability and young carers, is perceived as an issue for some siblings of disabled Children. One sibling in the focus group, now in his thirties, described:

“*the key reason for not resenting the constraints of my brother’s condition was the sense that managing my brothers CCHS was a family endeavour that I was very much involved in from a very young age*.” (FG2 P8)

The ability to deliver care was also identified in extended families. The support offered by extended family members was highly valued by all, but two families, one who didn’t have access to such care and the second where friends appeared to take this role. The competence and confidence required to undertake this care takes time to build up both on the part of the relative and the parent and Child, as seen in the following interview extract:

*“increasingly feeling confident that we can leave N and get further away and again just do the things that we would normally have done” (*10 290-291).

The ability of family members to gain this competence and confidence was perceived as being influenced by their level of contact and the training they received formally and informally. In this example the importance of competence and confidence is discussed:

“My sister often visits on a Monday and when it’s time to collect the older boys from school we would all go, or B and I would go, but today my sister said to leave N with her. She has had some training, but it was a while ago and she’s never used it much, but it was great that she felt confident enough to be left with him for a short while – so much more normal for us all” (15/10/B).

As in the following example parents worked at keeping extended family members competent and confident:

“Mum put N to bed to keep her training up” (11/8/U).

The complexity, nature and quantity of equipment which need to be set up, including ventilators, oxygen saturation monitors and suction machines was perceived as a concern in extended family care. Parents reported their confidence could be affected by mistakes or omissions and thus their confidence, to use an extended family member to help was altered as reflected in this passage:

“my mum would walk up to the little park up there and she would come back and say do you have a suction machine” (10 280-282).

When reflecting over a time period in the diaries, there was an acknowledgement of how significant the impact of this support is, which was not always acknowledged in day to day life:

“…. coz you do take things for granted… specially our parents, I think, you don’t realise, I’m like ooh parents coming” (8 265-266)

5.3.5 Facing Complications and other Unknowns

The most significant complication in CCHS which parents often have not been exposed to in the early days of diagnosis are ‘Sinus Pauses’. This represents a serious risk in Children with CCHS who have the potential to have or develop this complication. Sinus pauses are episodes, when their Child’s heart stops beating, due to cardiac pauses, thus creating immense fear and uncertainty. There were some significant concerns amongst the families who knew about this risk. A member of the group had recently died as a result of a sinus pause. A sinus pause can also be linked to anoxia (lack of oxygen to the brain sometimes unnoticed as it can self-resolve) which especially in very young Children can lead to brain damage. This concern is shared in the following interview extract

"*when N asks all sorts of questions about things we know his wee mind is active! We sometimes wonder if he had been diagnosed with a heart rhythm problem, sinus pauses, earlier would his mental development been a lot better than it is now for his age*" (E/4/1/U).

Not all the families were aware of this issue, until it was an agenda item at the CCHSUKSG as shown by the following diary entry:

"Following on from the Conference I now understand that all CCHS Children, whatever their mutation, are at risk of cardiac problems" (1/11/U).

There is also evidence of the parents asking for more in-depth information now they had a greater awareness of the issue, as in the subsequent journal records:

"Found out N had no pauses at all at previous halter tests I knew he had no significant pauses" (4/2/U).

The parents gained reassurance from having the Halter test undertaken (a 24-hour heart monitoring test undertaken to detect sinus pause) and reassurance from their medical team, which allowed them to refocus on the wider family life again. The families also recognised that it is an area where much is still unknown, but were comforted by knowing that medical advancements were occurring in this field. This is highlighted below:

"Received another email from N's consultant today saying he was discussing CCHS with other people in Europe and the US. This should clear up issues on testing requirements" (5/8/U).

The CCHSUKSG families compared the management of their Child and asked for further tests. The group is reassuring in the sense that they see other Children having and overcoming pacemaker operations (treatment for sinus pauses if identified) however it makes it an issue that is very close having lost one member to this complication.

5.4. Manageability

As a reminder, manageability is the behavioural component of the framework, or in other words, "the extent to which one perceives that the resources at one's disposal are adequate to meet the demands posed by the stimuli that bombard one" (Antonovsky 1987:17). From a salutogenic perspective when an individual or family have a high-level of manageability, they believe that any problem can be solved by their own actions and abilities or the support that they can access from those around them. This collection of themes considers this construct for the Children and families within this study.

5.4.1. Understanding the Wider Context of Care

As mentioned elsewhere, most of the Children were diagnosed in PICU. In terms of manageability, being many miles from home, made the pressures of splitting their time between hospital and home difficult, especially when there were siblings at home. This appeared to drive families to take family decisions, that they might not have otherwise done. Some of these were extreme, such as moving the whole family or changing the Children's schools to be together, as described below:

"So, we actually had to move the rest of the Children down and put them into erm school down near the hospital" (5 100-101).

The difficulties imposed on family life at this time were particularly hard on siblings especially when they had to move, as shown below:

"For the rest of the kids it was a big...huge thing for them …. N was only four and you know that he had to leave all their friends and start kind of a new life" (5 129-134).

Consequently, for discharge to be manageable for this group of families, it needed to be manageable within the wider context of the family. Discharge needed to be a process, where the Child and family were at the centre of making decisions, on how their new life might be (and that this should not be solely governed by a predetermined, medically driven process). The importance of discharge coordination, with a team which looked holistically at their health, medical and social needs was thus essential as evidenced below:

"… (local discharge coordinator) was dealing with N's condition …not the regional specialist unit …even as a NHS employee she seems to be very detached from the medical side of things and much more attached to the personal family side of things" (8 971-973).

As care for a Child with CCHS was viewed as specialist and overseen by regional Children's centres, the practical and financial issues of having an affected Child in intensive care were profound. These specialist centres were normally based in large cities, making commuting complex, expensive and time consuming, these issues were exacerbated by the long periods of time spent on PICU as illustrated:

"We were a year and a half in hospital" (8-95).

"It took about 2 ½ hours I say that's the average" (8-126).

One family found they had to give up work:

"It was obviously life changing for us coz... we both had to give our er.... give up our jobs" (8 *643).*

In terms of financial support, families gradually became aware of this, but it was felt that the various claim forms were bewildering. If professionals took time to support them this was much appreciated. Once completed, the benefit start date was set on discharge, but with many families, they had a phased discharge, so it was not until final discharge that benefits started making a real difference to family finances, as illustrated:

"Got awarded high rate for DLA and Mobility which should make things easier" (13/2/U)

5.4.2. Manageability of Care at Home

Manageability within the PICU was reported to be very different from manageability in the home and community environment. The true complexity and burden of highly technological care, was also only clear once the Child and family got home. Some families had a *phased discharge* package (this is when a family build up from a night at home to full time sequentially) whilst others did not. Families reported that they needed to know how to access skilled specialist advice from nurses, who knew the family, Child and their condition well, on an ongoing basis. They described needing a key contact. Families viewed the key contact role (commonly referred to as a named nurse / key worker in the literature) as being effective, when the role encompassed more than the technical and medical aspects of CCHS. The interviewee below outlines the wider aspects of the role:

"About the caring and the nurturing side as well and the impact that has on the Child and the family" (10 576-567).

Support staff also needed a range of skills including technical nursing skills. In addition, having knowledge of the Children and family, as the following respondent explained:

"to me is important: the person who is doing the training knew N didn't just know his equipment, but she actually knew him" (7 634-638).

Consistent nursing staff who knew the Child and family well, as described in the quote above (where it was provided) was vital to families. This component was found in surprisingly few of the family's experience of care. Where it was present, it was associated with teams who provided a holistic approach to care, encompassing case management support, ongoing Child support and training in addition to the care package. This appeared to be particularly important, as the Child and family came to terms with the enormous changes that have occurred in their Child and family life.

Access to emergency support and advice was felt to be required consistently or '*24/7'* in order to reduce the feeling of isolation in times of need. Further, it was suggested that this advice also needed to come from clinical and nursing teams that knew the Child and family well.

5.4.3. Manageable and Flexible Family Care Packages

Parents in the study expressed the importance of regular and sustainable care packages, in enabling them to safely manage the care of their Child. In the subsequent extract a family describe and reflect on the significance of their care package:

"...you know .... a third of our life minimum is, well more than that it's over a third of our life is spent with ...with a care team around us.... you do realise how big a part they are and erm... how much they do do help" (8314-322).

In CCHS whether mask or tracheostomy ventilated, there was widespread agreement amongst the study participants, that having carers to cover nights was essential to manage. Some parents however, as captured in the next diary entry, liked to have a night without professional carers to allow some privacy:

"made life easier having no care tonight as it is nice to get our life back every once in a while. Still look forward to Mondays when care resumes though" (27/8/E).

However, without sleep at night, parents felt that they became easily exhausted and could not manage in day to day life. In the interview excerpts below, parents outline how much is involved in caring for a Child with CCHS, but also the need to balance formal care with the parental role:

"shattered I definitely couldn't do it without ... I certainly couldn't work … umm... yeah just cause it's the amount of times like you have to get up 5 or 6 times in the night and it is shattering" (9 58-59).

".. the difficult thing I found was a lot of the time was I ... only want the carers in the house when I couldn't look after him, so I was never gonna be the mum to say I want 24/7 care" (7 578-580).

Flexibility was important to families, but to be able to do the things that most families take for granted, significant extra effort and flexibility was required. Some parents noted that their care providers worked with them to achieve this as below:

"A date has been agreed for a home assessment to be carried out at my mum and dad's house, if approved this will mean that N can go to his Grandparents for a sleep over, with his brothers and husband and I can have some proper respite" (9/10/E).

To have a family evening out required care staff to adapt as below:

"Out for family dinner with friends and were able to ask the carer to start a little later than normal, enabling us to stay out with the other family and not need to rush home" (20/10/E).

However, some families felt care was a service driven by a focus on staff to cover shifts and staff needs, rather than what was important to families. Families described family focused care as the ideal approach, where there was a team approach to care with the family part of, and central to decisions. Service driven care approaches had a significant effect on the family's ability to manage a normal family life as outlined below:

"*I think there is too much red tape, there's too much, too many walls, they make, umm you know carers are not allowed to be in the house on their own with the Child, umm there's no flexibility, you're told what your hours are, umm regardless of whether they fit, umm there's no flexibility at all umm and trying to change anything just it's just impossible. Umm those sort of things I would like to change. I would like them to listen to the families"* (11 340-346).

Conversely, there were examples where such listening and flexibility was apparent. For example, in one young person's personal health budget package a person focused approach was taken, as outlined in the following passage

"nurse [is] picking me up from club when I ring her and bringing me home" (6/7/E).

Parents felt it was important to be able to contact carers, if they were going to be home late due to an over running schedule or family event. However, as described below parents were often discouraged by health care providers from contacting their carers outside work hours:

"we're not allowed to have their mobile numbers so again if you are running late you can't text somebody to say oh listen …" (10 541-543).

There was a mix within data drawn between organisations, which tried to provide a flexible service that led to family driven care versus service providers which were service driven and did not include families in decisions, delivering care with generically trained staff, who didn't always know the Child and who worked non-flexible shifts. However regular, trusted, carers who were specifically trained for a Child and knew them and the family well and were willing to adapt to family need, such as supporting holidays were highly valued and vital in terms of manageability. However, it was recognised that the pool of carers needed to be sufficiently large to enable cover during times of sickness as described here:

"*maybe if there were six or eight people who came here on a regular basis and they were with N and they were with another Child, you if there was a bigger pool"* (10 465-467).

The majority of diary and interviews were highly complementary about the care staff, in particular the perceived skills possessed, as explained below:

"*had a temperature in the night and the carers looked after him brilliantly, they didn't even call me down to see him they just got on and cared for him while I slept. They are awesome"* (4/11/E C).

However, there were some carers who were not positively viewed, and some parents intimated that they had concerns that some carers went to sleep during the night shift, which could have had catastrophic consequences. Overall however, there was also significant dependence and trust in the care team without which life would be unmanageable as described below:

“life is planned with precision (laugh), you know umm everything is planned and you know, and the reliability of having the carers turn up every night is just, umm, you just couldn’t live without it” (11 233-236)

5.4.4. The Developing Child

There was a physical and cognitive developmental component in the diaries and interviews. The cognitive component has been considered in comprehensibility and the physical component will be considered here. Physical development has a significant impact on manageability. Anatomically as the Child grows, their airways and therefore their tracheostomy tubes become bigger making them less likely to block. When the Child is young, they can require frequent episodes of sleep requiring regular ventilation. They may also have varying activity patterns, but as the Child became older and less likely to sleep in the day, management becomes easier. The following interview extract describes this change:

“he is more predictable now if I went to the swings with him I wouldn’t necessarily take his ventilator, like within the village because I know when like he doesn’t drop like a hat in the pushchair and things like that so that has certainly made life easier” (9 339-341).

Many Children with CCHS have tracheostomy tubes initially, making speech or nonverbal communication harder to acquire. Once the Children could communicate their needs, they can engage those around them and when there is a problem, they can let others know what their needs are. The following extract illustrates how significant this change is to the manageability of the condition:

“with his speech improving it makes it easier for new people to be able to get into that inner circle” (2 295-296).

5.4.5. Equipment to Live

Equipment was a vital part of life with CCHS, supporting life or in warning of impending issues. Therefore, equipment being available, fully functional with back-ups for all key equipment was essential. Of equal importance was having easy to navigate systems to support this vital equipment, such as consumables and maintenance. The following extract shows the importance of this issue:

"we had a second humidifier, so when the current humidifier was continuously alarming at one am, so we could just quickly swap over and go back to sleep" (5/9/E)

In the example below, a parent discusses issues with an oxygen saturation monitor, which is a key instrument to warn if a Child is not breathing or ventilating effectively. The alarm limits that are safe for a Child and an adult are different, in particular the acceptable pulse rate. Adult alarm limits would lead to an oxygen saturation monitor alarming continuously on a small Child as their normal pulse rate is much higher than an adult. Conversely an adult has a much lower threshold for a low pulse known as bradycardia than a Child. Therefore, an adult minimum pulse alarm limit is much lower and would not trigger a low pulse alarm until well below the safe low pulse rate for a Child. On some monitors, like the one referred to below, setting of alarm limits is relatively complex but vital, as when switched on, they default to standard adult limits. To overcome needing to change these limits each time for a Child, the manufacturers have included a patient specific option, so a Child's limits can be programmed in so that as soon as the machine is switched on it defaults to the programmed Child specific limits. In the quotation below parents had not been shown this feature. This created an unnecessary risk in this case and prevented less confident carers, such as grandparents giving care:

"we....worked out after having the SATS machine for 4 years, ... that we don't need to programme it every night now….we have managed to programme it so that when you ….turn the button on it's set for N's settings and that's been something that's held back my mum and (partner's name) mum from actually looking after the kit, or shall we say putting him to bed when we've gone out it's been a case of we put...we put .. N to bed, and then we go out and so we could never go out, but now, all they've got to do is push one button and it's all done and so that's gonna make our lives ... an absolute bunch easier they should tell you that when you come out of the hospital …. how to do that, they should set it for your Child" (8 397- 427).

Servicing of equipment could be a significant logistical obstacle for some parents, but simple solutions such as home servicing, appeared to reduce these issues dramatically, as shown below:

*"EBME have made an appointment to come out during the half term break and service all N's equipment. This is much easier for us as all the equipment will be here instead of half of it being at school with N" (*22/11/U).

The ability to monitor oxygen saturations in the car was noted by several parents, as being particularly important to them. This is because a simple hand-held monitor, that runs on batteries, can monitor blood oxygen levels. Therefore, if a Child with CCHS start to go to sleep and stops breathing and their blood oxygen levels fall, this causes the monitor to alarm. This may also occur if the Child is unwell. This is illustrated below:

"I am able to monitor N's sats in the car due to small machine which alerted me that N was under the weather" (24/9/E).

The size, weight and importance of the equipment, necessitated easy access to transport, which was particularly important to parents' whose Children were under three years of age, and who needed frequent sleeps, requiring access to ventilation. The availability of blue badges was important - provided by local authorities for Children and adults with disabilities, these enabled families to park in disabled bays which are normally close to shops or facilities and have larger bays. For a group of Children, like the participants, who have significant and complex health needs and are dependent on extensive and heavy, bulky equipment this easy parking facility is very important. In the following diary record a special day out proved overwhelming, but easy access to the car and equipment appeared to help to resolve the issue:

"Disability Card for parking and easy access, but all day at the show wrecked N. Journey home had a sick boy very sweaty" (19/2/E)

5.4.6. Communities of Support

Extended family support was described by families in terms of direct care, by a trained family member or an untrained family member, with general awareness of CCHS (or indirect care where a family member helped with logistical issues or support for siblings). Family support was viewed by most families as a key component to their ability to manage. This provision enabled the responsibility of care to become more of a shared responsibility. This support helped families to return to a more typical family life and eased the pressure on families. The need to constantly watch a young Child with CCHS in case they went to sleep and stopped breathing could be particularly difficult. An example of valuable indirect care was identified when an adult with good awareness of CCHS and the potential risks was able to help, by being an extra set of eyes at busy social or leisure activity venues seen in the following diary extract:

"having four of us out to watch and look after N" (21/5/B).

However, this direct or indirect support was not always available or accessible to all families. When support was available it was seen as a "normal" form of family care and adaption, and was most commonly taken on by grandparents, though aunts and uncles also featured as a significant group. The type of care offered by extended families was similar to that the extended family offered other siblings, but was curtailed if the family member was not trained. The availability of trained extended family support allowed for parents to focus on siblings, their relationships and developing wider friendships.

The care of siblings was the most common and valued form of indirect extended family care, allowing siblings to access opportunities and activities, that may have been difficult for the affected sibling to attend. Indirect or untrained care also offered a second pair of eyes and an extra pair of hands to carry all the equipment, particularly on holidays and days out as in this example, where the advantages were pointed out by one parent:

“having uncle to carry emergency rucksack, so we could take N in the backpack rather than try to take the pushchair to the beach” (7/9/E).

For many of the Children there appeared to be close ties to the wider family network. The Children’s suitcases in scenario one and two contained a variety of family members from mummy and daddy, whole family, sibling, uncle and grandparents.

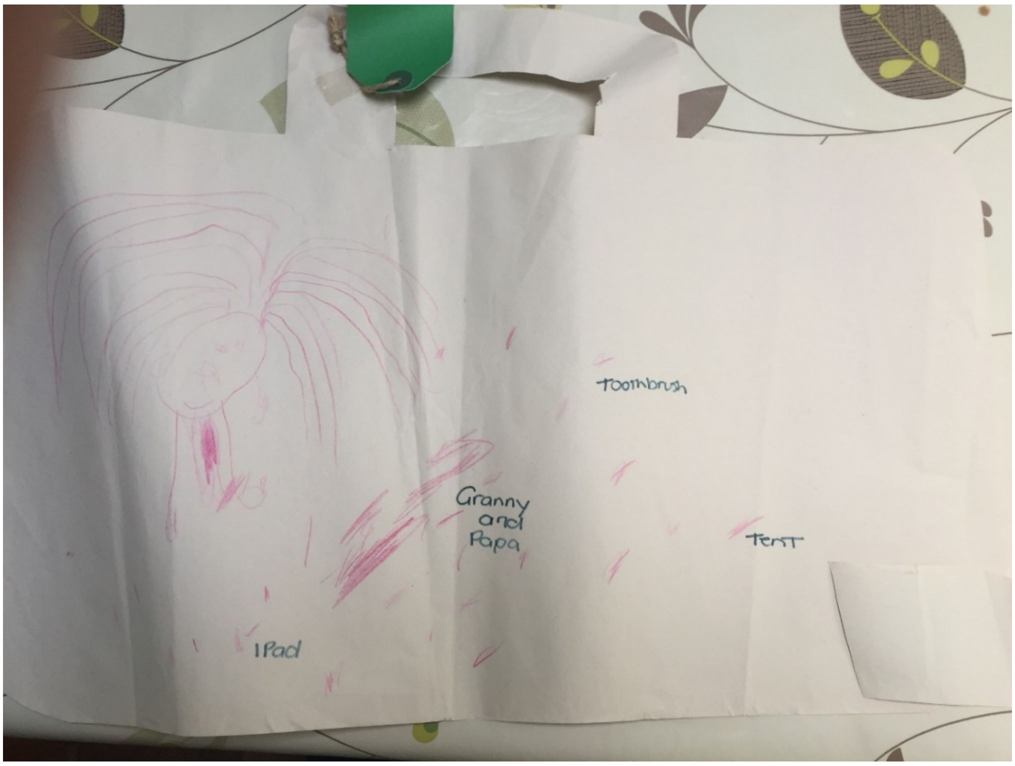


Figure 5.3 Extended Family Featured in One Child’s Suitcase Picture (reproduced with permission)

Pre-existing friends (prior to diagnosis) mainly featured in parent’s descriptions, as someone a parent went out with, while a competent and confident carer provided care. However, for many of the families there was a sense of a lack of understanding about the changes that had occurred for the Child and family within their friendship group, making them a limited option in supporting manageability in the community. This is highlighted in this interview quotation:

“the odd person doesn’t really have a clue about what life’s like and actually you know there’s been people invited, friends invited us to go youth hostelling holiday last year and you know let’s walk the west highland way with the kids and you go not really you have not figured my life out have you” (10 240 346).

However, in some families, some friends were reported to take a similar role to grandparents, in supporting the parent's Child and siblings and were seen as part of the same network of support, as the wider extended family. The diaries and interviews demonstrated the very significant role that the extended family, and to a lesser extent, friends play in enabling Children and families to adapt to the complexities and demands of the condition.

Being part of the CCHSUKSG had also facilitated strong friendships and support which is evident in both the diaries and the interviews. The group created a feeling of not being on your own and appeared to be for most participants, a vital source of support, as illustrated by the following interview quotation:

"because you do think you are all on your own and you think that no one else is, has this or is going through this and this is clearly not the case" (11 165).

For the Children, home and school friends were clearly important but so too were friends who had a shared experience of CCHS. Within the Children's playroom in scenario one and two there were well established friendships from previous meetings and new friendships that developed through the day. (S1 S2 RN1 16)

The wealth of knowledge of the CCHSUKSG was viewed as particularly important to new families and in meeting other families, to whom they did not have to explain their Children's condition. In short, they were *"in the same boat*" as demonstrated in the following diary extract:

"*Going to a group who are used to ventilated Children, don't have to explain everything*" (2/9/E).

Meeting other families with older Children and young people helped to create a sense of a possible future and helped to build hope. The active involvement in the group and helping new families or others going through a difficult time, could be a significant support to a new family's ability to manage, as revealed in the following diary entry made after the CCHSUKSG conference at which this study had been launched:

"Have a message from the FB support page saying that the new website we previewed at the social gathering in …….. is now up and running......Its brilliant and I was chuffed to see a pic of N on there. Really hoping that this will help new families realise CCHS is manageable" (13/11/E).

The link to the CCHS community via the CCHSUKSG allowed a harnessing of the group experience so "many heads" were seen to be beneficial. The group experience was also used to compare Children's symptoms, clinical care, care packages and what level of package support parents felt they needed. This could increase expectations for those with small care packages, but also challenged those who felt they needed very big care packages. The community support was also seen as helping to widen views on support packages typified in this passage:

“Discussion on Facebook tonight about respite and appears that many families get funding for three hours per week from Social work. Nothing like this has ever been raised to us - will now look into this as it would be great as N gets bigger if we could get help with afterschool care or attending clubs etc” (12/10/U).

It was felt by some participants that there was a risk with virtual discussion groups, that views could be misunderstood and cause fewer forthcoming members to withdraw. It was discussed in the interviews, that there were some incidences of negative comments on the CCHSUKSG’s social media page, but overall the participants were clear it was a positive support. As an information source, the group and its social media page achieved quick answers to questions. This learning and exchange were important in manageability. Families described how this was particularly important to them, as formal care providers were often hard to contact or slow to return calls.

5.5 Conclusion

The findings, after initial analysis and further consideration utilising Antonovsky’s (1987) three conceptual areas of meaningfulness, comprehensibility and manageability (in an adapted manner), have identified a rich account of how Children and their families adapt, learn, manage and regain control and meaning in their lives. From this account three themes emerged outlined below.

The next big Leap (adapted from *Meaningfulness Antonovsky (1987)*

This theme described the impact of the diagnosis and the initial period of intensive care in hospital, which lasted through the parent’s life. The ability to regain a focus on what was important to the family was key to adaption. The importance of Child and family centred care, which supported the ability to regain hope and a positive focus for their Child and family was striking. Hope provided the foundation for “taking the next big leap”, which encapsulated the positive drive behind striving for outcomes that were important for the Child and family, including accepting levels of risk to achieve these outcomes. Young Children were highly focused on their personal outcomes. The way care was delivered could support or impede these outcomes for Children and their families. Supporting features of effective care packages included reliability, consistency choice, flexibility a focus on supporting Child and family outcomes. Celebrating achievements was also important to Children and parents alike.

A life of learning and adaption (adapted from *Comprehensibility Antonovsky’s (1987).*

This theme describes the way children and families understood and conceptualised the CCHS, within the context of their daily lives. This understanding and confidence in initially, the parent’s ability to manage their Child’s condition, developed over time and through experience and was closely linked to the new challenges that each big leap brought about, such as starting school. The young Children who took part in the study initially saw CCHS as their parent's issue and their learning and conceptualisation was linked to their developmental stage. Learning was initiated by them wanting to achieve their next big leap. Taking an interest in their care then progressed to taking part in care procedures in a set context. Later understanding of the management of their condition included caring for their own medical needs out of context, in different and new situations. As the Child grew more independent the parents had to learn how to support the young person less directly, creating a challenge to the parent's understanding of LTV. The Children and families were focused on normalising their lives. Positive ongoing support helped them to be confident that they could cope with whatever came along.

Working together to make LTV manageable.

Working together to make life and LTV "manageable" adapted from *Manageability* Antonovsky (1987). This final theme encompassed the capacity to deliver the care each Child needed. Development affected the care needs of the Child requiring different levels and types of support at different stages. The requirement of care throughout the night made care packages very important to families, how much these care packages helped them manage was affected by the flexibility of delivery and importantly the consistency, skills and person who delivered the care. Equipment was essential to enable manageable care, providing early warning of problems and dependable respiratory support. Family or friends support at some level, were a feature of most family's resources and was integral to care, in particular the flexibility of care. The CCHSUKSG was a key support to families in providing ongoing information and emotional support and to the Children who had friendships through the group with Children who had a shared experience. Schools were an essential component of the Child's experience and were highly valued by parents, both in terms of manageability and emotional support.

All through this data set the Children and family's hopes and expectations of achieving as normal Childhood as possible were apparent. The Children with CCHS demonstrated a considerable ability to adapt and lead a normal Childhood. Parents shared that goal however, with significant additional responsibility.

The strongest theme overall was the focus on the next big leap. This theme characterised striving forward and was deeply motivating to Children and families. However, it is equally clear how complex and difficult this was to achieve. This complex mix is summed up in this final interview quote:

"I kind of feel a bit like ... you know… like a swan. It might look elegant but it's really not, its furious and its fast and its panicking and ummm stressed all underneath but yet trying to sort of look like yeah we know what we are doing this is really easy" (10 357-360).

Chapter 6. Discussion

6.1 Introduction

This final discussion chapter is the culmination of bringing the thesis together. It will draw together all the elements of the study to critically explore how the aim has been achieved However, firstly, the research aim and study questions are presented here as a reminder:

The overall aim of the study was to explore from the perspectives of Children and families what helped and enabled them to overcome the complexities that Long-term ventilation (LTV) brought into their lives. From this aim to questions were developed for the study outlined below

1. Does the salutogenic theory offer a new theoretical approach to underpin conceptualisations of the care for Children, and young people (Children) living at home with long-term ventilation support?
2. What enables Children, young people and families to overcome the complexities that home long-term ventilation brings into their lives?

6.2 Critical Discussion of the Methodology

Throughout the thesis Antonovsky's theory (1987) has been applied in order to study Long-Term Ventilated (LTV) Children. This is a novel and unique application in this field with this group. Arguably, Children with LTV requirements are some of the most medically complex individuals receiving care in the community. The predominant approaches in the literature, as set out in Chapter Two, are currently problem-focused or biomedical focused approaches. This context of complexity has made the approach taken in this research unusual, as the medical complexity of LTV treatment has led to the domination of biomedical, problem and risk-based models and the more positively focussed, health based, salutogenic approach is rarely applied. Therefore, the use of a different novel approach enabled the researcher to consider new approaches to the data collection and analysis and subsequently important new findings to be identified.

Conversely, proponents of pathogenic or traditional problem focused models however, could argue that using salutogenic theory to refocus care runs the risk of failing to recognise the technical and condition specific issues that a problem based, or pathogenic approach would uncover. This is true as the salutogenic theory is more commonly used in health promotion approaches. However, the findings of this study highlighted that a change of focus from diagnosis and treatment to seeing health as a process (as encapsulated in the approach of salutogenesis) was found in this study’s findings and seemed to work particularly well in understanding a lifelong chronic condition (this finding is similar to Cowley and Billings 1999). This study's findings identified how the Children and families were driven by their hopes and aspirations and integrated LTV into daily lives they aspired to lead; the researcher contends that once the initial diagnosis and treatment is initiated and LTV started, LTV (like any other long-term condition or complex problem) has to be adapted to and integrated into the Child and family's life and there needs to be a change of focus in assessment, support and care delivery to reflect this.

Furthermore, from the parent's perspectives in this study, also highlighted the potential for care to be entirely focused on medical outcomes, which can lead to parents feeling like 'nurses' rather than parents (Lindahl and Lindblad 2011:256, Kirk et al. 2004:460). At the same time parental findings reflected that there was a desire to lead "an ordinary life". The Researcher would argue that the findings presented in this study demonstrate the usefulness of the salutogenic approach in this context. However, pathogenic and risk factors remain important in safe and effective LTV care, but nonetheless this research is suggestive that a more integrated approach with salutogenic approaches is required in long-term home LTV care (discussed further in 7.5).

As outlined in Chapter Three,

this study utilised salutogenic theory utilised in an adapted manner. The study's qualitative approach and research questions required an ability to question, consider and challenge or develop salutogenic theory. The approach taken of using salutogenic theory within the diary questions and as a lens for the analysis required extensive thought, supervision, quality review and safeguards within the study design. Utilising theory in this manner had risks and benefits. A significant risk was that the integral nature of the usage of salutogenic theory would make its critique and development more difficult. This was extensively reflected upon (4.3, 4.4, 4.13.2, 4.13.8). The theoretical findings outlined in 6.4 demonstrate the level of eventual challenge to the salutogenic theory that evolved through the study.

Antonovsky (1987) defined the salutogenic approach as an approach that was aimed at gathering the whole story of the person. In practice, however (in later research) Antonovsky's approach has often been applied in a reductionist way (See for example Olsson, Hwang 2002). As described elsewhere, (see 3.18) Antonovsky's (1987) theory is classically applied by using the sense of coherence (SOC) questionnaire and scale (developed by Antonovsky 1987) to measure a person's SOC score (see 3.18). By taking one element of the salutogenic theory, namely the SOC questionnaire and applying this in isolation from other salutogenic concepts, it could however be argued that using a SOC Scale rather than using the concepts of comprehensibility, meaningfulness and manageability more broadly was not in keeping with the type of holistic approach required to capture the “whole story of the person” (Antonovsky 1987) or qualitative approaches per se. The decision taken in reference to the qualitative study base was that a more open methodology and data collection tool was required. This decision was considered at length (see 3.19, 3.21, 3.22) and underpinned by a further literature review of studies that had utilised this type of approach as described in 3.19.2. This approach was particularly effective in several ways. Firstly, this method enabled an open and questioning approach to be taken with the data and the salutogenic theory. Secondly, this facilitated the Child and family’s voices to be central to the study. It was hearing what worked for the participants through the Child and family voices that enabled rich and interesting findings to be identified.

The approach taken throughout this study in utilising salutogenic theory, was to use in an adapted manner, the three concepts of meaningfulness, comprehensibility and manageability described earlier (3.8) to underpin the qualitative epistemology and methodology. In turn this impacted on how the study was framed, including the phrasing of the questions, which were then used in a diary format, open interview questions and as a storyline for a participatory play-based scenario. In the diary, for example, as described elsewhere (Chapter Four), participants were asked three open questions broadly based on the concepts of salutogenic theory. Another example was the Children, as described elsewhere (4.11.4.3) where the salutogenic approach to collecting data was used in the river analogy, to explain the complex theory; this had the effect of the Children becoming actors within the scenario and this novel approach adapted a complex theoretical approach into a simple Children’s participatory play-based activity. It is however, a fair point that there could have been practical difficulties in framing understandable and useable questions on such complex constructs with this group, but the careful handling, as described in Chapter Four sought to minimise this. Thus overall, the researcher would suggest that in reality the salutogenic approach was a strong construct, but allowed for flexibility in line with the qualitative approaches.

The integration of salutogenic theory and the approach of a technique known as Appreciative Inquiry (AI) was carefully considered, as outlined in 4.4. AI describes the process by which data is gathered, to assess the way in which participants envisage their future and the unfolding of future developments. The use of AI was considered necessary to determine what Children and families would see as an ideal LTV service. The addition of AI, however, represented the addition of another approach to a study, where complex methodologies were already being applied. The decision to adopt AI may therefore be susceptible to the charge of over-complicating the study and this may be seen as a study limitation. However, the use of AI was limited and carefully explored (see 4.4) and was used to fill a clear gap identified with using salutogenic theory.

A further discussion point is that the positive focus used in the framing of questions and scenario could be argued to risk missing the true balance of the Children and families' experience (an experience which was complex and negative as well as positive). This concern was voiced, initially, by some of the families, however further quality checks including analysis of the data, revealed families found ways of expressing strongly held negative concerns. This concern was further reduced, as the literature review had demonstrated, that the existing research base, whilst rich in data around the problems Children and their families faced, was lacking robust evidence of what worked best for the Children and their families (see Chapter Two).

A particular issue with the methodology was that the number of Children and families in this study was very small, limiting the breadth of experiences that could be studied. The Children with CCHS are a unique subset of Children receiving LTV. Their cognition and physical abilities are only mildly affected. However, the majority of Children receiving LTV have more additional complex needs and disabilities (Wallis, Paton et al. 2010). This does therefore, pose issues around transferability to the wider LTV group with complex disabilities and would require further research to explore its use in the wider LTV group.

The process of keeping a salutogenic focused diary produced data in itself and is also a worthy discussion point for this chapter. Interestingly, four out of the eight families found the diaries easy to complete, whilst four found them more challenging. One participant left the study before starting the data collection, as they were facing significant adversity in their daily life and found finding positives difficult. Overall more of the respondents found the diaries interesting than an additional burden (this was particularly noticeable in those who had found doing so a challenge). The strong theme, in the group who found the exercise helpful, was that when the diaries were re-read their perspective of the time period was re-evaluated. On reflection, the participants tended to consider that the month recorded was far better than they had perceived it to be. Participants put this down to a tendency to focus on the day to day frustrations encountered in their lives, rather than what was going well. There is a growing body of evidence (Burns et al. 2007, Fredrickson and Joiner 2002, Garland et al. 2010) that contends that "positive emotions expand people's mind-sets in ways that little by little reshape who they are" (Garland et al. 2010:850). In addition, "inducing positive emotions" by asking patients about their "best times" (Burns et al. 2007:8) have meant that long-term positive diaries have been used as a way of trying to enact this change (McCullough 2003 quoted in Lambert et al. 2009:461).

The concept of finding it hard to only write down the positives for some participants was an interesting finding, as was the tendency to find it easier to identify issues and frustrations. Antonovsky (1987) linked this trait with the level of a person's sense of coherence. Those having a lower sense of coherence were more likely to report negative experiences (as outlined in 3.11). Another possibility is that the way the United Kingdom's healthcare system assesses, delivers, and evaluates care based on a risk and protect approach, predisposed respondents, when asked about their experience, to discuss what is not going well, rather than their achievements and success. In-depth consideration of this tendency is considered against the findings in (5.2.1 Paragraph 5-8).

As described elsewhere in Chapter Two, the family interviews were based on the three diary questions and participants were asked to highlight the most important issues for them from their diary entries prior to interview and from these two open questions were asked (see 4.11.3.1). In the interviews the participant's responses were used to further add to the discussion in the interview. The interviews were relaxed, as they took place in the participant's own home. This placed the interviewees at ease and enabled them to engage in frank and open discussions. The approach did bring an element of distraction from small Children and dogs, which lengthened transcription, but had little effect on final data capture. There was a risk that this extra step (of conducting the interviews) would add little to the study, but the data was significantly more detailed than the diary data and enabled a level of exploration that was not possible within the diary (thus reducing the risk of the diary methodology limiting respondent's responses).

Furthermore, the Arts-based data collection from Children was originally intended to be drawn from the picture pages in diaries and the scrap books and memento boxes. Unfortunately, this was ineffective with only two pictures being produced by the Children and no mementos being collected. The second approach used to engage the Children was in developing vignettes into a scenario-based approach, was a novel method developed for this study. This method was used to gain an understanding of the Children's perspective. This approach produced interesting data around what was important to the Children and who held the responsibility for their health. The activity risked not engaging the Children, but the use of play specialists and the careful planning of the day (and activity) achieved a high level of interest and data. No attempt was made however, to interpret pictures produced in the session. Only the Children's words and descriptions of their pictures were collected as data and incorporated into the findings, which fitted well with this study.

It is also worth discussing the use of content analysis which was utilised as a framework for the analysis undertaken during this study and fitted well with this study (4.13.1). The flexibility and content sensitivity of this method were therefore considered to be of central importance. The merit of this choice is discussed in depth in 4.13.1, 4.13.8. This method was utilised at different points and the findings of and the two sweeps of analysis undertaken, compared and contrasted to widen the analysis of the data and to consider the utility of a salutogenic approach. In the first sweep as described and critiqued in 1.13.5.1. an inductive approach was used to undertake the initial analysis of the data. The data was initially coded using a memo approach (Glaser

1978:72), and then considered through a process coding approach (Glaser 1978:72). A parent representative also independently considered the findings and commented on crossover or differences from their own experience. In this phase the analytical approach was separated from the underpinning theoretical approach, to create a data set without theoretical influence. However, in practice in a study where the methodology was intertwined with the underpinning theory, there was still a theoretical influence, as the questions and approach were rooted within an adapted salutogenic approach. The influence of theory on research has been a matter of significant debate since the inception of grounded theory (Denzin 1978:74). However, in this study real questions around how the findings worked together as a process for the participants remained. The decision was taken to explore the data further. After significant consideration and supervision, a decision was taken to conduct a second phase of analysis; in this phase the link to theory was overt and was carried out through a process of directed analysis. The findings were analysed within the context of Antonovsky's (1987) three concepts and considered the data utilising a process of inspection (Denzin 1978:74). There was a risk that categorising the results into Antonovsky's (1987) three concepts could overestimate the importance of these concepts to the Children and families. To explore this possibility, the data from the phase one conventional content analysis were compared and contrasted and considered in depth against the phase two directed analysis findings. Final categories were then decided upon.

However, a significant risk to the quality of the research in this type of analytical approach is that it could “force” rather than “create” data (Flick 2009, Mays and Pope 2000). Review of supervision notes and a detailed research diary supported the initial premise that this transition was not forced, and the initial data was still clear within the new theoretical codes based around the three concepts. To ensure credibility, trustworthiness and quality categories were subsequently explored by presenting the findings back to the participants, via a focus group, interviews, and a further Children’s participatory play-based scenario was undertaken to widen the Children data set. This allowed adult participants to consider whether they felt the findings reflected their experience, through a process of member checking and more data from the Children was produced to cross reference with the findings. During the final member check, reliability was also considered, by going through the preliminary findings of the study, to test whether the participants felt the findings linked to the data they had given. The focus group gave a group view, and this was cross referenced with three interviews with families who did not attend the focus group. This enabled both an individual and group view of the reliability of the phase two codes. The findings, whilst fundamentally identifiable as the original findings, were now split and grouped into Antonovsky’s three conceptual areas. This new focus was enlightening, as it brought together codes into stronger concepts that shed new light on the findings. Importantly this second sweep also created a set that could now be seen together as a process of adaption for the first time. Glaser cautioned about the risk of being subsumed by the great theories (Kelle 2005). However, in this study rather than being subsumed by salutogenic theory for the first time despite an extensive critique and quality review in chapter three, fundamental questions were raised within the Researcher around how salutogenic theory works as a process. The findings from phase one analysis on their own, whilst novel and interesting, did not identify the process in which adaption to a chronic condition occurred for the Children and families. To fully explore these questions required in depth consideration of the findings, in the context of the new directed analysis data. From this analysis the process of how families adapt became clear. These findings posed questions about how adaption occurred in this group of families and salutogenic theory and in particular how salutogenic theory could be used in qualitative research and in practice. This is an important discussion and critique, that is informed by the findings, so this is explored in depth in the following sections.

6.3 Critical Discussion of the Findings

Having critically discussed the methodology used in this study, the chapter will now move on to a critical debate of the study findings.

6.3.1 The Next Big Leap (adapted from Meaningfulness Antonovsky 1987)

As demonstrated in Chapter 5.2.1, meaningfulness at diagnosis for many families was pared back to its most basic level, with a struggle for life itself and hopes and expectations were reset to any life at all. In the face of this threat, meaning was often reset to the fight, initially for life and then for home. For some participants this continued into life at home where the response to every problem was to see it as a fight. This finding linked closely to the existing LTV literature (2.4.1). However, the data identified for the first time a very different perspective. Half the participants who reflected on their diary prior to the interview, noted the month had been far better than they perceived (the others had a general expectation that things would work out). They reflected that their recollection prior to reviewing the diary was of the struggles not the steps forward that they had made. There was a sense from the interview data that right back to PICU there had been a focus on problems and risks, rather than opportunities and hope. In many families this placed them at odds with the medical and care teams. Once home this continued with assessments and care delivery, which from all the respondent's viewpoint was based on problems and issues, with families conversely driven and focused upon hope, normality and opportunities, setting a scene for some for ongoing problematic relationships. This is an important and novel finding that had an ongoing effect on the ongoing perceptions of participants to the value of the care and support they received.

This sense of striving to achieve normality was conceptualised by the families as a series of leaps (described as 'the next great leap'). These leaps were highly meaningful to initially parents and later to the Children. The leaps tended to be centred around things that most families generally take for granted, such as traveling in the car or going to school. However, the participants found that undertaking everyday activities for the first time with a ventilated Child, (even these seemingly simple activities) presented a significant challenge. As Children and their families became more confident, these leaps became focused on the achievements commonly associated with Childhood, such as family holidays or swimming. These leaps were significantly more complex and often riskier for this group of Children and families (but were highly motivating). The Children were equally motivated by these leaps with three Children, during the study, changing from tracheostomy to mask ventilation, to enable them to undertake an activity they wanted to achieve. To support the Children and families in this endeavour, families described needing trust and well-developed relationships with their care teams. This finding was an important and novel finding of this study, whilst not linked to the LTV literature there are some cross overs with the wider literature which will now be considered.

There appears some crossover of these findings and the approach of family centred care. Family centred care is an approach that instead of caring for an individual Child, acknowledges that for Children the family is an interrelated and insuperable unit and therefore care needs to be focused on the whole unit, rather than just on the individual (Shields 2015). There are several key tenants of this approach, parents are the experts in their Child's care, the family is the Child's main source of support. Parents being involved in care reduces the Child's anxiety (Jonas, et al. 2014). The nurse's role is seen as recognising this and adapting care and care routines to fit the family unit. The nurse achieves this by gaining an understanding of what is important to each individual family unit. The approach of family centred care has been linked to salutogenic theory and the approach of providing "healing orientated practices and environments integrated into care (Jonas, et al. 2014). Even though questions were used that deliberately sought out what was working, it was clear that the central tenants of family centred care were often missing - that of a nurse who knows the Child and family well. Many families had access to Community Children's Nurses, but they were generally seen as not having the LTV skills required to support the family, so could not help them adapt their Child's care to the family's life. The majority of the Children had care providers, who saw their role as providing shifts, rather than providing holistic integrated care around the principles of family centred care. Indeed, these care providers were often completely separate from the local Children's services. Overall there was a critical gap, that of the named nurse working with the family, who was integrated into the local health, care and education system. The two Children and families that had an active named nurse who was part of their daily care package in the cohort expressed significant satisfaction with care delivery and described care that could be described as family centred. Interestingly, these respondents described teams who supported the family with each new leap; coming home, first holiday, starting school. At the time of the interview, in one of the families, the named nurse was supporting their discussions and planning with the Child's secondary school, which the Child was due to start the following year. The delivery of a highly complex intervention in the community such as LTV in isolation from a named nurse and strong integration and links with the wider multiagency team seems to have significant risks. Further research is required to explore the models and outcomes of the care delivery systems utilised with this group of Children and families.

Professionals, who identified any of the leaps considered as important to the Children’s families, and helped the Children and families achieve their goals, were seen as offering more effective and meaningful support. The families valued teams that celebrated their Child's achievements and understood the importance of achieving milestones of Childhood (such as swimming), by helping them balance risk with achieving these milestones. Striving for normality has been described in the LTV literature (Carnevale et al. 2006) but the concept of the next big leap and milestones of Childhood were at the time of this study not reported in the wider literature and were a novel outcome of the current study.

The Children demonstrated focus and determination to achieve their next great leap and although they largely (at the younger ages) saw their condition as their parent’s issue, if the condition became a barrier to them doing what they wanted, they became highly motivated in overcoming this perceived barrier. An example of this was moving from tracheostomy to mask ventilation as the tracheostomy precluded swimming (an activity several of the Children very much wanted to achieve). Two of the family diaries covered a time for the Children, where they had decided to move from tracheostomy ventilation to ventilation via a mask, to enable the Children to swim. The impetus for this leap however, came from the Children themselves. This focus the next big leap has some parallels with the Special Educational Needs and Disability reforms that have brought in England through the education and health care plan which is a strengths and outcomes-based approach (Department for Education and Department for Health and Social Care 2015). It also has links with family centred care as described earlier in this section. However, this finding has further implications than implementing these initiatives (discussed further in 6.5.1 and 7.1).

In discussing striving for normality and complex needs, it is important to consider perceptions and models of disability. The social model of disability emerged in the United Kingdom (UK) in the early 1970s. This model rejects the individual focus of the medical model that centres on impairment and rehabilitation; instead focusing on the barriers created by social organisation and design. (Such barriers take no account of disabled people and therefore exclude them from active involvement in society (Shakespeare 2006). The concept of “normality” was initially common to both medical and social models of disability. In the medical model the focus was on working towards medical “normality”, while the social model of disability centred upon normal social access. However, within disability groups there is an increasing focus on pride and celebration of difference (Oliver 1996) (This discourse equally challenges the role of the non-disabled in disability research, which is a further challenge for the researcher as a non-disabled, non-ventilated researcher). The social model has been critiqued, as being overly focused on the needs of the healthy wheelchair user, rather than those with more complex and acute need, where access is not the exclusive issue (Oliver 1996). The salutogenic model (Antonovsky 1987) differs from the UK social model of disability, as this clearly divides impairment and disability, rather than seeing health as a continuum, which is more akin to the Nordic model of disability which sees a continuum switching between the person and the environment (Owens 2015). This Nordic model of disability appears more closely aligned with the CCHSUKSG approach.

For the Children and their families within the CCHSUKSG, there was a clear group identity and a collective mission. Medical research and input to the group is a core part of their activity. Medical speakers form at least half of each of the group's agenda. However, the rejection of medical barriers to activities such as swimming, (identified in this study) that were seen as a milestone of Childhood and an important leap, may link to the collective group's rejection (at least in part) of the medical model's intrusion into their lives. For these Children and families, the emphasis instead was on a sense of wanting integration with wider society. The normality sought by this group of Children was in part a need for inclusion into the wider society around them and freedom from professional control. However, for this group of Children and families it went beyond control. There was a sense of activities, such as swimming, being deeply meaningful, fulfilling and creating a real drive.

The Importance of siblings was also clearly apparent across the data. Care providers who acknowledged sibling's importance and provided flexibility to meet the sibling's next big leaps (as well as those of the effected Child) were highly regarded. The lack of a clear focus on siblings in this study is a limitation and is an area which requires further focused study. This study's findings therefore highlight, that moving to a process focused on Child and family derived next big leaps would create more effective and health promoting health support than appears to have been delivered thus far to this group of Children and families. This focus on meaning and opportunities could be argued to be linked to the salutogenesis approach used in this study. If the families had been asked more openly about the problems they faced, these problems may have been more fully emphasised and may even have acted as motivators for change. However, there is an increasing body of evidence that questions problem focused frames of reference. The critique presented in the current study contends that reducing or removing problems is insufficient to understand human adaption (Sheldon and King 2001). Moreover, the literature review undertaken had already clearly identified a significant evidence base which had described the problems LTV Children and families faced (Chapter Two). The gap identified in this literature review and encapsulated within this study focused upon what worked for Children and families. Therefore, in considering the trustworthiness of the conclusions of this study the strength of the findings in identifying what works for this group of Children and families is the predominant area which was considered and critiqued.

When we consider the concepts of “hope” and “the next big leap”, one possibility is that a focus on hopeful outcomes was merely a defence mechanism in the face of the adversity (faced by Children and families) rather than a trigger to understand and manage their lives. The premise that meaning is purely a defence mechanism has been a matter of debate since the late 1940s.Viktor Frankl (2004) saw meaningfulness and hope as far more than a defence mechanism. Meaningfulness was rather the creator of the will to do and motivated people “to live and even die for the sake of [their] ideals” (Saldafia 2009:105). In the current study, a clear link was found in the data in both Child and adult participants, to demonstrate that significant effort was made in achieving meaning, not just in adversity, but in day to day life. This was represented across the findings, but was most clearly demonstrated in the next big leap. The next big leap involved significant effort and organisation to achieve. These leaps were also often linked to leaps that involve significant adaption for all families such as starting school or moving to secondary school, but with CCHS the complexity of the change was far higher. However, the boost of confidence and self-esteem achieved by the Child and family when they achieved it, in the way they had hoped, was also significantly higher.

The findings of this study appear to have links with positive psychology (Fredrickson 1998). Positive Psychology emerged at the turn of the century in response to growing concerns about the deficit approach of traditional psychology in the UK (Carr 2004). In the United States of America, Professor Seligman and a group of likeminded psychologists laid the foundation for what is now known as positive psychology. This new branch of psychology focused on the “scientific study of human strengths and happiness (Carr 2004). Three areas of study were identified, those associated with the past e.g. satisfaction, the present i.e. gratifications, (i.e. sense based) and finally the future such as optimism and hope (Seligman 2002). Having hope was a clearer motivator for change in this group of participants. Participants were motivated to take on the challenges of their Child’s condition to go home and get on and live. Children were motivated to take over aspects of the management of their condition for the first time. Positive Psychology has been linked by some to the broad concept of Salutogenesis (Mittelmark et al. 2017). However, there were also wider factors identified by participants, that went further than the personal attributes identified within positive psychology (considered the in following themes below). Moreover, an interesting feature of the findings was that despite the positively orientated questions used in the diary, interviews, and focus groups, set within a positive approach, there was also a problem-based discourse apparent. On further investigation it was clear that some participants had used inverse positives or interview reflections to share negative data or had identified issues that, although appearing negative, had induced a positive effect. An example of this was the “fight” where families identified battles (such as gaining rehousing or gaining the funding for a care package) as leading to positive effects. Such battles provided significant motivation where they were defined by a clear beginning and end. These fights were clearly meaningful and motivating. This part of the finding was apparent in the review (2.4.1-2.4.3). However, it was also apparent in the data that once home the effect of this fighting to resolve problems became less positive as the number of issues often become overwhelming. This was a new finding which has particular relevance to the care provided to LTV Children and families in the community. The interviews identified that these issues could become fixated upon leading to a loss of a wider focus on what the Child and family had achieved. This was identified from some of the self-reflections from participants in the interviews when they reread the diaries to highlight what was most important to them. They themselves identified their perception of the month had changed with them seeing it much more positively than they had initially perceived

One discussion point is that there was a tendency, that without positive questions, some participants moved to a problem-based discourse. This was noted in the focus groups and was an interesting finding, meriting further discussion. A psychological approach to this tendency connects this precedence of negative emotions with the way that negative discourse often reflects a clear danger and therefore need to trigger action. Such responses are therefore psychologically prioritised over the more long-term nature of positive adaptation (Seligman and Csikszentmihalyi 2000). Conversely Antonovsky (1987) identified the focus on a problem-based discourse as being linked to a person's sense of coherence. In the diaries, half but not all, participants noted the positive influence of the diaries on their contextualisation of the month covered in the diary. Antonovsky's theory would suggest that this is due to some of the parent's innate sense of coherence. Parents, with a lower SOC, would recall the month to be full of struggles and adversity, but when reflected on through the diary would see it far more positively. Those with a high SOC would not note any particular change to their perceptions of the past month. It would appear they simply put the difficult experience behind them or actively sought out the positives in the situation. One participant explicitly noted that her family described themselves as positive thinkers and therefore found the approach sought within the diary questions reflected her normal approach to life.

This phenomenon however was more apparent in the diaries, but was less obvious in the focus groups (where the precedence of negative events was more apparent) (Seligman and Csikszentmihalyi 2000). This finding of a solution focused discourse therefore needed to be assessed for trustworthiness. Such diary findings were tested through questions and focus group discussion. The interviews and focus groups supported the findings from the diaries. The researcher thus concluded that the findings had gained useful and relevant data to answer the research question, whilst maintaining trustworthiness.

Overall using the construct of meaningfulness, developed into the next big leap, offered a new understanding of how families adapt and overcome their Child's CCHS. It also identified a wide gap between the way professionals treated and supported families and what support the families themselves identified as most effective. The findings from this study identified findings of what worked for families and Children but also identified a way of reframing negative issues to capture solutions.

6.3.2 A Life of Learning and Adaption (adapted from Comprehensibility Antonovsky's (1987)

As described in 5.3, comprehensibility for this group of Children and families was strongly linked to their underpinning understanding of their Child's CCHS and its management. Most importantly however, comprehensibility considered how this understanding was contextualised within the Children and families' day to day life. Antonovsky (1987:140) saw this as the ability to define the nature and scope of the problem (including its duration) and to identify the likely impact and most useful resources to bring to bear. The participants described that learning through life experience was a key part of the process of adaption. For parents in this study, how they conceptualised CCHS changed with each move towards independence. Once a Child could move around independently the whole process of supervision, in case of airway or condition related issues, had to be rethought and then integrated into how they coped day to day. In this way parent's conceptualisation of how they managed CCHS was continually challenged by the Child's development and the journey of Childhood, in particular through the education system. As the Child's understanding and physical abilities changed so too did their wider comprehension. The young Children who took part in this study initially seemed to contextualise CCHS as their parent's condition. The start of separation from caregiver enabling self-expression and self-control usually start around two years of age (Carter et al. 2005). For the young Children, in this study, it appeared that separation in terms of their CCHS occurred much later. There are no LTV studies in this area, but studies of self-care in chronic illness have noted the link with self-care and Child development (Macorby 1984). The complexity of the Child's care and the protection given by parents is one potential explanation of this phenomenon. In research into cystic fibrosis (CF) it has been noted 47% of parents reported overprotectiveness (McCollum and Gibson 1970) and that Children with CF exhibited greater dependency on others (Droter 1978). There is also research that identifies that Children are often excluded from discussions around their condition (Angst and Deatrick 1996). There is moreover research in this area around types of education approaches and their efficacy (Bluebond-Langne, Angst, and Lask 2001). However, the current study identified that Children up to the age of ten, tended not to consider their CCHS if they were focused on an activity. This is an important new and novel finding. When asked, the Children very clearly saw the management of their CCHS at this point as their parent's issue. This was exemplified when the Children during the play scenarios explained that taking their parents with them on a trip meant they had all their medical equipment with them. Interestingly this did not appear to be because they did not know what to do. This was exemplified in scenario two, when prompted by the diver's discussion of sleeping and breathing under water, the Children described in great detail what they needed to breath at night (indeed some of the parents of the Children through the diaries and interviews had identified the Children could and did do their own basic care independently).This important and novel finding of this study, has implications for the understanding of Children's self-care in complex conditions particularly in different contexts and warrants further study.

A life of learning and adaption was also described in some of the diaries and interviews by parents as important for siblings. Rather than finding having a brother or sister with LTV a burden, (as described in much of the literature relating to siblings of LTV Children (Lindahl and Lindblad 2011) there was a wider perspective taken by participants in this study. Respondents reported that gaining an understanding of their sibling's condition and being able to get involved in all aspects of the family's daily life, including the affected Child's care, was helpful for the sibling and the affected Child and family. In the play room activities, it was noted that several siblings took an active role in the care of their affected brother or sister and had the necessary skills to do so. However, there were also concerns expressed by parents about the wider effects on siblings. This finding partially disputes the current research base, which identifies that having a brother or sister with LTV can create issues for siblings around loss of attention, restrictions on siblings, increased domestic responsibilities (Lindahl and Lindblad 2011:253). In addition, in other studies wider concerns were raised, including the effect on the sibling's schooling and the concept of siblings taking on more care related responsibilities as they matured, is described in Heaton et al. (2005:447). There have been almost no positive aspects of sibling care identified in the existing LTV literature at the time of this study. However, the diaries in the current study identified the importance of sibling care and the interviews noted how it allowed siblings time alone together. One adult sibling at the final focus group supported the value of sibling involvement in care as being essential for him to feel part of what was a major aspect of the family's life. He went on to describe that being excluded from training to safely care for a sibling effectively would have excluded them spending time together on their own. This was also supported by parents in two of the phase one family interviews. The data gathered in this study appears to suggest that siblings were far more positive about their situation, provided they were trained, included, and allowed to become involved with their brother or sister's care. This study therefore offers a separate explanation that isolation and distorted sibling relationships may result from not being trained in an affected sibling's care needs. This finding, although potentially important, should be treated with some caution however, as it was gained from small amounts of data from two siblings and a parental perspective from two family interviews. This therefore is an area that requires further study from the perspective of a larger group of siblings themselves.

For the extended family and friendship network that supported each family, to enable their support to be effective the data suggests they needed to understand CCHS at varying levels, from awareness to full training, depending on their involvement and role. When this was achieved, the families' normal patterns of family and friend support returned, greatly supporting the Child and the families' quality of life. This informal trained care was far more flexible and meaningful than even the best formal care packages, however it enhanced formal care rather than replacing it. This concept of integrating the Child's needs within the family's life was evidenced in Lindahl and Lindblad (2011:257) and is apparent in the wider disability literature.

The themes of learning through experience, growing up with CCHS and going to school with CCHS, described the initial process and ongoing contextualised learning and understanding of CCHS that the families and then Child went through. These themes also described the changing needs arising from Child development (as the Child grew, developed independence, and went through Childhood steps such as starting school). These areas were significant concepts in this study, which suggested that at each developmental stage new issues arose; many of which were complex to resolve due to the Child's CCHS. These issues were routed in the family context and support to overcome them was only of use when this family context was fully acknowledged. Neither on-going adaption nor Child development is noted in the LTV literature. The issues noted in the literature are more frequently around difficulties in accessing training and the problems LTV creates, rather than wider considerations of adaption, Child development and the process and stages of growing up. There were examples in the literature of Child development being considered at a fixed point to add context (Noyes et al. 1999:445), but not as a key area for focus throughout the duration of a care package.

This lack of focus on ongoing adjustment and Child development in such a complex condition is markedly different to the approaches commonly taken in other complex conditions such as Diabetes, CF and heart abnormalities, where the provision of long-term condition management support has been available for many years, either through long-term projects or on-going specialist nurse support. In this context, many specialist nurse approaches provide consistent support throughout the early days and weeks at home and throughout Childhood and adolescence within the home environment (Kirk et al. 2010, Llahana et al. 2001, RCN 2006*)*. Where long-term condition support was available to the participants in the current study however, it was hospital based and normally accessed via clinics. This was checked with participants in the focus groups and interviews and was supported by participants as an accurate reflection of their experience. This is a new and original finding which has the potential to be used to rethink the current conceptualisation of care packages in Children's LTV considered in 7.1. The data identified starting school as a particular area where the participants had little or no support, with parents being relied on to look after their Child in school or to train school teaching assistants in how to manage their Child's condition. School has been identified as a key issue in the wider literature around Children with complex conditions requiring support of self-care skills to enable Children to fully access school activities and opportunities (Kirk et al. 2010).

The findings in the study also identified “condition related learning and adaption”. The parents described for example how the CCHSUKSG had supported them when their Child had made the decision to move on from tracheostomy to mask ventilation. The group supported the Child and family by talking through their own experiences and sending short video clips of their Child putting on a mask. This wider understanding of how and why families learn about their Child’s LTV not in a one-off way, but as a lifelong journey effected by physical, emotional, and social development is a unique finding of this study. This could be attributed to the focus that using Antonovsky’s concept of comprehensibility brought to the analysis. The implications on practice of these findings are considered in 7.2.

The parents described their care as a care package focused on shifts. Most of the parents described little or no contact with nurses in their day to day care. Care consisted of allocated shifts with carers often inconsistently or generically trained to support ventilated Children with little or no understanding of the individual Child’s underlying condition (see Coad et al. (2013) for a replication of this finding). Participants therefore identified a need for a coordinated long-term condition management approach for their Child and family. Child specific training of carers within the family’s own home was seen as Child and family centred, contextually relevant and essential for enabling Children to be Children and families to live as normal a life as possible. Children and their parents who had consistent carers, who had been trained in this way described their care team much more positively than generically trained carers. Parents who had access to this ongoing LTV and CCHS support (more frequently through the CCHSUKSG than via the formal care provided to them) reported increased confidence and independence with grandparents, relatives and friends picking up aspects of the care, that formal carers had previously undertaken. The focus on a care package and shifts rather than a long-term condition management approach appears, from this small sample, to increase dependence (and with it conflict with care providers and commissioners), by not supporting families to learn and adapt as their child grew and developed. This is an important and new finding in LTV research. However, further studies would be required to confirm this emerging finding by taking a more focused longitudinal approach in a more in-depth study of LTV care provision.

The extended family, friends and indeed community care for the families in this group seemed to be given little consideration by care package providers or clinic-based staff. The families’ care packages consisted of care largely devoid of any long-term condition management, training or support being offered to the extended family or to the wider friendship group which is similar to the findings of (Margolan, Fraser and Lenton 2004) some 13 years prior to this study. The approach of building on family’s strengths and social networks has a substantial evidence base in the wider Children’s disability literature (Judge 1998). For the families in this study, education in long-term condition management was professionally delivered in clinics and informally via the CCHSUKSG. This group was important in understanding the Child's condition in the context of their daily lives and their Child's on-going development. This finding is an addition to the current LTV literature, which reported surprisingly infrequently on the benefits of peer support via support groups.

The possibility was explored, that the Child and family's understanding of their care was "polarised", between the outpatient care they received in hospital and their care package at home. Care for Children requiring LTV was, at the time of writing, funded differently to most other NHS care. The large care packages required to manage Children's LTV at home necessitated bespoke care packages, which were not available within existing NHS contracted provision, therefore they were separately assessed and funded by the NHS. Hospital acute services offered initial care and ongoing clinic-based care. Indeed, most of the families had some sort of specialist nurse support at the acute centre. Therefore, it was considered important to determine if the initial adaption to life at home with CCHS, child development or issues occurring around going to school were areas covered at clinic. It was also appropriate to consider the possibility that the participants' perception of their care (as presented in diaries, interviews, and focus groups) was solely focused on the aspects in the care package they received at home (rather than also including clinic activity). The final focus group and interviews explored this possibility and it was articulated by the parents, that the clinic-based education received by families met some of their needs, but what was missing was a focus on learning and adaption in its broadest sense throughout the developing Child and family's individual journey, within the time and context of where the issues were arising at home.

6.3.3 Working Together to Make Life and Long-Term Ventilation Manageable adapted from Manageability Antonovsky (1987)

The study's findings as set out in 5.4 identified that families' and Children's ability to manage a condition which requires 24-hour skilled care and supervision had a number of different, complex components.

Firstly, discharge was made easier when there was a planned family focused approach, including planning around dates that were important to families rather than professionals. This gave milestones for families to focus on during what was often a protracted discharge process (Children were all diagnosed at birth or within their first year so had no recollection of this time). It was considered if this finding had come about as the milestones the families were describing were memorable events and therefore remembered more easily on recollection, than professionally derived dates based on condition, training, or community readiness for discharge parameters. However, in the focus groups and final interviews, it was clear that these family derived milestones were linked to hope and were therefore deeply meaningful as described above (6.3.1). The need for planning is clear in the existing literature (2.4.1); however, the importance of planning for family milestones is a new finding. This finding is small but new and suggests a small but important step towards making discharge more personalised. However further research is required into this area.

The often-protracted discharge process families had experienced presented significant difficulties for families which were hard to manage, in particular when there were siblings who had their own needs. For many, hospital care was provided far from home, making the logistical and financial issues particularly difficult. These problems were already clear in the existing research identified in the Literature review (2.4.1) however, the importance of a named nurse providing in-reach from the community provider taking over care was a new finding. Such a process was becoming increasingly difficult to enable under the care package commissioning models present in particular in England (but was also noted to be missing in the family experience in other part of the UK). The majority of families in this study had packages of shifts rather than complete packages of care. The approach of in-reach is linked to case management approaches soon after diagnosis and was at the time of this study declining. This finding comes from two parents and therefore requires further study, into its trustworthiness and transferability and may prove to be an area worthy of further study to gain a full understanding of the potential value of this approach.

Care packages which aided manageability at home were those that were sustainable and flexible. Inflexible care made the family's life difficult to manage and limited the family's ability to lead as normal as life as possible. This is in keeping with existing LTV research see (2.4.2 - 2.4.7). The families recognised the importance of sustainable care and the size of care team required to provide it, but felt that numbers of carers and rotation of staff went beyond sustainability and was more for organisational reasons (and was not compatible with the family centred care they required for their Children).

The data identified a number of concerns around this rotation. The first concern was that a Child may struggle to communicate with a changing care team due to their tracheostomy, making speech problematic. Alternatively, a Child may just be nervous of these strangers, who were present in their room overnight. This finding came from the diaries and was corroborated in the interviews and focus groups, where parents were concerned about a Child being scared or unable to communicate. This fear made overnight care of limited value (in terms of manageability), as the parents were too concerned to sleep. This is a new and potentially important finding around the effect on Children of a lack of consistency in the care team. For the Children and families in this study, a well-known and trusted carer had a significant impact on their confidence and the Child's contentment, making care by this group of well-known and trusted carers of particular value to the Children and families. This is an important and novel finding. This finding may have wider utility with the more disabled wider LTV group, where limited communication is a common feature. There was at the time of this study, no research around Children's care providers perspectives to gain an understanding of how they view rotation of staff in terms of service delivery but most importantly the Child's communication needs. This is an important and novel outcome of this study and is important to consider in practice. This finding requires more research to move practice on more widely. This is an area which would seem to warrant further study.

For the families and Children studied, there was a clear developmental component in living with and adapting to LTV. For example, risk decreased as Children could vocalise their needs to those around them. As Children developed augmented methods of communication (or they chose to change from tracheostomy to mask ventilation), they could easily verbalise their needs. However, with each increase in independence came new concerns for parents as they moved from direct care givers to overseers of care. For example, a Child's request to join a group, such as Scouts, where direct parental supervision was not practical, was perceived by some parents as worrying. Managing these continually changing needs challenged the manner in which parents and Children developed to manage CCHS in their day to day lives. This finding links with the personalisation of care approach which was a predominant policy driver at the time of this study within the NHS (NHS England 2018), but which was largely absent from the family's experience of the care provided in all but three families. Most importantly however, this process of changing concerns and needs demonstrates the ongoing difficulties parents face as their Child grows and develops. This theme of working together to make LTV manageable has hitherto not been addressed in the wider LTV literature. A longitudinal study is required to identify fully the impact of Child development on the care of LTV Children.

The literature review highlighted that there were no LTV studies in Children which had used a salutogenic approach. Indeed, in most research in LTV was heavily weighted towards what was not working in health care delivery, and effectiveness of care packages. This approach was equally apparent in quantitative and qualitative studies, which have both focused largely on researching the workload and problems that LTV brings into parents and children's lives. This it appears, has resulted in a research evidence base with a problem-oriented focus on the delivery of care packages and the burden of care.

6.4 Overall Consideration of the Theoretical Approach Taken in Reference to the Research Questions

To recap, question two asked 'Does the salutogenic theory offer a new theoretical approach to underpin conceptualisations of the care, for Children and young people living at home with long-term ventilation support?' The salutogenic theory (Antonovsky 1987), considered in this study set a foundation for the creation of a novel methodology which delivered new understanding, insight and understanding of the experience of Children and their families on how they successfully integrate LTV in to their lives. This approach also identified significant differences between the approach taken in Children's LTV by healthcare professionals and what families found helped them get on and live their lives. These differences are considered in depth above. The differences identified the importance of professional support being aligned with the way in which Children and families adapt to life with LTV. Thus, the researcher would contend salutogenic theory has been useful in understanding the conceptualisations of Children and their families living with LTV.

Throughout this study, the researcher has been mindful of the study's qualitative base. The challenge of using a theory as a base in qualitative research was carefully considered and challenged at each step. Interestingly this critical reflection in reference to the findings led to a challenge and remodelling of the salutogenic theory itself. This is how this evolved; the original salutogenic theory published by Antonovsky (1979) identified the concepts of comprehensibility and to a lesser extent manageability. Antonovsky later identified meaningfulness as a third and most important component and published this in his second book on salutogenic theory (Antonovsky 1987). He theorised that this process started by a person making sense of their world in terms of their understanding, practical management and most importantly how it fitted into their lives. Through this process they would be enabled to manage and would find meaning in their life creating a vital strength and ultimately meaningfulness.

Figure 6.1   Antonovsky's Ordering of his Three Concepts

Utilising this theory within this study, it became clear from the data (through the process of directed analysis) that the sequence of the three concepts was important to understanding the Child and family's experience. However, the sequence was different to the order originally identified by Antonovsky (1987). This evolved through the analysis of the family data and interviews. This is a unique, novel aspect of this study: how this emerged was that the families wrote and talked extensively about meaningfulness. The data on meaningfulness was not just of a greater quantity; it was fundamentally richer and appeared to be of much greater significance to them and was a key to how they managed life with CCHS. Further comparison between the conventional and directed data (Figure 4.14), identified the importance of codes linked to meaningfulness where there appeared to be a clear sequential process. This process, started by families regaining a focus on something meaningful to them, in the new situation that they found themselves in. Families were driven by this new-found meaningfulness to go on and achieve comprehension of how to care for their Child's condition and fit LTV into their families' lives and make it manageable. As the Child became older, the diaries and Child's data specifically suggested it was not a quest for comprehension of the condition that started the process (as suggested by Antonovsky 1987) of them beginning to take on ownership and management of their condition. Rather it was when they found an outcome they wished to achieve, and to accomplish this outcome required them to take control of an aspect of their condition. Only then did they gain the drive to comprehend and manage this aspect of their LTV. This process applied equally to siblings with the meaningfulness of independent time with their sibling, or a desire to be involved with a key aspect of family activity created a quest for understanding how to manage their brother or sister's condition. This order is represented below as a process in the order the Children and families described.

The process of adaption followed by Children and families as 'identified' in this study as following a cycle started by a quest for Meaningfulness followed by » » Comprehensibility » » Manageability. This then continued at home, as an ongoing cycle, within the Children and families lives as depicted below.

Figure 6.2 Reordering of Concepts to Match Child and Families' Experience

The approach of professionals, identified by this group of families, was that they were focused on identifying, treating and trying to solve problems. In addition, they provided education to help families understand the Child's condition and its technical management within hospital, prior to discharge. Care packages were utilised to enable parents to manage at home, with limited ongoing professional support being delivered through periodic clinic visits. Home care packages were focused on shifts and delivering the support the professionals' assessments had determined was required to reduce the burden and maintain a Child with CCHS safely at home represented below.

The approach of professionals up to discharge was:

Assessment and treatment of problems » teaching and helping families understand condition » care provision. Once home, Education dropped off to add hoc clinic support and the focus was identified as:

Assessment and treatment of problems »provision of care package represented below:

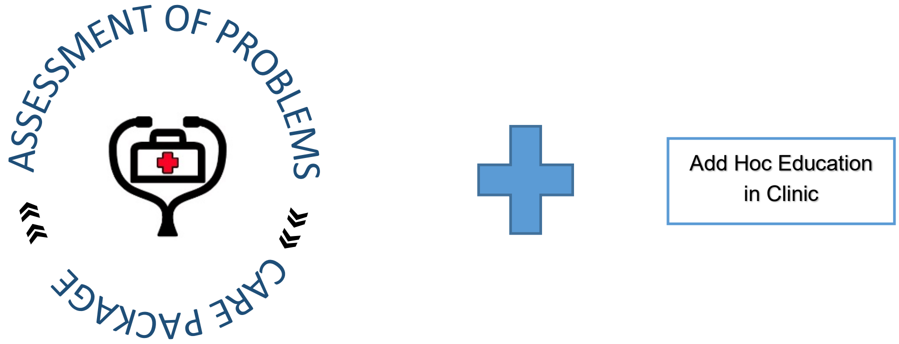


Figure 6.3 Professionals Approach to LTV in the Community

This study therefore, has identified the need to modify salutogenic theory to use it in qualitative research and practice development. The lack of challenge around the underpinning salutogenic theory as described in 3.6 appears from the literature largely to have come about due to the rapid rise to popularity of the Sense of Coherence Scale (SOCS), as described in 3.19. The SOCS uses questions from each of the three domains of comprehensibility, manageability and meaningfulness in a mixed questionnaire making the ordering of the component part of little significance in its use. The SOCS has been studied and critiqued extensively (Erickson and Lindstrom 2005). The qualitative literature utilising the wider underpinning salutogenic theory is much smaller (see 3.21) and contains no challenge of the theory, or the order of the theory as outlined above. Antonovsky did not reorder the components in his description of them, or consider the theoretical application required to reflect the conceptual precedence of meaningfulness, which Antonovsky (1987) had himself identified. However, this study has found the ordering is important, as it is the importance of a meaningful aim (the next big leap) which motivated the families to learn about and manage their Child's LTV.

The development of salutogenic theory, in particular its ordering, both arose from and helped consider the data in a way that has identified important new findings. The most important of which was a fundamental mismatch in what services saw as the process of adaption to LTV and strove to provide, compared to what was important to how families and Children adapted to a long-term condition, therefore, services and resources were not as effectively organised and targeted as they could be. Professional practice and the majority of the LTV research at the time of this study was based on a pathogenic or problem focused approach of identifying and solving problems under pinned by teaching Children and families and providing care packages to help families manage the Child's condition (however this teaching largely ended at discharge except for occasional clinic interventions). Meaningfulness was seen by professionals as something families achieve when problems were resolved, knowledge imparted, and the family have a manageable burden. Children's services provide care for what for most, if not all parents are the most important part of their lives, their Children. It could therefore be argued that meaningfulness is implicit in Children's health care. The very fact that a parent's Child is being given chance to be healthy, or even to live, is by their definition meaningful. However, the findings from this study suggest that meaningfulness is not the outcome, it is the drive which motivates families to comprehend and manage and as such, meaningfulness is vital to start adaption. LTV like the majority of the NHS's work is about a long-term chronic condition. Recovery is not cure. Recovery is the start of a very different life for the Child and family. The focus in the current study on discovering what is important to families and Children, found their hopes and aspirations appeared to motivate families and Children to take on the complexities they faced, to achieve the best life for the Child and family. This approach equally has the potential to inspire professional staff to meet their Child and families' aspirations, rather than a more traditional problem focused discourse.

To critique this finding, diary notes from the launch and parental reflections in the interviews were considered. At the launch and in the interviews, respondents had identified that the third question "things that made life/care clearer or easier to understand (comprehensibility) was the most difficult question to answer as such things did not occur every day. This questioned the trustworthiness of an argument around quantity of responses, as it had already been ascertained that comprehensibility was unlikely to have occurred frequently. The questions were reordered in the diary to reflect this feedback, which could have prioritised meaningfulness, as the first daily question the participants answered. However, all three questions were very brief and fitted on one page making them all immediately visible to participants enabling parallel, rather than sequential answering of the questions. The advisability of a reordering of meaningfulness, comprehensibility and manageability was borne out in the diaries and interviews, as the data on meaningfulness was not only more plentiful it was also the richest. The qualitative analysis undertaken in this study identified the importance of both adults and Children finding a focus that was important to them to start the adaption process. The quality and trustworthiness of this (and the other findings) was tested in a final focus group, followed up by interview at home. The ordering was supported in both the focus group and the home interviews. The Children's participatory play-based scenarios provided additional evidence of the precedence and importance of the concept of meaningfulness. The participatory play-based scenario data was dominated by data around meaningfulness and the drive it created, conversely less was identified around comprehensibility and manageability.

This final section in reference to the research aim and questions has brought together the overall aim, research questions and findings. This study has identified new and novel findings which bring together a rich set of findings that set out what works for Children and families in their day to day lives. This has in turn identified the usefulness of salutogenic theory in understanding family conceptualisations of life and care with LTV. However, this study has also raised important questions about salutogenic theory and how it works which have particular relevance to its use in qualitative research and practice situations.

The modelling of how Antonovsky conceptualised salutogenic theory working and how the data suggests this process of adaption works was of particular significance. The new modelling informed by the data as outlined above, when used to compare how families adapt to LTV compared to how professionals deliver care and support, was fascinating. This comparison led to a new understanding of where the family and professional approaches were poorly aligned and often contradictory to each other. The adaptation of the theory is an additional and welcomed outcome of this study.

6.5 Conclusion

This study opened by identifying the emergence of the research question, from the Researchers observations of practice in Long-term ventilation (LTV) home care packages and the literature around this area of practice (see Chapter One). From this emerged question two as outlined below:

What enables Children, young people and families to overcome the complexities that home long-term ventilation brings into their lives?

Despite a significant literature base (considered in Chapter Two) around LTV home care, identifying the problems and issues, these Children and their families faced. Very little had been written at the time of this study around what worked in practice for this group of Children and families. To answer this question therefore required a way of looking at this area of practice anew.

The introduction goes on to detail the Researcher's previous academic and practice related links with the salutogenic theory, which this study has utilised to identify and consider question one; Does the salutogenic theory offer a new theoretical approach to underpin conceptualisations of the care, for Children, and young people (Children) living at home with long-term ventilation (LTV) support?

The salutogenic theory (Antonovsky 1987) considered in this study, set a foundation for the creation of a novel methodology, which delivered new understanding and insight of the experience of Children and their families, around how they successfully integrate LTV in to their lives. This approach also identified significant differences between the approach taken in Children's LTV by healthcare professionals and what families found helped them get on and live their lives. These differences are considered in depth in 6.5.1. The differences identified the importance of professional support being aligned with the way in which Children and families adapt to life with LTV. Thus, the Researcher would contend salutogenic theory has been central in understanding the conceptualisations of Children and their families living with LTV, however it requires adaption to be effective for children and families in practice.

Chapter 7. Recommendations

This chapter draws the study to conclusion, by moving the findings and discussion on to the recommendations, in reference to the research questions.

The following chapter aims to ground the recommendations in the three overarching themes, the next big leap, a life of learning and adaption, and working together to make LTV more manageable, by moving the themes on to application. Overall these findings encapsulated in the three themes, if adapted could move service provision much closer to the expectations of Children and families. These changes would centre health support on strengthening each family's resources, by helping them to develop personal, family and community assets. This approach as well as health promoting would make care significantly more personalised.

7.1 Recommendations for Practice

Care should be focused on Child and family derived aims. This study is suggestive that meaningful self-selected aims (next big leap) motivated Children and families alike to take control of the Child's LTV, within the daily context of their lives. To foster this potential for family development, an asset-based approach needs to be central to care provision. Assessment approaches need to be changed to capture family's aims (see Appendix 12 for an example). Service delivery should be adapted to build family resources to achieve these aims, by supporting skill development and practical resourcefulness. Care packages need to support family aims and be commensurate with the family's objectives.

A clinical nurse management approach. This approach of "working together to make LTV more manageable" should start with early in-reach into acute care services of a named nurse, who will manage the Child's discharge from acute care. This should be achieved by undertaking a strength-based assessment and analysis of the outcomes the family's wish to achieve. From this assessment the nurse and family should work together to create a personalised and integrated multiagency provision around the Child. The clinical nurse management approach must then continue throughout the Child's Childhood. The role encompasses clinically and strategically supporting the Child and family through the daily challenges they face, by organising care around the family's aims and objectives, which this study identified were often the "next big leaps" of childhood including, first trips out, holidays, life events, starting school, through to transfer to adult services. With each new leap or challenge the children and families, supported by easy access to nursing support, would be enabled to utilise and adapt both their informal support networks and their formal care provision to achieve the aims that are important to them.

Long-term condition management nursing support. This is a well-researched and widely accepted component of complex care for most Children with complex chronic conditions. The exclusion of Children with some of the most complex of care needs (those with LTV) from this support, is at odds with current research and recognised good practice. This study has identified that long-term condition support would be most effective in assisting children and families through "a life of learning and development" when it is embedded in the day to day care of Children with CCHS. As the Children grow, develop and face new contexts, challenges and experiences the way the Children and families comprehend and manage LTV, requires continual adaption, skilled advice and training. This is important to Children and families alike. Such change could promote independence and therefore improve Children and families' quality of life, by helping them develop their capabilities, strengths and resources.

Family centred care. This is a well-recognised component of effective Children's care and is of particular importance in community care. The merging of clinical management and a long-term condition management role (which have significant cross over), into one nurse role would combine to provide a foundation of family centred care, enabling easy access to support of a nurse the family knows well, within the context of the Child and family's daily lives. Families and Children, as the Child develops, could be supported and empowered to achieve the outcomes that are important to them. The inclusion of siblings, other relatives and significant others is implicit in such an approach, and with training and support could enable families to use traditional family and friend-based support, to help achieve the normality they strive for as a family. This approach could also be provided where a parent or young person chooses a personal health budget by clinically supporting its development and the training of the staff within it.

Children and families need to be supported within the local multiagency Children's services. The additional support up to and beyond what existing universal and specialist Children's services can provide, needs to be carefully commissioned and integrated into existing Children's services. This type of integration enables multiagency support to be seamlessly provided around the Child and family. In personal budgets, this would allow training and support of family employed staff to still be provided and the links with local multiagency Children's services maintained. This level of integration would enable early recognition of areas, that need planning and additional support, such as starting school, sibling support and other Child and family derived aims.

Consistent Staff trained specifically to understand each Child, their Congenital Central Hypoventilation Syndrome and associated individual care needs. Children and their parents need confidence and trust in those caring for them, this includes Child specific competence and confidence in all their clinical care needs. In addition, staff need to understand the Child's wider preferences and needs, including communication and other key areas by knowing the Child and family well.

Care delivery needs to be sustainable, consistent and flexible. To have the greatest benefit for Children and families and to achieve the above objectives, teams and organisations need to be flexible and adaptable. Care provision, if not flexible and responsive, can provide a significant barrier to Children and family's achieving their aims. Employing institutions need to recognise the uniqueness of this type of care delivery and be flexible in application of organisational approaches to teams striving to deliver this very different type of care.

Personal health budgets should be encouraged. Personal health budgets should be utilised for delivering personalised care by family employed staff, supported and trained by long term condition nurses. The current lack of clinical support for families choosing this option is leading to a low take up of a personalised service, that this study identified many families covet.

Children's trained staff should be used in the care of Children. To provide effective meaningful care for Children and their families, such care must be sensitive to each Child's complex needs and their on-going development. Equally important is an understanding and sensitivity to the complexities of family centred care (this was a recommendation in 'Learning from Bristol' (Kennedy 2001) and the 'Quality Standard for Services providing Long-Term Ventilation for Children and Young People' (West Midlands Quality Review Service 2015) and should be implemented.

7.2 Recommendations for Theory Development

In keeping with qualitive perspectives, theory development was an important outcome of this study. Utilising a pre-existing theory was a risk in this study, but the process of using the data to consider findings, but also challenge the underpinning theoretical model has enabled important theory development to take place through the study and theoretical recommendations to be made.

This study questions the current ordering of the concepts of Antonovsky's theory (1987) in the qualitative study of Children and their families and in their care. The study purports a new ordering of meaningfulness, comprehensibility and manageability. This study argues that establishing meaningfulness for families is essential for them to be engaged and to gain comprehensibility of their Child's care needs and ultimately the ability to manage the care their Child needs.

LTV care is largely, due to its complexity and risk, rooted in a problem-based model. The study has explored a salutogenic theory model to identify what Children and families identify as important. The study suggests that the salutogenic model has the potential to reroute care into a more effective and health promoting approach. This study does not suggest that biomedical models have nothing to offer this group of Children and families. However, the study does suggest that once a diagnosis has been made and LTV has commenced, care can be refocused to a salutogenic emphasis on assessment and ongoing support though the lifespan. However biomedical care will always remain part of the mix of support that Children and families require particularly at times of acute illness. Becker (2010) has produced a model of health that uses both biomedical and salutogenic models and this provides a possible framework for developing more holistic models of care.

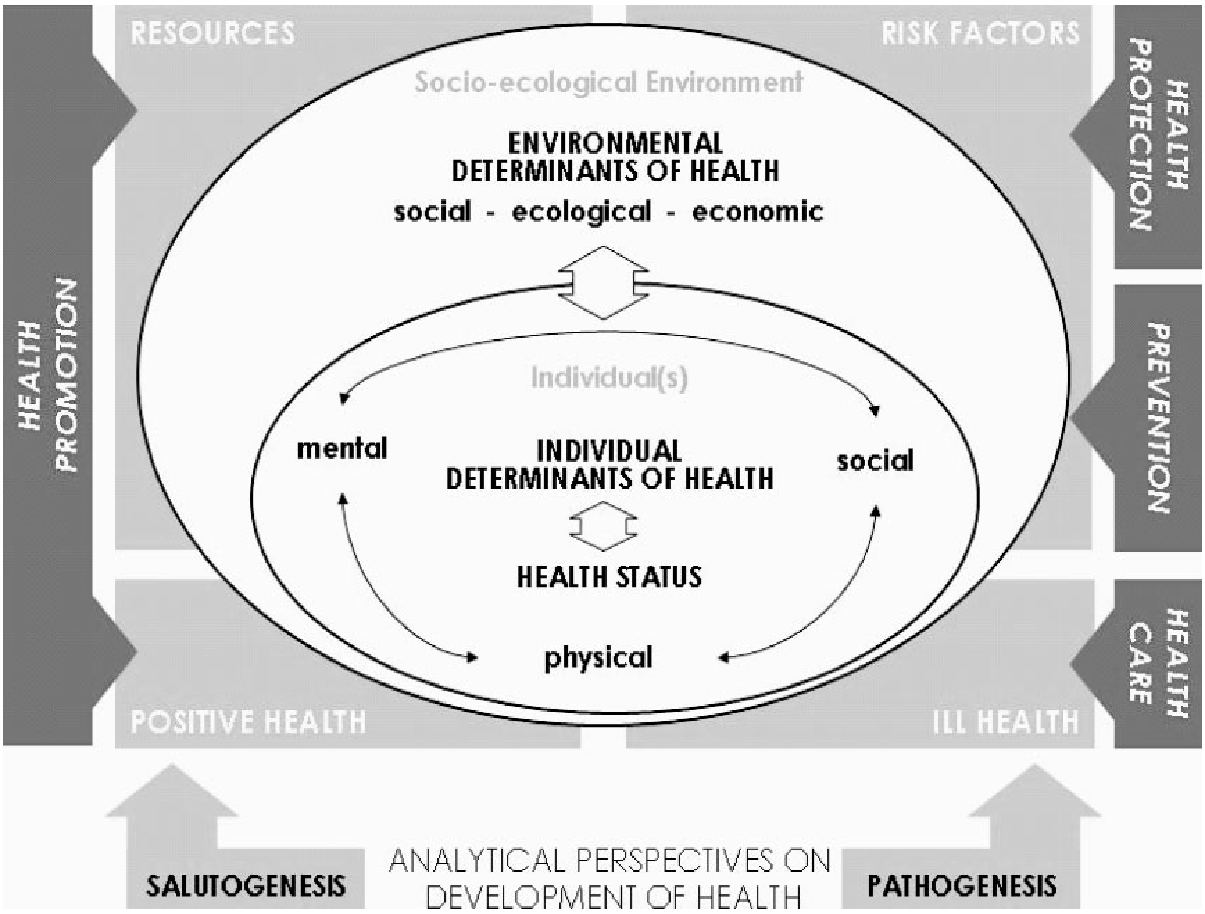


Figure 7.1 Analytical Perspectives on the Development of Health; Becker et al. (2010)

Appendix 11 shows an example of an assessment tool developed out of the outcomes of this study, which blends the salutogenic and pathogenic approach into a single integrated assessment. This kind of blending of approach would enable a clear focus on salutogenic approaches, whilst recognising clinical needs and planning for the risks explicit in-home LTV care.

7.3 Methodological Recommendations

The use of a diary approach added a longitudinal dimension of time and reflection into the findings and provided a wider understanding than that obtainable from an interview method alone. The methodology of using diaries to gain a more complete picture of a Child's care (beyond the day to day frustrations of complex care) is an area identified in this study, which could usefully be applied in future research.

Children's participatory scenarios are an engaging new method, which has a wider potential in Children focused research. The use of this method in Children with other complex conditions would be an interesting progression, building upon the experience of this study. Using participatory scenarios with Children with more complex disabilities is a potentially more challenging, but interesting area for this approach to be considered and developed.

7.4 Closing Thoughts

This study has concluded with recommendations, which if implemented have the potential to deliver a more personalised, empowering and health promoting approach to providing care for Children and families with CCHS. However, the approach is limited in transferability across the wider field of LTV (without further study), due to the specific needs of Children with CCHS; most notably high levels of physical and cognitive ability and the ability for most to come off ventilation during the day. Other studies have noted the effect of these issues on findings (Lindahl and Lindblad 2011:251). A larger study is therefore required to determine if the approach used in this study has wider applicability in LTV and complex disability. Long-term ventilation is a central component of families' lives, requiring an integrated family centred approach to care including: personalisation of care, integral long-term condition management and caseload management support. A change from service driven provision to an approach of achieving and developing family and community resources, to achieve Child and family aims, allows for more creative models of care and most importantly, a shared belief amongst professionals and families, that they are working together to achieve Child and family derived outcomes.

Areas for further Research. It would be interesting to reproduce this study across the wider none CCHS group of LTV Children and families, particularly those with more complex physical disabilities.

For the young Children in this study, it appeared that separation from their parents in terms of their management of their CCHS occurred much later than expected. There are no LTV studies in this area but studies of self-care in chronic illness have noted the link with self-care and Child development (Macorby 1984). This would seem a useful area for further research.

The finding about involving siblings in care, although potentially important, should be treated with some caution however, as it was gained from small amounts of data from two siblings and a parental perspective from two family interviews. This therefore is an area that requires further study from the perspective of a larger group of siblings themselves.

The need for planning for discharge is clear in the existing literature (2.4.1); however, the importance of planning for family milestones is a new finding. This finding is small but novel and suggests a small but important step towards making discharge more personalised. However further research is required into this area.

7.5 Prologue to the Research Journey

After 31 years of nursing with 17 years' experience of working in the field of Children's LTV, it could have been very difficult to break free of the preconceptions I had built up. The rigor and change of focus enabled by the PhD process and the focusing of this study on the salutogenic approach helped me to question every aspect of the process of assessing, planning and delivering LTV care from a completely different paradigm, from that in which I had previously operated. The process has been inspirational and challenging. This approach has changed not just my view of LTV care, it has challenged my views of nursing and the current approaches applied in the National Health Service (NHS) to the management of long-term conditions.

The Children and families involved in this research have been inspirational to work with. By approaching them via their long-established support group, I have been able to be a researcher, rather than part of the system. By being outside the system for the first time in 31 years of nursing practice, I have been freed up to think in a fundamentally different way (explored in 4.6).

The grant funding, which underpinned this study from the WellChild charity, has allowed me to trial innovative, effective and fun approaches to involving Children in research. I started with an approach that Child and family research should be more than do no harm to me, it had to be beneficial not just to future Children, but to the Children who were actually taking part. The provision of a play room for annual events, staffed by play specialists, set parents free to support each other and learn more about their Child's conditions. For the Children, the play room has been an important addition to their CCHSUKSG experience. For some, the family diaries charted real progress with their independence and progress with their Child's condition, which started with their interaction with their peers in the playroom. My team and I have been so inspired with the feedback from this activity, that we have continued to support the playroom at CCHSUKSG events through our team’s volunteer service.

As a student who has struggled all my life with dyslexia, this study has been one of the greatest challenges of my life, however the creativeness that thinking differently, which has been associated with dyslexia, (Everatt et al. 1999) has enabled me to develop new and novel approaches to each part of the study. Going forward in reference to the findings of this study, I plan to put the problems and restrictions of dyslexia behind me and recognise the creative and novel approaches that thinking differently enables.

A prototype model has been developed and evaluated alongside this study (Appendix 12). I thought this radically different system would be very challenging for staff and families immersed in a problem focused approach. However, the positive reception of this model has been very promising, not just from staff, but from the children, young people and their families. The openness and excitement generated by a new approach, has opened the possibility of operationalising the findings of this research, through a new practice tool thereby, enabling further research into the effectiveness and impact of this new approach I now realise that this PhD, rather than being at an end, is the start of a new chapter.

References.

ACT, (2007) *The Transition Care Pathway. A Framework for the development of Integrated Multi-Agency Care Pathways for Young People with Life-threatening and Life-Limiting Conditions*. Bristol: ACT

Adler, P. A., Adler, P. cited in Baker, S. E., Edwards, R. (eds.) (2012) *How many qualitative interviews is enough? Expert voices and early career reflections on sampling and cases in qualitative research*. Southampton. National Centre for Research Methods

Alaszewski, A. (2006) *Using Diaries for Social research*. Introducing Qualitative Methods Series. London: Sage

Alexander, E., Rennick, JE., Carnevale, F., Davis, M. (2002) 'Daily struggles: Living with Long-Term Childhood Technology dependence'. *Canadian Journal of Nursing Research*, 34 (4), 7-14

Angst, D. B., Deatrick, J. A. (1996) 'Involvement in Health Care Decisions: Parents and Children with Chronic Illness', *Journal of Family Nursing*, 2(2), pp. 174–194. doi: 10.1177/107484079600200205.

Antonovsky A (1998) cited in McCubbin, H. I., Thompson, E. A., Thompson, A. I., Fromer, J. J. Eds (1998) *Stress, Coping, and Health in Families. Sense of Coherence and Resiliency*. London: Sage

Antonovsky, A. (1996) 'The salutogenic model as a theory to guide health promotion'. *Health Promotion International* 11 (1), 11-18

Antonovsky, A. (1979) *Health, Stress and Coping*. London: Jossey-Bass Ltd.

Antonovsky, A. (1990) 'A somewhat personal odyssey in studying the stress process'. *Stress Medicine* 6 (2) 71-80 cited in Mittelmark, M.B., Sagy, S., Eriksson, M., Bauer, G., Pelikan, J.M., Lindström, B., Espnes, G.A. (Eds.) 2017 *The Handbook of Salutogenesis*. Switzerland: Springer International Publishing

Antonovsky, A. (1987) *Unravelling the Mystery of Health. How People manage Stress and Stay Well*. London: Jossey-Bass Ltd

Antonovsky, A., Sourani, T. (1988) 'Family Sense of Coherence and Family Adaptation.' *Journal of Marriage and Family* 50 (1), 79–92

Apers, S., Rassart, J., Luyckx, K., Oris, L., Goossens, E., Budts, W., Moons, P., & For The I-Detach Investigators. (2016) 'Bringing Antonovsky's salutogenic theory to life: A qualitative inquiry into the experiences of young people with congenital heart disease.' *International Journal of Qualitative Studies on Health and Well-being* 11(1)

Aslam, S (2018) 'Pinterest by the Numbers: Stats, Demographics & Fun Facts' [online] available at <https://www.omnicoreagency.com/pinterest-statistics/> accessed 19 May 2018.

Bach, J. R., Gupta, K., Reyna, M., Hon, A. (2009) 'Spinal Muscular Atrophy Type 1: Prolongation of Survival by Noninvasive Respiratory Aids'. *BS PEDIATRIC ASTHMA, ALLERGY & IMMUNOLOGY*, 22 (4). DOI: 10.1089/pai.2010.0002

Bagnoli, A. (2009) 'Beyond the standard interview: the use of graphic elicitation and arts-based methods'. *Qualitative Research* 9 (5), 547–570 London: Sage

Bannister, D. (1966) ‘Psychology as an exercise in paradox.’ *Bulletin of the British Psychological Society*, 19(63), 21-26

Barter, C., Renold, E. (1999) ‘The Use of Vignettes in Qualitative Research’ [online] available at <http://sru.soc.surrey.ac.uk/SRU25.html>

Bartley, M., Schoon, I., Mitchell, R., Blane, D. (2011) ‘Resilience as an asset for healthy development’ [online] available from <http://www.ucl.ac.uk/capabilityandresilience/resilienceasanassetforhealthydevelopment.doc> [11th November 2011]

Bauer, G., Davies, J. K., Pelikan, J. (2006) ‘The EUHPID Health Development Model for the Classification of Public Health Indicators’. *Health Promotion International* 21 (2), 153-159

Becker, C. M., Glascoff, M. A., Felts, W. M. (2010) ‘Salutogenesis 30 Years Later: Where do we go from here?’. *International Electronic Journal of Health Education*, 13 (25)

Bluebond-Langne, R., Angst, D., Lask, B. (Eds) (2001) *Psychosocial Aspects of Cystic Fibrosis.* London. Arnold Publishers

Bolger, N., Davis, A., and Rafaeli, E. (2003) cited in Alaszewski, A. (2006) *Using Diaries for Social research*. Introducing Qualitative Methods Series. London: Sage

Bonmatí-Tomás, A., Malagón-Aguilera, M., Bosch-Farré, C., Gelabert-Vilella, Juvinyà-Canal, D., Gil, M., Bonmatí-Tomás et al. (2016) ‘Reducing health inequities affecting immigrant women: a qualitative study of their available assets’. *Globalization and Health*, 12 (37)

Bragg, S., Buckingham, D. (2008*) ’Scrapbooks’ as a resource in media research with young people. in Doing Visual Research with Children and Young People* ed by Thomson, P. London: Routledge, 114–131

Breathe on (n. d.) available from <http://www.breatheon.org.uk>

Briassoulis, G., Filippou, O., Natsi, L., Mavrikiou, M., Hatzis, T. (2004) ‘Acute and chronic paediatric intensive care patients: current trends and perspectives on resource utilization’. *QJM. An International Journal of Medicine*, 97(8), 507-18

Bryman, A. (1984) ‘The Debate about Quantitative and Qualitative Research: A Question of Method or Epistemology?’. *The British Journal of Sociology*. 35 (1), 75-92

Burns, Andrea. B., Brown, Jessica. S., Sachs-Ericsson, Natalie., Ashby Plant, E., Curtis, Thomas., Fredrickson, Barbara. L, Joiner, T. (2007) *Upward spirals of positive emotion and coping: replication, extension, and initial exploration of neurochemical substrates* NIH Public Access Author manuscript, London: Elsevier Ltd

Carlin, J., (2005), *Including me. Managing complex health needs in schools and early years settings*. London: Department for Education and Skills, Council for Disabled Children.

Carlson, J. A. (2010) Avoiding Traps in Member Checking. The Qualitative Report, 15(5), 1102-1113. [online] available from <http://nsuworks.nova.edu/tqr/vol15/iss5/4> [June 2018]

Carnevale, F. A., Alexander, E., Davis, M., Rennick, J., Troini, R. (2006) cited in Linahl, B., Lindblad, BM. (2011) ‘Family Members’ Experiences of Everyday Life when a Child is dependent on a ventilator: A Metasynthesis Study’. *Journal of Family Nursing* (17), 241-269

Carnevale FA, Alexander E, Davis M, Rennick J, Troini R. (2006) 'Daily living with distress and enrichment: the moral experience of families with ventilator-assisted children at home'. *Pediatrics. American Academy of Pediatrics*. 117(1), 48-60

Carr, A. (2004) *Positive psychology The Science of happiness and human strengths*. Hove East Sussex. Brunner-Routledge

Carter, B., Cummings, J., Cooper, L. (2007) 'An exploration of best practice in multi-agency working and the experiences of families of children with complex health needs. What works well and what needs to be done to improve practice for the future?'. *Journal of Clinical Nursing* 16 (3), 527-39

Carter, B. (2005) Cited in Carter, B., Cummings, J., Cooper, L. (2007) 'An exploration of best practice in multi-agency working and the experiences of families of children with complex health needs. What works well and what needs to be done to improve practice for the future?'. *Journal of Clinical Nursing* 16 (3), 527-39

CCHS Support Group (n. d.) available from <http://cchssupport.co.uk/about.html>

CCHS UK. (n.d.) *What is Congenital Central Hypoventilation Syndrome*? [online] available from <https://static1.squarespace.com/static/590e03c1725e25240d277e1a/t/5919749ef7e0ab8d479fb777/1494840482841/UK+CCHS+info.pdf>

Center on Salutogenesis. (2015) available from http://www.salutogenesis.hv.se/eng/Salutogenesis.5.html

Centre for Reviews and Dissemination (2009) *Systematic Reviews. CRD's guidance for undertaking reviews in health care*. University of York.

Children's Hospices UK, (2011) *A Guide to Support Ventilated Children & Young People in Children's Hospices*. Bristol: Children's Hospices UK

Coad, J. (Lead) (2013) *BREATHING MATTERS – Improving Care with a Training Needs Analysis of Ventilated Children, Young People, Families and Professionals. Final Project Report*. Coventry University, CCFAR, Department of Health

Coad, J. (2007) 'Using art-based techniques in engaging children and young people in health care consultations and/or research'. *Journal of Research in Nursing* 12 (5), 487–497

Contact a Family (n. d.) [online] available from <http://www.cafamily.org.uk/medical-information/conditions/c/congenital-central-hypoventilation-syndrome/?s=congenital+central+hypoventilation+syndrome>

Cooperrider, D., Whitney, D., Stavros, J. M. (2009) *Appreciative Inquiry Handbook for Leaders of Change*. London: Berrett-Koehler

Council for Disabled Children. (2011) *Managing My Way*. London. National Children's Bureau

Council for Disabled Children. (2010*) Guidelines on the discharge from hospital of children and young people with high support needs*. London: National Children's Bureau

Cowley, S., Billings, J. R. (1999) 'Resources revisited: salutogenesis from a lay perspective'. *Journal of Advanced Nursing*, Blackwell Science Ltd, 29(4), 994-1004

Dabrowska, A. (2008) 'Sense of coherence and coping with stress in fathers of children with developmental disabilities'. *Polish Psychological Bulletin* 39 (1), 29

Daniel, M., Mathias, A. (2012) ‘Challenges and coping strategies of orphaned children in Tanzania who are not adequately cared for by adults’. *African Journal of AIDS Research* 11(3), 191–201

Davis, L. J. Ed. (2006) *The disability studies reader 2nd edition*. London. Routledge

Day Good, K. (2012) ‘From Scrapbook to Facebook: A history of personal media assemblage and archives’. *New Media & Society*, 15 (4), 557-573

Dempster, T., Cooper, L., Petty, G. (2011) ‘Grief and bereavement − supporting siblings of a children's hospice service Grief and bereavement − supporting siblings’ *BMJ Support Palliative Care* 1, 218 doi: 10.1136/bmjspcare-2011-000105.40

Denzin, N. K. (1978) cited in Miles, M., Huberman, M. (1994) *Qualitative Data Analysis. An Expanded Sourcebook. Second Edition*. London: Sage

Denzin, N. K., Lincoln, Y. S. (2008) *Strategies of Qualitative Inquiry, Third Edition.* London: Sage

Department for Education and Skills (DFES). (2007) *Aiming high for disabled children: better support for families*. London: HM Treasury

Department for Education and Skills, Department of Health (DH). (2004) *National Service Framework for Children, Young People and Maternity Services*. London: Department of Health

Department for Education and Skills. (2003) *Every Child Matters*. Norwich: The Stationary Office

Department for Education, Department for Health and Social Care. (2015) *SEND code of practice: 0 to 25 years Guidance on the special educational needs and disability (SEND) system for children and young people aged 0 to 25, from 1 September 2014*. London. Department for Education

Department of Health. (2010) *National Framework for Children and Young People’s Continuing Care*. London: Department of Health

Department of Health Independent Review Team. (2007) *Independent Review of palliative care Services for Children and Young People: Economic Study. Final Report*. York: York Health Economics Consortium.

Department of Health (2000) *Framework for the Assessment of Children in need and their Families*. Norwich: The Stationary Office.

Department of Health and Social Care (2016) *Children and young people’s continuing care national framework Process for assessing, deciding and agreeing continuing care for children with complex health needs*. [Online] available at <https://www.gov.uk/government/publications/children-and-young-peoples-continuing-care-national-framework>

Dietscher, C., Winter, U., Pelikan, J. (2017) *The Application of Salutogenesis In Hospitals* cited in Mittelmark, M.B., Sagy, S., Eriksson, M., Bauer, G., Pelikan, J.M., Lindström, B., Espnes, G.A. Eds. (2017) ‘The Handbook of Salutogenesis.’ Switzerland. Springer International Publishing

Droter, D. (1978) cited in Bluebond-Langne, R., Angst, D., Lask, B. (Eds) (2001) *Psychosocial Aspects of Cystic Fibrosis*. London. Arnold Publishers

Dybwik, K., Nielsen, E.W., Brinchmann, B.S. (2011) ‘Home mechanical ventilation and specialised health care in the community: between a rock and a hard place’. *BMC Health Services Research*, 11 (115), 1-8

Dybwik, K., Tollåli, T., Nielsen, E. W., Brinchmann, B. S., Dybwik et al. (2011) ‘Fighting the system: Families caring for ventilator-dependent children and adults with complex health care needs at home’. *BMC Health Services Research*, (11), 156

Eaves, S. (2014) ‘From Arts for Art’s Sake to Arts as Means of Knowing: A Rationale for Advancing Arts-Based Methods in Research, Practice and Pedagogy.’ *The Electronic Journal of Business Research Methods* 12 (2), 147-160

Edwards, R., Holland, J. (2013) *What is Qualitative Interviewing*? London. Bloomsbury

Edwards, E. A., O’Toole, M., Wallis, C. (2004) ‘Sending children home on tracheostomy dependent ventilation: pitfalls and outcomes’. *Archives of Disease in Childhood* 89, 251-255

Edwards, A., Talbot, R. (1999) *The Hard-pressed Researcher: A Research Handbook for the Caring Professions (2nd edition)* London: Longmans

Eide, P., Kahn, D. (2008) ‘Ethical issues in the qualitative researcher--participant relationship.’ *Nursing Ethics* 15 (2),199-207 doi: 10.1177/0969733007086018

Einberg, E., Svedberg, P., Enskär, K., Nygren, J. M., (2015) ‘Friendship Relations from the Perspective of Children with Experience of Cancer Treatment: A Focus Group Study with a Salutogenic Approach. *Journal of Pediatric Oncology Nursing* 32(3), 153–164

Emmons, R.A., & McCullough, M.E. (2003). Counting blessings versus burdens: An experimental investigation of gratitude and subjective well-being in daily life. *Journal of Personality and Social Psychology*, 84, 377–389.) cited in Lambert, N. M., Graham, S. M., Fincham, F. D., Stillman, T. F. (2009) ‘A Changed Perspective: How Gratitude can Affect Sense of Coherence Through Positive Reframing’. *The Journal of Positive Psychology* 4 (6), 461-470

Enright, E., Hill, J., Sandford, R., Gard, M. (2014) ‘Looking beyond what's broken: towards an appreciative research agenda for physical education and sport pedagogy’ *Sport, Education and Society*, 19 (7)

Eriksson, M., Lindstrom, B. (2008) ‘A salutogenic interpretation of the Ottawa Charter’. *Health Promotion International* 23 (2), 190-199

Eriksson, M., Lindstrom, B. (2005) ‘Validity of Antonovsky’s sense of coherence scale: a systematic review’. *Journal of Epidemiology & Community Health* 59, 460-466

Evans, R. (2014) *Children who are Long Term Ventilated – Pathfinder Project Engaging with Families and Children / Young People An independent quality improvement review* by the Patient Experience Network for NHS England. London: TSO

Everatt, J., Steffert, B., Smythe, I., (1999) ‘An eye for the unusual: creative thinking in dyslexics’. *DYSLEXIA,* 5, 28–46

Finch, J. (1987) ‘The Vignette Technique in Survey Research.’ *Sociology*, 21, 105-14

Fine-Goulden, M.R., Ray, S., Brierley, J. (2015) ‘Decision making in long-term ventilation for children’. The *Lancet Respiratory Medicine* 3(10), 745–746

Finkel, RS., Chiriboga, C.A., Vajsar, J., Day, J.W., Montes, J., De Vivo, D.C., Yamashita, M., Rigo, F., Hung’ G., Schneider, E., Norris, D.A., Xia, S., Bennett, C.F., Bishop, K.M.

(2016) ‘Treatment of infantile-onset spinal muscular atrophy with nusinersen: a phase 2, open-label, dose-escalation study’. *The Lancet*, 388 (10063), 3017-3026

Flensborg-Madsen, T., Ventegodt, S., and Merrick, J. (2006) ‘Sense of coherence and health. The construction of an amendment to Antonovsky’s sense of coherence scale (SOC II)’. *TSW Holistic Health & Medicine* 1 169–175.

Flensborg-Madsen, T., Ventegodt, S., Merrick, J. (2005) ‘Why is Antonovsky’s Sense of Coherence Not Correlated to Physical Health? Analysing Antonovsky’s 29-item Sense of Coherence Scale (SOC-29)’. The *Scientific World Journal* 5, 767-776

Flick, U. (2009) *An Introduction to Qualitative Research.* Los Angeles: Sage Publications.

Fowler, M. D. M. (1988) ‘Ethical Issues in Nursing Research’ Issues in Qualitative Research, 10 (1), 109-111 https://doi.org/10.1177/019394598801000112

Frankl, Viktor. E. (2004). *Man’s Search For Meaning. The Classic Tribute to Hope from the Holocaust.* UK, Rider, an imprint of Edbury Publishing.

Fraser, K., Miller, M., Aldridge, J., McKinney, P. A., Parslow, R. C. (2011) *Life-Limiting and life-threatening conditions in children and young people in the United Kingdom; national and regional prevalence in relation to socioeconomic status and ethnicity. Final Report for Children’s Hospice UK*. Leeds: Division of Epidemiology, University of Leeds

Fraser, S., Lewis, V., Ding, S., Kellett, M., Robinson, C. eds. (2004). *Doing Research with Children and Young People.* London, UK: Sage.

Fredrickson, B. L., Joiner, T. (2002) ‘Positive Emotions Trigger Upward Spirals toward Emotional Well-Being’. *Psychological Science* 13 (2), 172-175

Fredrickson, B. L., (1998) ‘What Good Are Positive Emotions?’ *Review of general psychology*, 2(3), 300–319. doi: 10.1037/1089-2680.2.3.300

Fusch, P. I., Ness, L. R. (2015) ‘Are we there yet? Data saturation in qualitative research.’ *The Qualitative Report* 20(9), 1408-1416. Retrieved from <http://www.nova.edu/ssss/QR/QR20/9/fusch1.pdf>

Garland, E. L., Fredrickson, B. L., Kring, A. M., Johnson, D. P., Meyer, P. S., Penn, D. L., (2010) ‘Upward spirals of positive emotions counter downward spirals of negativity: insights from the broaden and build theory and affective neuroscience on the treatment of emotional dysfunctions and deficits in psychopathology’. *Clinical Psychology Review* 30, (849-864) 172-175

Generic Processes of Appreciative Inquiry. (n. d.) The center for appreciative inquiry. Las Vegas USA. Available from <https://www.centerforappreciativeinquiry.net/more-on-ai/the-generic-processes-of-appreciative-inquiry/ > [3 May 2017]

Gilgoff, R. L., Gilgoff, I. S. (2003) Long-Term Follow-Up of Home Mechanical Ventilation in Young Children with Spinal Cord Injury and Neuromuscular Conditions’. *The Journal of Pediatrics* 142, 476-480

Glaser, B. G. (1978) cited in Miles, M., Huberman, M. (1994) *Qualitative Data Analysis. An Expanded Sourcebook. Second Edition*. London: Sage.

Glazier and Strauss (1978) cited in Antonovsky A (1987) *Unravelling the Mystery of Health. How People manage Stress and Stay Well*. London. Jossey-Bass Ltd.

Goodsell, T. L., Seiter, L. (2011) 'Scrapbooking: Family Capital and the Construction of Family Discourse.' *Journal of contemporary Ethnography*, 40 (3), 318-341 doi.org/10.1177/0891241611399437

Grant, G. J., Booth, A. (2009) A typology of reviews: 'An analysis of 14 review types and associated methodologies'. *Health Information & Libraries Journal* 26, (2) 91-108

Green, J., Thorogood, N. (2014) *Qualitative Methods for Health Research*. Third Edition. London: Sage.

Hazel, N. (1995) *Elicitation Techniques with Young People, Social Research Update*, 12, Department of Sociology, University of Surrey, available online <http://www.soc.surrey.ac.uk/sru/SRU12.html>

Heaton, J., Noyes, J., Sloper, P., Shah, R. (2005) 'Families' experiences of caring for technology-dependent children: a temporal perspective.' *Health and Social Care in the Community* 13 (5), 441-450

Hewitt-Taylor, J. (2004) 'Children who require long-term ventilation: staff education and training'. *Intensive and Critical Care Nursing* 20, 93—102

Hintermair, M. (2004) 'Sense of Coherence: A Relevant resource in the Coping Process of Mothers of Deaf and Hard-of-Hearing Children?'. *Journal of Deaf Studies and Deaf Education* 9 (1), 15-26

Howe, K. R. (2004) 'Against the Quantitative-Qualitative Incompatibility Thesis or Dogmas Die Hard'. *Educational Researcher* 17 (8), 10 –16

Hsieh, H-F., Shannon, S. E. (2005) 'Three Approaches to Qualitative Content Analysis'. *Qualitative Health Research* 15 (9), 1277–1288

Hughes, C. (n. d.) DEVELOPING REFLEXIVITY IN RESEARCH [online] available from <https://www.warwick.ac.uk/.../researchprocess/developing_reflexivity_in_research.doc>

Hughes, R. (1998) 'Considering the Vignette Technique and its Application to a Study of Drug Injecting and HIV Risk and Safer behavior.' *Sociology of Health and Illness* 20 (3) 381-400

Hull, J., Aniapravan, R., Chan, E., Chatwin, M., Forton, J., Gallagher, J., Gibson, N., Gordon, J., Hughes, I., McCulloch, R., Russell, R. R., Simonds, A. (2012) 'Society guideline for respiratory management of children with neuromuscular weakness.' *Thorax* 67: i1ei40. doi:10.1136/thoraxjnl-2012-201964

Hutchfield, K. (1999) 'Family-centred care: a concept analysis'. *Journal of Advanced Nursing* 29 (5), 1178-1187

Jardine, E. & Wallace, C., (1998) cited in Noyes, J., Hartmann, H., Samuels, M., Southall, D. (1999) 'The experiences and views of parents who care for ventilator-dependent children'. *Journal of Clinical Nursing* 8, 440-450

Jardine, E., O'Toole, M., Paton, J.Y., Wallis, C. (1999) 'Current status of long term ventilation of children in the United Kingdom: questionnaire survey'. *British Medical Journal* 318, 295

Jensen, B.B., Dur, W., Buijs, G. (2017) *The Application of Salutogenesis in Schools* cited in Mittelmark, M.B., Sagy, S., Eriksson, M., Bauer, G., Pelikan, J.M., Lindström, B., Espnes, G.A. Eds. (2017) The Handbook of Salutogenesis. Switzerland. Springer International Publishing

Johnson, B. R., Onwuegbuzie, A.J. (Oct. 2004) 'Mixed Methods Research: A Research Paradigm Whose Time Has Come' *Educational Researcher* 33, (7) 14-26

Judge, S. L. (1998) 'Parental Coping Strategies and Strengths in Families of Young Children with Disabilities'. *Family Relations* 47 (3), 263-268

Kai-Wei, K., Wang, A. B. (2008) 'Caregivers' Experiences at Home with a Ventilator-Dependent Child., Qualitative Health Research 18 (4), 501-508

Kandel, I., Merrick, J. (2007) 'The Child with a Disability: Parental Acceptance, Management and Coping'. *The Scientific World Journal* 7 1799-1809

Karns, J. T. (2002). Scrapbooking during traumatic and transitional events. Journal of Clinical Activities, Assignments and Handouts in Psychotherapy Practice, 2(3), 39–47. cited in King, J., Prout, B., Stuhl, A., & Nelson, R. (2016) 'Scrapbooking as an intervention to enhance coping in individuals experiencing grief and loss'. *Therapeutic Recreation Journal* l, (2), 181–185

Kelle, U. (2005) '"Emergence" vs. "Forcing" of Empirical Data? A Crucial Problem of "Grounded Theory" Reconsidered.' Forum Qualitative Sozialforschung / Forum: *Qualitative Social Research*, [S.l.], 6 (2), Available at: <http://www.qualitative-research.net/index.php/fqs/article/view/467/1000>. Date accessed: 22 May 2018.

Kennedy, I. (2001) 'Learning from Bristol. *The Report of the Public Inquiry into children's heart surgery at the Bristol Royal Infirmary 1984-1995*. London. The Stationary Office

Kennedy, I. (2010) *Getting it Right for Children and Young People. Overcoming cultural barriers in the NHS so as to meet their needs*. A Review by Professor Ian Kennedy'. London. Department of Health

Kenten, C. (2010) cited in Edwards, R., Holland, J. (2013) *What is Qualitative Interviewing?* London. Bloomsbury

Kirk S, Beatty S, Callery P, Milnes L, Pryjmachuk S. (2010) *Evaluating Self-Care Support for Children and Young People with Long-Term Conditions. Final report*. NIHR SDO programme' [online] available at <http://www.netscc.ac.uk/hsdr/files/project/SDO_FR_08-1715-162_V01.pdf>

Kirk, S., Glendinning, C. (2003) 'Developing services to support parents caring for technology-dependent child at home.' *Child: Care, Health & Development* 30 (3), 209-218

Kirk, S., Glendinning, C., Callery, P. (2004) 'Parent or Nurse? The experience of being the parent of a technology-dependent child.' *Journal of Advanced Nursing* 51 (5), 456-464

Kirk, S., Glendinning, C. (2002) 'Supporting expert parents - professional support and families caring for a child with complex health care needs in the community'. *International Journal of Nursing Studies* 39, 625 – 635

Lambert, N. M., Graham, S. M., Fincham, F. D., Stillman, T. F. (2009) 'A Changed Perspective: How Gratitude can Affect Sense of Coherence Through Positive Reframing' *The Journal of Positive Psychology* 4 (6) 461-470

Langeland, E., Vinje, H. F. (2017) *The application of Salutogenesis in Mental healthcare settings* cited in Mittelmark, M.B., Sagy, S., Eriksson, M., Bauer, G., Pelikan, J.M., Lindström, B., Espnes, G.A. Eds. (2017) 'The Handbook of Salutogenesis.' Switzerland. Springer International Publishing

Leavy, P. (2009). Method Meets Arts: Arts-based Research Practice. Guilford Press: New York:149 cited in Eaves, S. (2014) 'From Arts for Art's Sake to Arts as Means of

Knowing: A Rationale for Advancing Arts-Based Methods in Research, Practice and Pedagogy.' *The Electronic Journal of Business Research Methods* 12 (2), 147-160

LIMA, M. F., De Paulo, L. F., Higarashi, L. H. (2015) Technology-dependent children: the meaning of home care – a descriptive study. [Online] *Brazilian Journal of Nursing,* [S.I.], 14 (2) 178-89, Available at:<http://www.objnursing.uff.br/index.php/nursing/article/view/5191/pdf_524> [14 June 2018].

Lindahl, B., Lindblad, B.M. (2013) 'Being the parent of a ventilator-assisted child: perceptions of the family-health care provider relationship when care is offered in the family home.' *Journal of family Nursing* 19, (4)489-508. doi: 10.1177/1074840713506786

Lindahl, B.' Lindblad, BM. (2011) 'Family Members' Experiences of Everyday Life when a Child is dependent on a ventilator: A Metasynthesis Study'. *Journal of Family Nursing* 17, 241-269

Lindstrom, B., Eriksson, M. (2007) 'Antonovsky's sense of coherence scale and its relation with quality of life: a systematic review'. *Journal of Epidemiol Community Health* 61, 938-944

Lindstrom, B., Eriksson, M. (2006) 'Contextualizing salutogenesis and Antonovsky in public health development'. *Health Promotion International* 21 (3), 238-244

Lindstrom, B., Eriksson, M. (2005) 'Salutogenesis'. *Journal of Epidemiol Community Health* 59, 440-442

Livingstone, S., Haddon, L., Görzig, A., Ólafsson, K. (2011) Risks and safety on the internet: the perspective of European children: full findings and policy implications from the EU Kids Online survey of 9-16-year olds and their parents in 25 countries. EU Kids Online, Deliverable D4. EU Kids Online Network, London, UK.

Llahana, S.V., Poulton, B. C., Coates, V. E. (2001) 'The Paediatric diabetes specialist nurse and diabetes education in childhood.' *Journal of Advanced Nursing*, 33 (3), 296-306

Londal, K. (2010) 'Children's Lived Experience and their Sense of Coherence: Bodily Play in a Norwegian After-school Programme'. *Child Care in Practice* 16 (4), 391-407

Maccorby, E. E. (1984) cited in Bluebond-Langne, R., Angst, D., Lask, B. (Eds) (2001) *Psychosocial Aspects of Cystic Fibrosis.* London. Arnold Publishers

MacLure, M (2008:174) cited in St. Pierre, E.A., Jackson, A.Y. (2014) 'Qualitative Data Analysis After Coding'. *Qualitative Inquiry* 20 (6), 715 – 719

Maddox, C., Pontin, D. (2013) Paid carers' experiences of caring for mechanically ventilated children at home: Implications for services and training. *Journal of Child Health Care* 17, (2) 153–163 DOI: 10.1177/1367493512456113

Margalit, M., Kleitman, T. (2006) 'Mothers' stress, resilience and early intervention'. *European Journal of Special Needs Education*, 21, (3) 269-283

Margolan, H., Fraser, J., Lenton, S. (2004) 'Parental experience of services when their child requires long-term ventilation. Implications for commissioning and providing services'. *Child: Care, Health & Development* 30 (3), 257-264

Mattiuzzi, P. G. (2007) 'The sense of Coherence' [online] available from <http://everydaypsychology.com/2007/04/sense-of-coherence.html> [9th May 2012]

Mays, N., Pope, C. (2000) 'Assessing quality in qualitative research' *Qualitative Research in Health Care* 320, (50)

McCarthy, P.G., Sebaugh, J.G. (2011) 'Therapeutic scrapbooking: a technique to promote positive coping and emotional strength in parents of pediatric oncology patients'. *Journal of Psychosocial Oncology*, 29(2): 215-30.

McCollum, A. I., Gibson, L. E. (1970) cited in Bluebond-Langne, R., Angst, D., Lask, B. (Eds) (2001) *Psychosocial Aspects of Cystic Fibrosis.* London. Arnold Publishers

McCubbin, H. I., Thompson, E. A., Thompson, A. I., Fromer, J. J. Eds (1998) *Stress, Coping, and Health in Families. Sense of Coherence and Resiliency*. London. Sage publications Ltd.

McDougall, C. M., Adderley, R. J., Wensley, D. F., Seear, M. D. (2013) 'Long-term ventilation in children: longitudinal trends and outcomes.' *Archives of Disease in Childhood*, 98, 660-665. doi: 10.1136/archdischild-2012-303062

McGarry, J. (2007) 'Nursing relationships in ethnographic research: what of rapport?' *Nurse Research*,14(3) 7-14

Mendes, M. A. (2013) 'Parents' Descriptions of Ideal Home Nursing Care for Their Technology-Dependent Children.' *PEDIATRIC NURSING* 39 (2)

Miles, M., Huberman, M. (1994) *Qualitative Data Analysis. An Expanded Sourcebook. Second Edition*. London. Sage

Miles, M., Huberman, M., Saldana, J. (2014) *Qualitative Data Analysis. A Methods Sourcebook. Third Edition*. London. Sage

Miske, L.J., Hickey, E.M., Kolb, S.M., Weiner, D.J., Panitch, H.B. (2004) 'Use of the mechanical in-exsufflator in pediatric patients with neuromuscular disease and impaired cough'. *Chest*. 125(4), 1406-12

Mittelmark, M.B., Sagy, S., Eriksson, M., Bauer, G., Pelikan, J.M., Lindström, B., Espnes, G.A. (Eds.) 2017 *The Handbook of Salutogenesis*. Switzerland. Springer International Publishing

Morgan, A. (2012) *Revitalising evidence based public health: an assets model*. [online PPT] available from <http://www.phirn.org.uk/seminar%206/Antony%20Morgan.ppt> [9th May 2012]

Morgan, A., Ziglio, E. (2007) 'Revitalising the evidence base for public health: an assets model'. *Promotion & Education* 14, 17-22

Morrow, S. L. (2005) 'Quality and Trustworthiness in Qualitative Research in Counseling Psychology.' *Journal of Counseling Psychology* 52 (2), 250–260

Morris, J. (2001) 'Social exclusion and young disabled people with high levels of support needs'. *Critical Social Policy* 21, 161-183

Muscular Dystrophy UK, (2015) 'Breaking point. The crisis in accessible housing and adaptations'. Muscular Dystrophy UK.

Naaldenberg, J., Vaandrager, L., Koelen, M., Leeuwis, C. (2011) 'Aging Populations' Everyday Life Perspectives on Healthy Aging' *Journal of Applied Gerontology*, 31 (6), 711 – 733

National Health Interview Survey (1973) cited in Antonovsky, A. (1979) *Health, Stress and Coping*. London: Jossey-Bass Ltd

NHS England. (2018) https://www.england.nhs.uk/ourwork/patient-participation/patient-centred/planning/

NHS Estates. (2008) *Ward layouts with single rooms and space for flexibility* 2008 STATUS IN WALES. London: TSO

Nicholl, H. (2010) 'Diaries as a Method of Data Collection in Research'. *Paediatric Nursing* 22 (7), 16-20

Nilsson, K.W., Leppert, J., Simonsson, B., Starrin, B. (2010) 'Sense of coherence and psychological well-being: improvement with age.' *Journal of Epidemiology Community Health*. 64(4) 347-52. doi: 10.1136/jech.2008.081174

Noyes, J. (2006) 'Comparison of ventilator-dependent child reports of health-related quality of life with parents reports and normative populations.' *Journal of Advanced Nursing* 58 (1), 1-10

NOYES, J. (2002) 'Barriers that delay children and young people who are dependent on mechanical ventilators from being discharged from hospital'. *Journal of Clinical Nursing,* <https://doi.org/10.1046/j.1365-2702.2002.00565>

Noyes, J. (2000) 'Ventilator-dependent children who spend prolonged periods of time in intensive care units when they no longer have a medical need or want to be there'. *Journal of Clinical Nursing* 9, 774-783

Noyes, J., Hartmann, H., Samuels, M., Southall, D. (1999) 'The experiences and views of parents who care for ventilator-dependent children'. *Journal of Clinical Nursing* 8, 440-450

Noyes, J., Lewis, M., (2005) *From Hospital to Home*. Essex. Barnardo's

Nursing and Midwifery Council. (2013) *The code: Standards of conduct, performance and ethics for nurses and midwives*. London. Nursing and Midwifery Council

O'Brien, M.E. (2002) 'Living in a House of Cards: Family Experiences with Long-Term Childhood Technology Dependence.' *Journal for specialists in pediatric nursing*. 7 (1) 7 - 15

Oelofsen, N., Richardson, P. (2006) 'Sense of coherence and parenting stress in mothers and fathers of preschool children with developmental disability'. *Journal of Intellectual and Developmental Disability*, 31 (1), 1-12

Oliver, M. (1996) *Defining Impairment and Disability: Issues at Stake' in Exploring the Divide* ed. by Colin Barnes, C., Mercer, G. Leeds. The Disability Press, 29-54

Olsson, M.B., Hwang, C.P. (2002) 'Sense of coherence in parents of children with different developmental disabilities.' *Journal of intellectual disability research*, 46 (7), 548-59.

Owens, J. (2015) 'Exploring the critiques of the social model of disability: The transformative possibility of Arendt's notion of power'. *Sociology of Health and Illness*, 37(3) 385 - 403

Paediatric Intensive Care Audit Network: *A Decade of* Data (published September 2014): Universities of Leeds and Leicester. [Online] Available from: <http://www.picanet.org.uk/Audit/AnnualReporting/PICANet_A_Decade_of_Data_2014_Annual_Report_Summary.pdf.> [Accessed 27/02/2015].

Parab, C. S., Cooper, C., Woolfenden, S., Piper, S.M., (2013) ‘Specialist home-based nursing services for children with acute and chronic illnesses’. *Cochrane Database of Systematic Reviews*, 6. DOI: 10.1002/14651858.CD004383.pub3.

Petrelli, D., Whittaker, S. and Brockmeier, J. (2008) AutoTopography: *What Can Physical Mementos Tell us about Digital Memories*? In: CHI 2008. CHI 2008, 5-10 April 2008, Florence (Italy). ACM.

Petrelli, D., Whittaker, S. (2010) ‘Family memories in the home: contrasting physical and digital mementos’. *Personal and Ubiquitous Computing* 14 (2), 153–169

Pimlott, B. (2002), cited in Alaszewski, A. (2006) *Using Diaries for Social research. Introducing Qualitative Methods Series*. London. Sage.

Redouane, B., Cohen, E., Stephens, D., Keilty, K., Mouzaki, M., Narayanan, U., … Amin, R. (2016). Parental Perceptions of Quality of Life in Children on Long-Term Ventilation at Home as Compared to Enterostomy Tubes. *PLoS ONE*, 11(2), e0149999. http://doi.org/10.1371/journal.pone.0149999

Robinson, R. (1990) cited in Noyes, J., Hartmann, H., Samuels, M., Southall, D. (1999) ‘The experiences and views of parents who care for ventilator-dependent children’. *Journal of Clinical Nursing* 8, 440-450

Roper, H., Quinlivan, R., Workshop Participants. (2010) ‘Implementation of "the consensus statement for the standard of care in spinal muscular atrophy" when applied to infants with severe type 1 SMA in the UK’. *Archives of Disease in Childhood*. 95(10), 845-900

Rose, G. (2001): cited in White, A., Bushin, N., Carpena-Mendez, F., Laoire, C. N. (2010) ‘Using Visual Methodologies to Explore Contemporary Irish Childhoods’. Qualitative Research 10 (2) 143 - 158

Royal College of Nursing (2014) *Safeguarding children and young people – every nurse’s responsibility: RCN guidance for nursing staff*. London: RCN.

Royal College of Nursing, (2012) *Defining staffing levels for children and young people’s services. RCN Standards for clinical professionals and service managers*. London. RCN

Royal College of Nursing guidance (2006) *Specialist nursing services for children and young people with diabetes*. London. RCN

Russell, P. (2003) ‘Access and Achievement or Social Exclusion? Are the Government’s Policies Working for Disabled Children and Their families?’. *Children & Society* 17, 215-225

Sackett, D. L., Wennberg, J. E., (1997) ‘Choosing the best research design for each question’ *BMJ*, 315:1636

Saks, M., Alsop, J. (2013) *Researching Health Qualitative, Quantitative and Mixed Methods. Second Edition*. London. Sage

Saldafia, J. (2009) *The Coding Manual for Qualitative Researchers*. London. Sage

Sale, J.E.M., Lohfeld, L.H., Brazil, K. (2002) 'Revisiting the quantitative-qualitative debate: Implications for mixed-methods research'. *Quality and Quantity*, 36 (1), 43-53. DOI: 10.1023/A:1014301607592

Schreudera, E., Rijnders, M., Vaandragerb, L., Hassinkc, J., Enders-Slegersd, M., Kennedye, L. (2014) ‘Exploring salutogenic mechanisms of an outdoor experiential learning programme on youth care farms in the Netherlands: untapped potential?' *International Journal of Adolescence and Youth*, 19 (2), 139–152

Sarvey, S. I., (2008) 'Living with a machine: the experience of the child who is Ventilator dependent.' Issues in *Mental Health Nursing*, 29,179–196

Schrodinger, E. (1968) cited in Antonovsky A (1987) *Unravelling the Mystery of Health. How People manage Stress and Stay Well*. London. Jossey-Bass Ltd.

Secure Email Standard ISB 1596 NHS Digital (n. d.) [online] available at <https://digital.nhs.uk/services/nhsmail/the-secure-email-standard> accessed 22 May 20189

Seear, M., Kapur, A., Wensley, D., Morrison, K., Behroozi, A. (2016) 'The quality of life of home-ventilated children' *Arch Dis Child*, 101, 620-627

Seligman, M. (2002) cited in Carr, A. (2004) *Positive psychology The Science of happiness and human strengths*. Hove East Sussex. Brunner-Routledge

Seligman, M.E., Csikszentmihalyi, M. (2000) 'Positive psychology. An introduction.' The American Psychologist, 55(1), 5-14

Shakespeare, T. (2006) *'The Social Model of Disability' in The Disability Studies Reader 2nd edition*. ed. by Davis, L. J. London. Routledge, 173-184

Sheldon, K. M., King, L. (2001) 'Why Positive Psychology Is Necessary.' *American Psychologist*, 56 (3), 216-217 DOI: 1O.1O37//OOO3-O66X.56.3.216

Shenton, A. K. (2004) 'Strategies for Ensuring Trustworthiness in Qualitative Research Projects' *Education for Information,* 22 (2), 63-75

Shields, L. (2015) 'What is family-centred care?' *European Journal of Person Centered Healthcare*, 3 (2),139-144

Sidey, A., Widdas, D. Eds (2005) 'Textbook of community Children's Nursing. Second Edition'. London. Elsevier

Simonelli, F., Guerreiro, A. I. F., Simonelli, I., Pasquale, C. D. (2010) 'Six assertions about the salutogenic approach and health promotion'. *Italian Journal of Public Health* 7 (2), 94-101

Sivberg, B. (2002) 'Coping Strategies and Parental Attitudes. A comparison of parents with children with Autistic Spectrum Disorders and Parents with non-autistic Children'. *International Journal of Circumpolar Health* 61 (2), 36-50

Smith, J. K., (1983) 'Quantitative Versus. Qualitative Research: An Attempt to Clarify the Issue' *Educational Researcher* 12 (3) 6-13

Social Policy Research Unit, (2003) cited in Sidey, A., Widdas, D. Eds (2005) *Textbook of community Children's Nursing. Second Edition*. London. Elsevier

Spratling, R. (2015) 'Defining Technology Dependence in Children and Adolescents.' *Western Journal of Nursing Research* 37(5) 634–651 DOI: 10.1177/0193945914526002

Springgay, S., Lewin, R., Wilson Kind, S. (2005) cited in Eaves, S. (2014) 'From Arts for Art's Sake to Arts as Means of Knowing: A Rationale for Advancing Arts-Based Methods in Research, Practice and Pedagogy.' *The Electronic Journal of Business Research Methods* 12 (2), 147-160

St. Pierre, E. A., Jackson, A. Y. (2014) 'Qualitative Data Analysis After Coding.' Qualitative Inquiry 20 (6), 715-719

Steier, F. (1991) *Research and Reflexivity.* London. Sage

Stokes, R. H., (2010) 'Paediatric occupational therapy: Addressing parental stress with the sense of coherence'. The Canadian Journal of Occupational Therapy [online]. Available from <http://www.readperiodicals.com/201002/1970424341.html> [9th May 2012]

Strang, S., Strang, P. (2001) 'Spiritual thoughts, coping and 'sense of coherence' in brain tumor patients and their spouses'. *Palliative Medicine* 15, 127-134

Streubert, H., & Carpenter, D. (1999). Qualitative Research in Nursing: Advancing the Humanistic Perspective (2nd ed.). Philadelphia, PA: Lippincott Williams & Wilkins

Strümpfer, D. J. W. (1995) The origins of health and strength: from 'salutogenesis' to 'fortigenesis'. South African Journal of Psychology. 25(2), 81–9. Cited in Bonmatí-Tomás, A., Malagón-Aguilera, M., Bosch-Farré, C., Gelabert-Vilella, Juvinyà-Canal, D., Gil, M., Bonmatí-Tomás et al. (2016) 'Reducing health inequities affecting immigrant women: a qualitative study of their available assets'. *Globalization and Health*, 12 (37)

Sudbury, J., Noyes, J. (1999) Voices and Choices of Children and Young People Who Use Assisted Ventilation Bibliography and Analysis of the Literature. University of Salford & Joseph Rowntree Trust

Sullivan G (1993) cited in Noyes, J., Hartmann, H., Samuels, M., Southall, D. (1999) 'The experiences and views of parents who care for ventilator-dependent children'. *Journal of Clinical Nursing* 8, 440-450

Tan, J. S., Docherty, S. L., Barfield, R., Brandon, D. H. (2012) 'Addressing Parental Bereavement Support Needs at the End of Life for Infants with Complex Chronic Conditions.' *J Palliat Med*, 15(5), 579–584 doi: 10.1089/jpm.2011.0357

The NHS Confederation (2009) *Aiming high for disabled children: delivering improved health services*. London. NHS Confederation.

The Nuremberg Code (1947) *BMJ* 1996; 313 :1448

The Top 20 Valuable Facebook Statistics (2018) [online] available at <https://zephoria.com/top-15-valuable-facebook-statistics/> accessed 21 May 2018

Thedin, J. B. (2014) 'What makes teenagers continue? A salutogenic approach to understanding youth participation in Swedish club sports.' *Physical Education and Sport Pedagogy* 19 (3), 239-252

Thurmond, V. A. (2001) 'The Point of Triangulation.' *Journal of Nursing Scholarship*, 33(3), 253-258

Together for short lives, (2013) *A Core Care Pathway for children with Life-Limiting and Life-Threatening Conditions. Third edition*. Bristol. Together for Short Lives

Toly, V. B., Musil, C. M., Zauszniewski, J. A. (2014) 'Resourcefulness training intervention: A promising approach to improve mental health of mothers with technology-dependent children.' *Applied Nursing Research* 27, 87–90

Toly, V. B., Musil, C. M., Carl, J. C. (2012) 'Families with Children Who Are Technology Dependent: Normalization and Family Functioning.' *Western Journal of Nursing Research* 34(1) 52–71 DOI: 10.1177/0193945910389623

Trowbridge, K., Mische-Lawson, L. (2014) ‘Families with Children with Medical Complexity and Self-Management of Care: A Systematic Review of the Literature.’ *Social Work in Health Care*, 53 (7), 640-658, DOI:10.1080/00981389.2014.916776

Udo, K. (2005) ‘Make “emerge” or “force” the empirical data? A crucial problem of reconsidered theory.’

United Kingdom Central Council UKCC. (1994) cited in Sidey, A., Widdas, D. Eds (2005) *Textbook of community Children’s Nursing. Second Edition*. London. Elsevier

Wallis, C., Paton, J.Y., Beaton, S., Jardine, E. (2010) ‘Children on long-term ventilatory support: 10 years of progress.’[online] *Archives of Disease in Childhood*. Available from <http://adc.bmj.com/content/early/2010/11/24/adc.2010.192864.full.html> [25 November 2010]

Wang, C. H. Et Al. (2010) ‘Consensus Statement on Standard of Care for Congenital Muscular Dystrophies.’ *Child Neurology*. 25(12), 1559–1581

Wasserman, I. C., Mcnamee, S. (2010) ‘Promoting compassionate care with the older people: a relational imperative.’ *International Journal of Older People Nursing*, 5, 309–316 doi: 10.1111/j.1748-3743.2010.00252.x

While, A.E., Cockett, A. M., Lewis, S. (2003) ‘Children and Young People requiring Home Assisted Ventilation in the South of England: Incidence Receipt of Care Support and Components of Care Package’. *Children and Society* 18, 207-217

White, A., Bushin, N., Carpena-Mendez, F., Laoire, C. N. (2010) ‘Using Visual Methodologies to Explore Contemporary Irish Childhoods’ *Qualitative Research* 10 (2) 143 - 158

Whiting, L., Miller, S. (2008) ‘Traditional, alternative and innovative approaches to health promotion for children and young people’. *Paediatric Nursing* 21 (2), 45-50

Whiting, M. (2012) ‘Impact, meaning and need for help and support: The experience of parents caring for children with disabilities, life-limiting / life-threatening illness or technology dependence.’ *Journal of Child Health Care* 17(1), 92–108 DOI: 10.1177/1367493512447089

Whittemore, R., Knafl, K. (2005) *The integrative review: updated methodology. Methodological issues in nursing research*. USA. Blackwell Publishing

Whittemore, R. S., Chase, C., Mandle, L. (2001) ‘Validity in Qualitative Research.’ *Qualitative Health Research*, 11 (4), 522-537

Wilson, S., Morse, J. M., Penrod, J. (1998) ‘Absolute involvement: the experience of mothers of ventilator dependent children’*. Health & Social Care in the Community*, 6, 224–233

Wilson, W. (2018) ‘Disabled Facilities Grants for home adaptations. *House of Commons Library Briefing Paper*, Number 03011

Winfield, N.R., Barker, N.J., Turner, E.R., Quin GL. (2014) Non-pharmaceutical management of respiratory morbidity in children with severe global developmental delay.’ *Cochrane Database of Systematic Reviews*, 10. DOI: 10.1002/14651858.CD010382.pub2.

Wolfe, I., Macfarlane, A., Donkin, A., Marmot, M., Viner, R. (2014) *Why children die: death in infants, children and young people in the UK. Part A*. London, Royal College of Paediatrics and Child Health

World Health Organisation (WHO) 1998 cited in Mittelmark, M.B., Sagy, S., Eriksson, M., Bauer, G., Pelikan, J.M., Lindström, B., Espnes, G.A. (Eds.) 2017 *The Handbook of Salutogenesis*. Switzerland. Springer International Publishing

Youngblut, L. M., Brennan, P. F., Swegart, L. A. (1994) ‘Families with medically fragile children: a study. Pediatric Nursing 20, 463-468 cited in Sidey, A., Widdas, D. Eds (2005) ‘*Textbook of community Children’s Nursing*. Second Edition’. London. Elsevier

Appendices.

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Appendix 1. Long Term Ventilation Review Table

NAME OF STUDY & REF	Type of study Data collection methods **QUESTIONS CLEARLY DEFINED. (POP, INTERVENTION, COMARKERS, STUDY DESIGNS**	SEARCH STRATEGY	MINIMISATION BIAS	ASSESSMENT OF PRIMARY STUDIES	ANALYSIS	CONCLUSIONS
Children who require long-term ventilation: staff education and training Hewitt-Taylor, Jaqui Intensive & Critical Care Nursing, 2004, Vol.20(2), pp.93-102 **C 17**	Qualitative study Method -survey Questions clear Clearly defined population Focus and intervention clear Design robust	For literature not described	piloted	Comprehensive	Not identified	1. Education needs to consider psychosocial. 2. Education needs to look at reality for families. 3. Education should highlight needs of siblings.
Long-term ventilation in children: longitudinal trends and outcomes McDougall, Catherine M ; Adderley, Robert J ; Wensley, David F ; Seear, Michael D Archives of Disease in Childhood, 9 September 2013, Vol.98(9), p.660 [Peer Reviewed Journal] BMJ Publishing Group Ltd and Royal College of Paediatrics and Child Health **E 1**	Quantative study method cross sectional survey of data base search, clearly defined population	Search of data base of the only centre initiating ventilation in British Columbia	Use of same data collection process throughout the 15 years	Comprehensive capture of other large-scale prevalence studies	Used same definition of ventilation as UK studies	10-fold increase in 15 years 10-year survival rate 91% 42% discontinued ventilation at 10 years 26% transitioned 1. 19% of children tracheotomy ventilated 81%

						NIV,57% ventilated 24 hours per day 2. invasively ventilated children stable numbers NIV increasing Numbers now plateauing
Parental perceptions of quality of life in children on long-term ventilation at home as compared to enterostomy tubes Redouane, B. ; Cohen, E. ; Stephens, D. ; Keilty, K. ; Mouzaki, M. ; Narayanan, U. ; Moraes, T. ; Amin, R. PLoS ONE, February 2016, Vol.11(2) [Peer Reviewed Journal] **E3**	Quantative study 3 standardised quality of life measures Clearly defined questions	Not presented	Standardised tools 47 children and families 1 centre all patients who met criteria	comprehensive	statistical	Study found that Health related quality of life was low for both children on long term ventilation and those on tracheotomy tubes and lowest for those dependants on both
Current status of long-term ventilation of children in the United Kingdom: questionnaire survey Jardine, E; O' Toole, M; Paton, J Y; Wallis, C British Medical Journal, Jan 30, 1999, Vol.318(7179), p.295(1) [Peer Reviewed Journal] **E5**	Postal survey Quantative study Clear questions	Overview given	All paediatric centres	Literature cited not described or search technique described	statistical	Comparative now updated
Long-term follow-up of home mechanical ventilation in young children with spinal cord injury and neuromuscular conditions Gilgoff, Rachel L.; Gilgoff, Irene S.	Quantative Retrospective Survey 20 years 1 clinic looked at survival and readmission rates	One centres data base 20 years retrospective search in notes	One centre but was undertaken by a consultant involved in	None	Kaplin meier estimate of survival curve and mantel cox log rank statistics	1 .7 re hospitalisations per year Survival 84% at 5 years 71% at 10 years

The Journal of Pediatrics, May 2003, Vol.142(5), pp.476-480 [Peer Reviewed Journal] **E2**			care of all of the children			30/39 children attended school
Daily living with distress and enrichment. B2	Qualitative methodology Semi structured interviews and fieldwork within the family home Questions complex but clear	12 families recruited through Quebec program for home ventilator assistance	Process for data collection and analysis robust and described in detail Interviews took notes and audio taped discussion	Underpinning literature not discussed	Comparative analysis, to identify significant patterns similarities and differences Richard Zanner's interpretive framework	1-confronting parental responsibility 2- daily threat of death 3-seeking normality 4- living in isolation 5 -what about the voice of the child 6 -questioning moral order overwhelming 7-deep enrichments 8- stopping treatment or institutionalisation would affect both enrichment and distress 9- sibling envy 10- normalising home
Sending children home on tracheostomy dependent ventilation: pitfall and outcomes D3	Case review Identifying numbers around key questions Question clear	Case review of the 39 most specialist children discharged from 1 UK national	Robust process for data collection and presentation	Discussion of findings based on a wide look at associated literature	Formulation of a data set to log the child's journey to home and too consider against literature	1- 9.6 months to discharge. 2-7 died but not due to home care 3-15 were weened of ventilation

		specialist hospital				4 returning the child home doesn't end paediatric responsibility
D9 Family members experience of everyday life when a child is dependent on a ventilator. Linghi, Sweden	Qualitative metasynthesis Wide Question focused on standards and improving practice. Clear inclusion exclusion criteria	12 Qualitative studies 1998-2006 Clear criteria	Structured process based on recognized model of metasynthesis	Methodology choice and rational considered against the literature studies included/excluded against a clear rational	Themes sub themes concepts and phenomena of interest collected and then considered together pear review, research notes, direct quotes were al used to strengthen trustworthy	Themes 1 the risk of being excluded 2 attending school means interaction but brings about special needs 3 living in the company of a machine thoughts about health and being alive 4 experiences of being a sibling 5 experience of being a parent of a LTV child 6 on the meaning of time and place equipment Notes process of lifelong learning and the uniqueness of the child in that learning Notes risk of exclusion And importance of

						Siblings Family centred care
J Clin Nurs. 1999 Jul;8(4):440-50. The experiences and views of parents who care for ventilator-dependent children. Noyes J[1], Hartmann H, Samuels M, Southall D. UK **B11**	Qualitative study Questionnaires Interviews Review of medical and nursing notes. Questions not clearly identified	7 children diagnosed with CCHS	Triangulation of data	Thematic review of existing data	Content analysis	1-Effects of hospitalisation Battle to get home Different support packages Holidays difficulties in getting on holiday Views of professional staff
J Adv Nurs. 2007 Apr;58(1):1-10. Comparison of ventilator-dependent child reports of health-related quality of life with parent reports and normative populations. Noyes J[1]. UK **B12**	Quantative approach Quality of life tools. these were then compared to the scores achieved by mainstream children and families Clear questions and aims	Purposeful selection from across own population of long-term ventilated children	Standardised tool Pilot work to consider tool with this group of children Expert reference group oversite	Built on an earlier study	Standardised test administered with professional panel oversite. Small sample size	The LTV children had significantly lower scores than a generalised cohort Rises the importance of considering how to raise self esteem Comparison of parent's perceptions and scores of what their child felt compared with children's scores showed significant areas of discrepancy which has issues for parental proxy in these areas
BMC Health Serv Res. 2011 May 23;11:115. doi: 10.1186/1472-6963-11-115.	Qualitative research using a grounded their approach. Study took place in Norway	3 out of 11 of the families	Formal care givers only	Current literature in this area considered.	Careful consideration of power	Looks at whether a family accepts them or not linked to

Home mechanical ventilation and specialised health care in the community: Between a rock and a hard place. Dybwik K[1], Nielsen EW, Brinchmann BS. **B13**	Comes from the perspective of the formal care giver Focus groups were used to collect the data Clear aims and targeting of the study	studied had an LTV child the rest were adults Sampling was steered by the data and ceased at the point of theoretical saturation	Not described more widely	Generalised overview	differentials in the group	perceived competence Being a guest Who decides on care How much can we take pressures on home carers Challenges of family members being part of a care team Continuity of care and competence of carers 4
Children and Young People Requiring Home Assisted Ventilation in the South of England: Incidence, Receipt of Care Support and Components of the Care Package While, Alison E.; Cockett, Andrea M.; Lewis, Samantha *Children & Society*, v18 n3 p207-217 Jun 2004 **B 14**	Two false Qualitative study including postal questionnaire and telephone interview Clearly articulated questions	All trusts in the south west UK identified and key respondents approached	No overall information but a detailed breakdown of questions is included Identified that CCN directory used to identify nurses may have led to incomplete data set	Generalised overview	Not described	0 almost ¾ had a physical disability in addition to LTV 44 percent of care was delivered by CCN teams or hospital outreach teams 40% attended mainstream school

Voices and choices: young people who use assisted ventilation; their health and social care, and education Author: NOYES Jane Publisher: Stationery Office Publication year: 1999 Pagination: 105p.,list of orgs.,bibliog. Place of publication: London **C 1**	Large scale literature review including all studies identified using terms of ventilator, dependant, technology and medically fragile	Clearly identified	Identified the search didn't identify literature about siblings and wider family mostly due to a dearth of such literature at the time	Rigid review and acceptance criteria	Systematic search	P6 Helpful discussion of search strategies in this area P 10 risks of using LTV rather than the restrictions society imposes P19 importance in play of there being no surveillance Education of carers 28 Language that disables and oppresses 31 Children and families seeing provided services as a challenge rather than support
Absolute involvement: the experience of mothers of ventilator dependent children BMC Health Serv Res. 2011 May 23;11:115. doi: 10.1186/1472-6963-11-115. Home mechanical ventilation and specialised health care in the community: Between a rock and a hard place. Dybwik K[1], Nielsen EW, Brinchmann BS. **C20**	Clear question Qualitative study based on grounded theory Interviews over the phone	16 mothers of children attending a camp for ventilated children	No specific details identified except for a clear process	Good overview of the literature	Grounded theory Process clearly identified	1-model of absolute involvement p226 2- Table of strategies good p227 3- Learning proceeded teaching but trigger for learning not identified 4- 4 thankfulness and appreciation P 228 5- Reframing time 228

						6- To manage a consistent reality mothers had to confront each unacceptable behaviour 229 7- Infringing on the marital bedroom to get assistance 229 8- Being normal swimming 229 9- Having to do unpleasant tasks to child raised internal discord230 10- Increase in confidence when nurses became friends 11- Rules of engagement ways of reducing conflict

Appendix 2. Complex Care Review Table

NAME OF STUDY & REF	QUESTIONS CLEARLY DEFINED. (POP, INTERVENTION, COMARKERS, STUDY DESIGNS	SEARCH STRATEGY	MINIMISATION BIAS	ASSESSMENT OF PRIMARY STUDIES	ANALYSIS	CONCLUSIONS
E7 How families of children with complex care needs participate in everyday life Woodgate, Roberta Lynn ; Edwards, Marie ; Ripat, Jacquie Social Science & Medicine, November 2012, Vol.75(10), pp.1912-1920	Qualitative approach Ethnographic approach Question wide Population clear Longitudinal 18 months	1 major city in Canada 40 family's Snowballing technique	Mixed methods Use of photographs taken by parents to focus decision	Wide consideration of concepts	Isolated items and patens Organised patterns Identifying attributes Discovery of relations ships across the above to develop themes Mixed cohort of needs affects some findings	Meaningful participation Having a life Harnessing resources 1 p 1915 For Children to find participation meaningful there had to be choice they needed to feel safe and comfortable Risk is linked to choose 2 p 1915 Accessibility is essential p1916 Meaningful participation linked to having a life Contributing to society is attached to meaningful life Harnessing resources enabled access that is a skill and availability issue 1916 New abilities can link to increased need of support 1917
E14 "Fighting the system": Families caring for ventilator-dependent children and adults	Qualitative research using a grounded theory approach	Four specialist hospitals in Norway	In depth interviews	Short but focused and helpful summary	Open coding Selective coding	1 p2 isolation in own home The fight

with complex health care needs at home. (Research article) (Report) Dybwik, Knut ; Tollali, Terje ; Nielsen, Erik W. ; Brinchmann, Berit S. BMC Health Services Research, July 4, 2011, Vol.11, p.156	General description of aims				Grounded theory approach	Immense bureaucracy of the system Lack of interest or insight into the needs of families Setting limitations 2 p 4 Lack of competence and continuity- Loved one could die without it P4 3 to compensate for the lack of competence the families kept themselves readily available which increased there already heavy burdens p 6 6 4 lack of continuity of care 52 care givers in 2 years p4 5 Being indispensable Worth fighting for 7 issues the same for children and adults
E15 Supporting 'expert' parents--professional support and families caring for a child with complex health care needs in the community Kirk, Susan ; Glendinning, Caroline International journal of nursing studies, August 2002, Vol.39(6), pp.625-35	Qualitative study Clear questions	Family's Purposefully selected from 3 specialist hospitals professionals from across in and	Interviews with a guide system enabling participants freedom to lead the interview	Extensive and comprehensive	NUD*IST program, codes and categories	Working with expert parents Professional acknowledged parental expertise and most didn't feel threatened by it

		outpatient settings and commissioners				Some parents felt there were professionals who were challenged by this Support Three dimensions identified by parents and professionals Emotional, practical, information p629 1 Liked professionals who knew them and were easy to contact p629 2 Giving
E16 Nursing's contribution to research about parenting children with complex chronic conditions: An integrative review, 2002 to 2012 Rehm, Roberta S. Nursing Outlook, September-October 2013, Vol.61(5), pp.266-290	An Integrative review	PUBMED CINAHL	Search terms Chronic illness Medically fragile Medically complex Technology dependant,	review	Categories, synthetisation on cross study comparison	Major themes summarised on p285 1 2 notes the need to support parents emotionally, help parents work with health care providers, Need to build on strengths p 288 3
Being the Parent of a Ventilator-Assisted Child **E17** Lindahl, Berit ; Lindblad, Britt-Marie Journal of Family Nursing, 2013, Vol.19(4), pp.489-508 [Peer Reviewed Journal] jfn.sagepub.com	Qualitative study Phenomenological-hermeneutic model	5 couples caring for ventilated children in Sweden Contacted through two clinics	Clear process	Good overview	Based on ricour 1988 2 different researchers considering each text	Home care managers should foster compassionate and professional care to foster interconnectedness with the family

						1 p502 There should be training given to carers on that p504
E19 The quality of life of home-ventilated children and their primary caregivers plus the associated social and economic burdens: a prospective study Seear, Michael ; Kapur, Akshat ; Wensley, David ; Morrison, Kelly ; Behroozi, Ariana Archives of Disease in Childhood, 3 July 2016, Vol.101(7), p.620 [Peer Reviewed Journal] BMJ Publishing Group Ltd and Royal College of Paediatrics and Child Health	Qualitative study Clear questions	1 large LTV service	2 validated questionnaires Large cohort 90 children Interview in clinic Demographics Daily workload score Paediatric quality of life Paeds QL Caregiver impact scale Social and economic costs	Helpful overview		shifts that have had a profound effect on home care recognition that life can be happy with extreme disability, shift to home and portable technology.1 p620 use ¼ of primary care givers reported reduced quality of life 2 P623 3 overall 53.3% of all care givers reported health problems requiring daily medication mainly anxiety, depression, hypotension, arthritis 4 from the ventilated child's point of view ventilation is a successful strategy
E20 Paid carers' experiences of caring for mechanically ventilated children at home Maddox, Christina ; Pontin, David Journal of Child Health Care, 2013, Vol.17(2), pp.153-163	Qualitative study		Blaikie s (2009) abductive strategy Small study 3 carers Diaries Semi structured interviews		QSR Nvivo7 Recursive exploration across the data	Parents as experts 1 idiosyncratic practice from parents and a feeling of being judged P156 Role definition tensions Boundary tension child minding etc 3 157

						Carers developing meaningful Mixed emotions relationships with children and parents 4 Support mechanisms Importance of On call p159 Work needed on helping carers to share feelings and in bereavement support 5 160 Coercion to care for children in hospital? leading to coercion of carers to go outside boundaries in family home 6 Parental dependence on carers
E21 Technology-dependent children: The meaning of home care - a descriptive study de Lima, M.F. ; de Paulo, L.F. ; Higarashi, I.H. Online Brazilian Journal of Nursing, 2015, Vol.14(2), pp.178-189	Qualitative study Symbolic interactionalism	1 hospital PICU		limited	Content analysis	1 poor breaking of the diagnosis 2 lack of sibling and partner involvement P181 3 concerns about abilities when away from the hospital environment 182 4 the way news is broken shapes future interactions with health care professionals 184

Daily struggles: living with long-term childhood technology dependence Alexander, Eren ; Rennick, Janet ; Carnevale, Franco ; Davis, Michael Canadian Journal of Nursing Research, Dec 2002, Vol.34(4), pp.7-14 **E22**	Qualitative study Grounded theory	1 hospital PICU Quebec CCHS Neuromuscular conditions 5 children		limited	Content analysis	1 struggling daily- 2 getting over the hump 3Starting to breath Need for ongoing support4

Appendix 3. Technology Dependent Review Table

NAME OF STUDY & REF	QUESTIONS CLEARLY DEFINED. (POP, INTERVENTION, COMARKERS, STUDY DESIGNS	SEARCH STRATEGY	MINIMISATION BIAS	ASSESSMENT OF PRIMARY STUDIES	ANALYSIS	CONCLUSIONS
Families With Children Who Are Technology Dependent Toly, Valerie Boebel ; Musil, Carol M ; Carl, John C Western Journal of Nursing Research, 2012, Vol.34(1), pp.52-71 **E24 – full article**	Quantitive study Clear questions Clear population Well designed	Hospitals in Chicago USA	Piers-Harris children's self-concept scale School agers coping strategies inventory Child behaviour check list Family adaptability and cohesion evaluation scale	Yes, short but focused	SPSS version 19	Mothers of children with technological care needs are at high risk of clinical depression this puts family functioning at risk
E26 Resourcefulness training intervention: A promising approach to improve mental health of mothers with technology-dependent children Toly, Valerie Boebel ; Musil, Carol M. ; Zauszniewski, Jaclene A. Applied Nursing Research, February 2014, Vol.27(1), pp.87-90	Longitudinal randomised controlled Pilot trial Clear question 4 weeks of journaling	1 large Midwestern hospital in the USA	22 families Standardized tools for data collection Randomised control trial All Caucasian	Short but focused review	SPSS version 19 Not clear how interviews were analysed	Most commonly used RT skills- Really on families and friends Seek professionals and experts Organise activities Use positive self-talk P88 1 Journaling gave an outlet to express thoughts and feelingsP89 2 Reduction in negative emotions and depressive cognitions are believed to

						proceed depression they were reduced in this study P89 3
Parent or nurse? The experience of being the parent of a technology-dependent child Kirk, Susan ; Glendinning, Caroline ; Callery, Peter Journal of Advanced Nursing, Sep 2005, Vol.51(5), pp.456-464 **B6**	Qualitative grounded theory Clear question	3 specialist UK hospitals	24 children researcher sought parent to parent rapport	2 parts, 1 – LTV, 2 - Parenting	Consistent comparative method.	1. Transformed meaning of home pg 459 2. Home – public space pg. 459 3. Undertaking medical tasks on your own child pg 459 4.wanted to define themselves as parents not nurses pg 460 5. parents and nurses carrying out procedures but different meanings pg 461 6. exparental knowledge spot minute changes lacking in professional care pg 461 7. appropriate care, risks of conflict pg 462
Parents' Descriptions of Ideal Home Nursing Care for Their Technology-Dependent Children Mendes, Michele Pediatric Nursing, Mar/Apr 2013, Vol.39(2), pp.91-6 **E28**	Qualitative descriptive study Clear Questions	I home care agency North east USA	Small study 4 families 3 interviewed as a couple and one mother.	Short overview	Qualitative content analysis	Competence Technical competence Critical thinking skills A get going approach 1 P92 Caring “caring for the human being not just the body” “these kids depend on people to bring them some of the world” “being like a mum a companion or a friend”

						A caring positive manner help families cope P92-93 Parent control Parents expected nurses to acknowledge the parental expertise and expected nurses to respect their wishes Some parents felt how they interact with nurses affects care delivered so cautiously treated their nurses well P93 1 Fitting in Understanding the difference between hospital and home care Flexibility Boundaries Parental boundaries are important to set Organisation Consistency Considering the fit Being open to feedback or complaints p94 2 All noted the defining moment of the first meeting with a health care

						provider and how it affected subsequent relationships. 3 95 Urged early interventionists to ask families about past experiences with services and then adapt their interventions to family's aspirations quoting vecca and fenburg (2000)
E30 Factors Related to Depressive Symptoms in Mothers of Technology-DependentChildren Toly, Valerie Boebel ; Musil, Carol M Issues in mental health nursing, July 2015, Vol.36(7), pp.518-27	Quantative study using six instruments Clear Questions	A large Midwest hospital	Large study	Comprehensive targeted review of literature	SPSS 15.0	Noted that 40 % of Mothers had scores which would indicate they are at significant risk of depression 1 P523 2 factors identified to affect this normalisation and resourcefulness. Resourcefulness is the only one known to be responsive to psychological interventions 2 P253
Family members' experiences of everyday life when a child is dependent on a ventilator: a metasynthesis study Lindahl, Berit ; Lindblad, Britt-Marie Journal of family nursing, May 2011, Vol.17(2), pp.241-69	Qualitative metasynthesis	Sweden Clear process of review and audit trail	12 studies 1998 – 2006 inc Canada, UK and USA		Thematic review	Gap in LTV literature is family strengths and longitudinal research. See full list on front of article

B9						
E24 Dependent: normalisation and family functioning Toly, Valerie Boebel Journal of Pediatric Health Care, 2009, Vol.23(5), pp.e30-e31	Quantitive study Using 5 validated tools Clear Questions	A large Midwest Hospital USA	Large study 103 mothers	Comprehensive targeted Literature Review 67 Nurses and organising care issues	SPSS 15.0	38.9% of mothers at risk of depression. High levels of family nursing correlated with lower normalization so ether the presence of nursing care hinders this process or the nursing by targeting complexity is greater in at risk families 2 vp65 Mothers level of depressive symptoms is a predictor of family functioning3 p 66
Developing services to support parents caring for a technology-dependent child at home Kirk, S ; Glendinning, C Child: care, health and development, May 2004, Vol.30(3), pp.209-18; discussion 219 **B5**	Qualitative study	3 specialist hospitals in North England	24 children 25% vented Face to face at home interviews	Short targeted review	Constant comparative metod Glaser (1992)	I.E professionally neglected emotional aspects of learning pg 213 2.E Parent or Nurse pg 213 3.E cumbersome equipment restricted activity Needing trained babysitters Professionals threatened by their expertise Poor coordination.
Families' experiences of caring for technology-dependent children: a temporal perspective Heaton, Janet ; Noyes, Jane ; Sloper, Patricia ; Shah, Robina	Qualitative approach to gain a ??? perspective Clear aims	Hospitals and 1 hospice in North of England	Interviews with all family members Photos by participants	Good focused review	Framework analysis approach Richie and Spencer 1995	Rhythms and Routines E1. Pg 446 Social activities were adapted around social routines . Effects of care regime

Health & Social Care in the Community, Sep 2005, Vol.13(5), pp.441-450 **B10**			Timeline drawings			E3 Siblings giving , Domestic support, care of siblings and Technological care
E31 Home-care nurses as strangers in the family Coffman, Sherrilyn Western Journal of Nursing Research, 1997, Vol.19(1), p.82(15)	Qualitative Phenomenology Clear Question	Florida USA	10 nurses with a minimum of 6 months home care nursing experience Themes checked with participants.	Short Focused review	Focus on the phenomenon Holistic approach to text	1 P83 Take charge skills that work so well in hospital can produce failure in the home environment were protecting and developing the family is key 2 P83 focusing narrowly on the child short changes family's Overall theme- Stranger in the family "you re part of the family, but you are still a stranger"2 P86 "You are a guest" Advocating for the child Need to dress appropriately to the social situation you are attending with the child Shifts to fit family Primacy of the focus on the child can cause friction Bonding with the child To close and issues of objectivity arise Need to have high expectations for the child Blending with the family Maintain family boundaries

						Not crossing the line of parenthood Working as a team Holding a job-linked to American system 3 p 94 empowering the family is a Health promotion activity Conclusion!!
Absolute involvement: The experience of mothers of ventilator-dependent children Wilson, Sharon ; Wilson, Sharon ; Morse, Janice M. ; Morse, Janice M. ; Penrod, Janice ; Morse, Janice M. Health and Social Care in the Community, 1998, Vol.6(4), pp.224-233 **C20**	Qualitative study, grounded theory	Summer camps for vent children 14 vent	2 phone interviews	Short, targeted literature review	Grounded theory	Use of hope, use of support group, approaches to get what want persistent approach 3: 228 Risk of gifts 4: 229 Swimming 5:229 Destabilising nurse 7 Don't see care as their sole responsibility 8
E35 Families With Children With Medical Complexity and Self-Management of Care: A Systematic Review of the Literature Trowbridge, Kelly ; Mische-Lawson, Lisa Social Work in Health Care, 09 August 2014, Vol.53(7), p.640-658	Review of the Quantitive Literature Using a social work model to explore the literature to identify family strengths and self-management	Worldwide literature search	Systematic review		450 abstracts reduced to 31 and then 7 after in-depth review Then analysed using a social work framework	1 P642 social workers overwhelmed by the professional problem solving model. 2 P659 a number of studies demonstrate the degree of disability is not a predictor of family function 3 P650 problem solving approach positive or negative accounted for more than 20% of the

						variance of parental mental health 4 651 strong link in the literature to a deficit focus /however acknowledges it is there in the Qualitative literature! 5 P653 nots the focus on the psychological support but no focus on motivations and abilities of families 6 P654 a critique of the current complexity of tech equalling more issues for care giver 7 p655 positives of living with a child with complex care needs
E36 Caregivers' Experiences at Home With a Ventilator-Dependent Child Wang, Kai-Wei ; Barnard, Alan Qualitative Health Research, Apr 2008, Vol.18(4), p.501	Qualitative Phenominographic approach Clear Question	Australia	All data 17 families reviewed by two other researchers	Targeted short Literature review	7 research steps as described by Dahlgren and fallaburg 1991	It's a new world –being home - positive Ambiguous social identity-being parent and a carer The difficulty is having carers at home Social isolation-"not matter how my best friends are they don't Know and they don't understand" ,people stareing1P 505- Respite helps

						Positives-became stronger and more resilient 2 P505 3 demands and challenges of care giving were unexpected and stressful 3 P506 Intimidation and intrusiveness in the sanctuary of home 4 p506
Parenting a Technology Assisted Infant: Coping with Occupational Stress Murphy, Kathlene. Social Work in Health Care, 10 April 1997, Vol.24(3-4), p.113-126 [Peer Reviewed Journal] Taylor & Francis Group **Xxxxxxx but keep**	Article not research					
E37 Living with a machine: The experience of the child who is ventilator dependent Sarvey, Sharon I. ; Sarvey, Sharon I. Issues in Mental Health Nursing, February 2008, Vol.29(2), pp.179-196	Qualitative Phenomenological approach Clear aim to get a first-person experience of being ventilated	USA	8 children Participant review	Targeted short review	Hermeneutic analysis	at the start of life all humans have caregivers but in LTV this relationship can't change 1 p185 machine and person intermeshed 2 P186 human and mechanical other to exist 3 P186 "some of the nurses ...they are afraid..so they watch me all the time 4 P187 The others mechanical and human place restrictions 5 P187 I am still a person- the reaction of others 6 P 188

						- Underestimating cognitive ability Can/can't- theme Normal different theme Children's matter of fact approach and seeing life as a whole with passions and interests 7 P193 1 very negative view of being tied in the yard 8p194

Appendix 4. PowerPoint Presentation Describing Salutogenic Theory for the Launch

WellChild
the national charity for sick children

COFAR
Centre for Children and Families Applied Research

MAKING IT COUNT

South Warwickshire NHS
NHS Foundation Trust

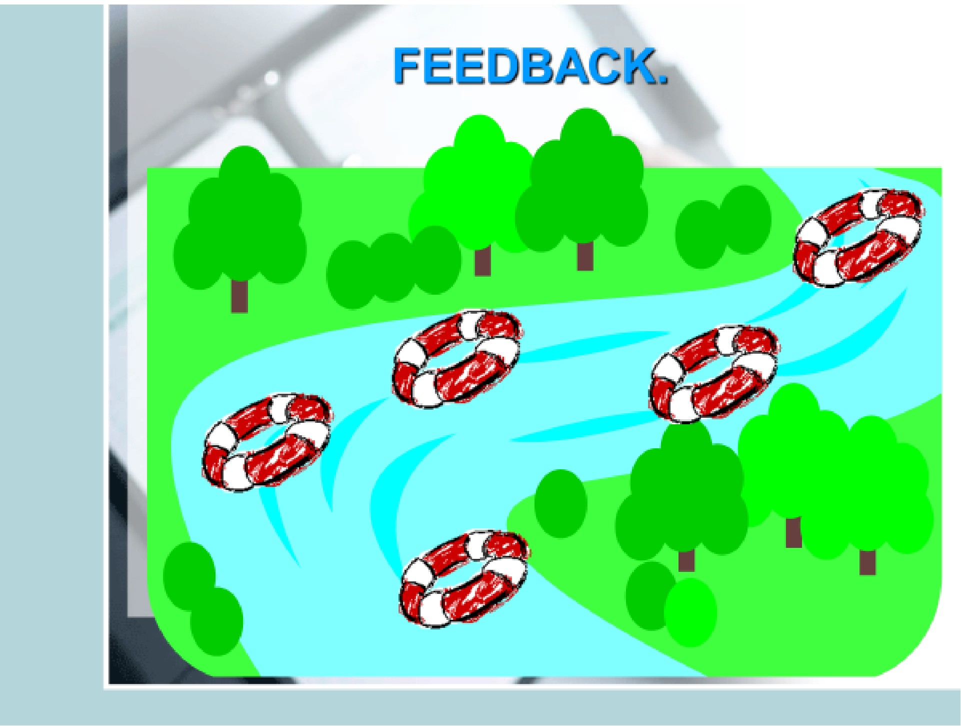
David Widdas
Andree Dignon
Jane Coad

Coventry University

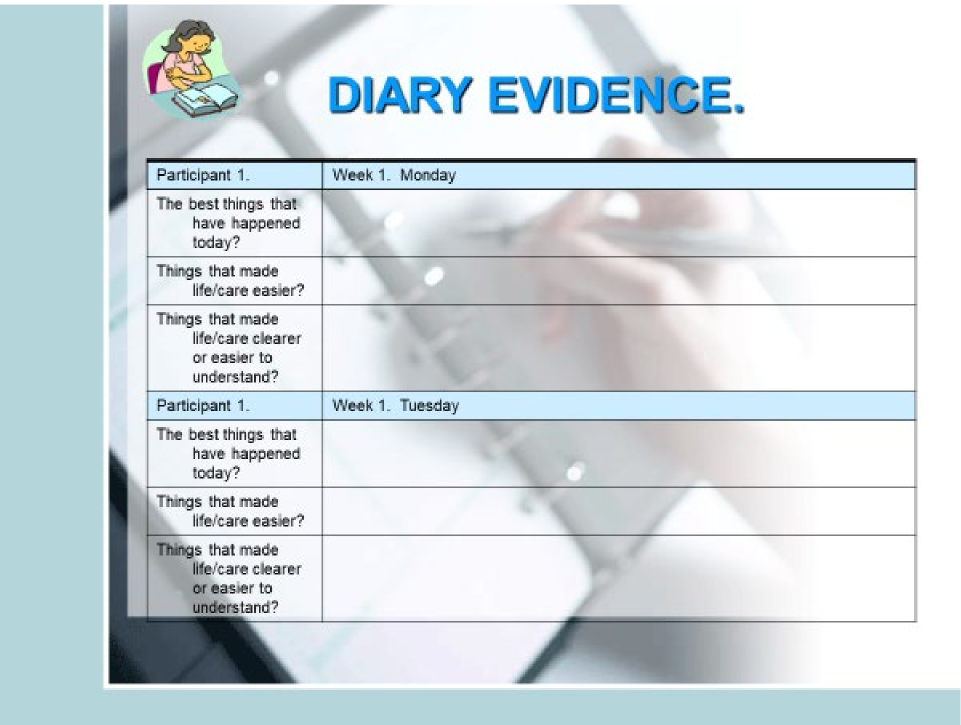
RIVER – MODERN MEDICINE.

ANTONOVSKY'S – ALL IN THE RIVER.

- The best things that have happened this today?
- Things that made life / care clearer or easier to understand this today?
- Things that have made life / care easier today?



FEEDBACK.

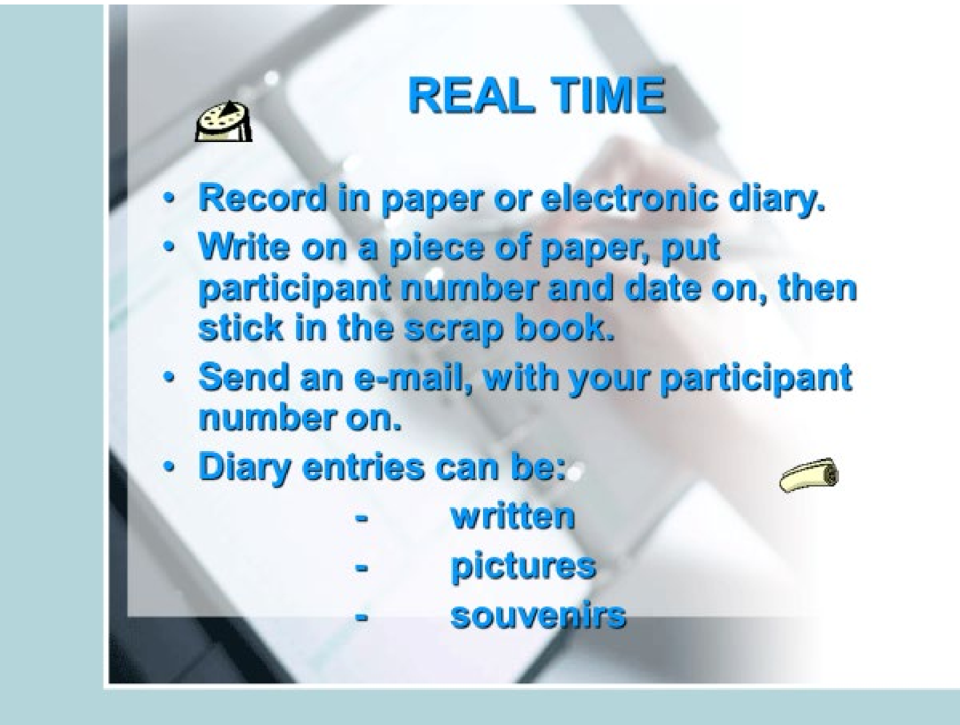


DIARY EVIDENCE.

Participant 1.	Week 1. Monday
The best things that have happened today?	
Things that made life/care easier?	
Things that made life/care clearer or easier to understand?	
Participant 1.	Week 1. Tuesday
The best things that have happened today?	
Things that made life/care easier?	
Things that made life/care clearer or easier to understand?	

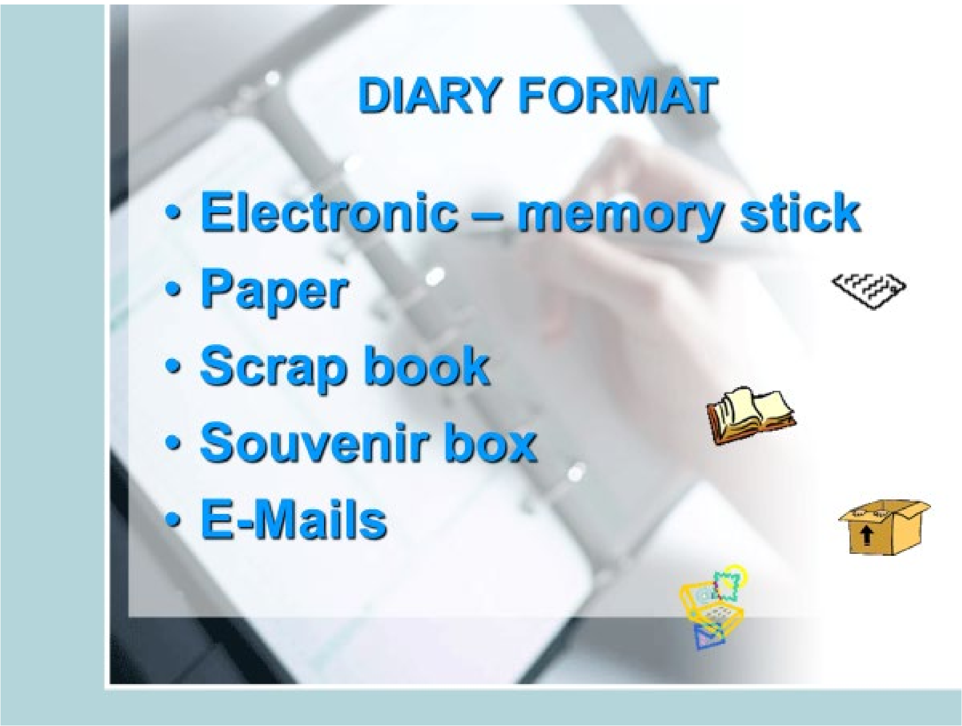
FAMILY DIARY

- Any family member.



REAL TIME

- Record in paper or electronic diary.
- Write on a piece of paper, put participant number and date on, then stick in the scrap book.
- Send an e-mail, with your participant number on.
- Diary entries can be:
  - written
  - pictures
  - souvenirs



DIARY FORMAT

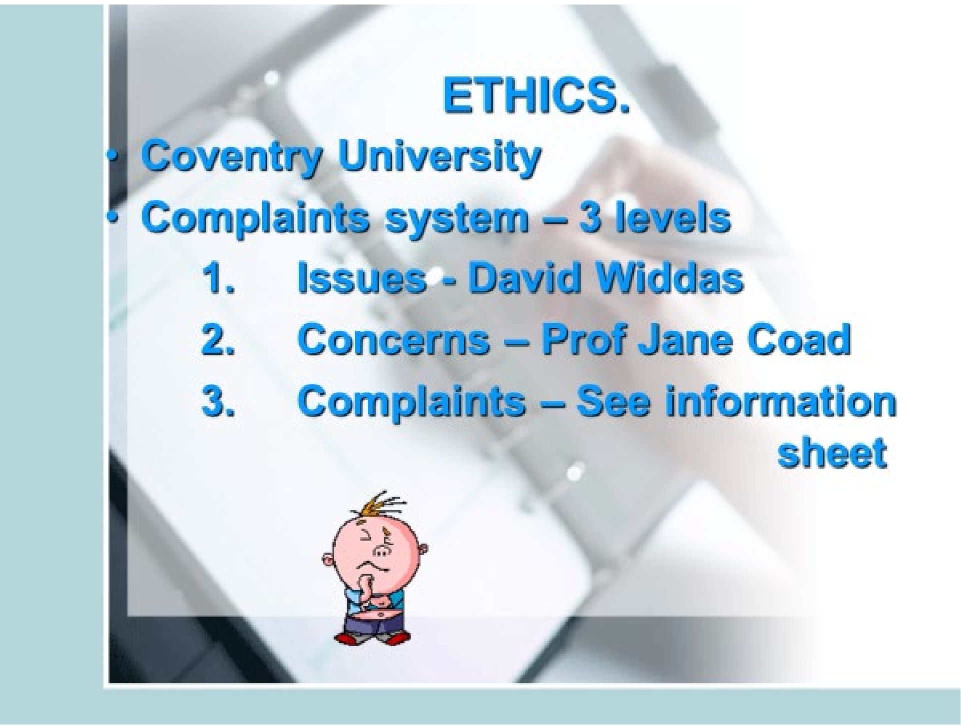
- Electronic – memory stick
- Paper
- Scrap book
- Souvenir box
- E-Mails

FINAL INTERVIEW

- Discuss diary entries, words, pictures, souvenirs.

DATA SECURITY

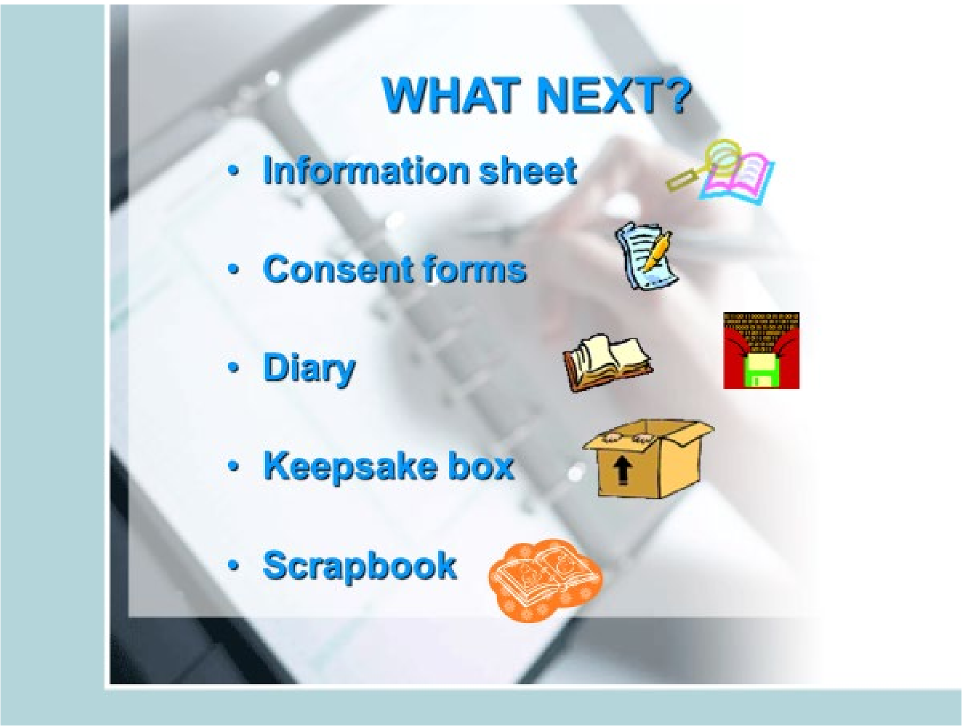
- Personal collection.
- Stored on NHS server.
- Made anonymous in writing up.
- NHS.net



ETHICS.

- Coventry University
- Complaints system – 3 levels
  1. Issues - David Widdas
  2. Concerns – Prof Jane Coad
  3. Complaints – See information sheet

Come Join Us



WHAT NEXT?

- Information sheet
- Consent forms
- Diary
- Keepsake box
- Scrapbook


THANK YOU

Appendix 5. Participant Information Sheet – Phase 1

INFORMATION SHEET

Making it Count.

Name of Lead Researcher: David Widdas

Supervisor: Prof Jane Coad

Introduction

Thank you for reading this information. You are being invited to take part in a research project about services for children and families who have children receiving long-term ventilation at home.

Before you make your decision, it is important for you to understand why the research is being undertaken and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. If anything is not clear to you or if you would like more information, then please feel free to ask David Widdas whose details are at the end of this information sheet.

What is the purpose of this research?

This research aims to identify and build on the factors that families believe make their child's care package work for them and their family.

Why have I been chosen?

We have been given permission by Linda as chair of the Congenital Central Hypoventilation Syndrome group to approach members within the group. As a parent of a child who requires ventilation, you have a unique insight into what makes care packages more effective for your child and the whole family.

What will happen to me if I take part?

If you do decide to take part, you should keep this information sheet as it provides details about the study.

You will be invited to keep a daily diary for one month which identifies the positive factors for you and your child and family.

There will also be an interview be after the diary keeping month to gain deeper understanding of your perspective of the positive factors that work for your child and family.

You do not have to take part in this research and can withdraw at any time without having to give a reason.

What are the possible disadvantages and risks of taking part?

We do not think that there are any disadvantages or significant risks for you taking part in this research as we are focusing on what works for you rather than issues and concerns.

What are the possible benefits of taking part?

We will publish the outcomes of this research with the aim of helping future families to benefit from packages of care developed on what families identify is effective and family focused.

What happens when the research study stops?

Once you have taken part, you will not need to do anything else. We will send you a short summary of the research study when it is finished.

Who do I contact if this study raises difficult issues for me?

For issues about how the research conducted please contact:

Will my taking part in this study be kept confidential?

All the information that is collected about you during the project will be kept strictly confidential and your name and other details will be removed in the final reports so that you cannot be recognised.

Who has reviewed the study?

This study has been reviewed by the Ethics Committee, at Coventry University.

Who can I contact for any more details?

Thank you very much for your time in reading this information leaflet

Appendix 6. Participant Information Sheet – Phase 2

Making it Count.

Name of Lead Researcher: David Widdas

Supervisor: Prof Jane Coad

Introduction

Thank you for reading this information. Your child/children are being invited to take part in the second part of a research project looking into services for children and families who have children receiving long-term ventilation at home.

Before you make your decision, it is important for you to understand why the research is being undertaken and what it will involve. Please take time to read the following information carefully and discuss it with others if you wish. If anything is not clear to you or if you would like more information, then please feel free to ask David Widdas whose details are at the end of this information sheet.

What is the purpose of this research?

This research aims to identify and build on the factors that families believe make their child's care package work for them and their family

Why have I been chosen?

We have been given permission by Linda as Chair of the Congenital Central Hypoventilation Syndrome Group to approach members within the group. The first phase of the research looked at the views of Parents and this phase looks at the views of children who receive ventilation and their brothers and sisters as they have a unique insight into what makes care packages more effective for your child and the whole family.

What will happen to me if I take part?

If you do decide to let your children take part in the study, you should keep this information sheet as it provides details about the study.

The children's play room, which is next to where the CHSG meeting is being held, will run play activities for the two days. Some of the play activities will be themed around the positive factors that help them manage, understand and motivate them around their condition. We will observe what is said in these sessions and keep notes. Any drawings or work created in these sessions will be photographed and given back to the child

Your Child/Children do not have to take part in this research; and if you choose for your child to take part you can withdraw them at any time without having to give a reason or the child can leave the session and re-join you in the CHSSG event.

Children who are not in the research as you, or they, have chosen not to participate in the research can take part in any of the activities; but no data will be collected about their involvement in the session.

What are the possible disadvantages and risks of taking part?

We do not think that there are any disadvantages or significant risks for you taking part in this research as we are focusing on what works for families rather than issues and concerns.

What are the possible benefits of taking part?

We will publish the outcomes of this research with the aim of helping future families to benefit from packages of care developed on what families identify is effective and family focused.

What happens when the research study stops?

Once you have taken part, you will not need to do anything else. We will send you a short summary of the research study when it is finished.

What if something goes wrong and I want to make a complaint?

Some materials have been removed from this thesis due to Third Party Copyright. Pages where material has been removed are clearly marked in the electronic version. The unabridged version of the thesis can be viewed at the Lanchester Library, Coventry University

Will my taking part in this study be kept confidential?

All the information that is collected about your child during the project will be kept strictly confidential and their name and other details will be removed in the final reports so that you cannot be recognised.

Who has reviewed the study?

This study has been reviewed by the Ethics Committee, at Coventry University.

Some materials have been removed from this thesis due to Third Party Copyright. Pages where material has been removed are clearly marked in the electronic version. The unabridged version of the thesis can be viewed at the Lanchester Library, Coventry University

Thank you very much for your time in reading this information leaflet

Appendix 7. Participant Consent Form

Making it Count: can gathering and studying children and family narratives using a salutogenic based approach, help refocus children's complex care packages in a health enhancing way?

Name of Lead Researcher: David Widdas

Supervisors: Prof Jane Coad, Andree Digon

Consent form **[Parents; Young people over 16 competent to read]**

Participant Reference Code:	Please initial box
I have read the information sheet	
I know that I do not have to take part and that if I do take part I can withdraw at any time without having to give a reason.	
I agree to take part in the study as set out in the information sheet	
I understand that all data collected will be kept securely and no one apart from David Widdas will access them. It will be kept on a password protected, secure NHS server.	
I agree to the audio recording of post diary interview. I understand this data will be downloaded onto an NHS server and destroyed after transcription. The only people to hear this recording will be David Widdas and the transcriber.	
In the final report I understand that all personal names will be removed I understand you will do the same about reports from the study.	
I understand that all hard copies will be kept in a locked filing cabinet until they are destroyed at the end of 2017.	

Name of participant	Signature	Date
Name of Researcher	Signature	Date

1copy for participant/1 copy for researcher

Appendix 8. Description from the Literature of Antonovsky's Three Concepts

Comprehensibility.

Believe the challenge is understood (Antonovsky 1996).
The cognitive component (Lindstrom, Eriksson 2006).
Things are potentially understandable (Hintermair 2004).
Predictability (Londal 2010).
Consistency, structure, clarity (Flensborg-Madsen, Ventegodt, Merrick 2005).
Sensible (Sivberg 2002).
Making sense of what is happening (Stokes 2010).
Do you feel that you can understand things, that things make sense and are not confusing? (Mattiuzzi 2007).
Do you feel things are predictable or expected in other words do you feel like you know what going to happen next, or that you know what is coming? (Mattiuzzi 2007).
Can you tell me about things today that have helped to make things more understandable, sensible, clear, less confusing, or predictable for you or your family?

Manageability.

Believe the resources to cope are available (Antonovsky 1996).
The instrumental or behavioural component (Lindstrom, Eriksson 2006).
Problems can be solved either with their own resources or with the help of others (Hintermair 2004).
Good load balance (Londal 2010).
Coper (Kandel, Merrick 2007).
Using coping strategies flexibly (Oelofsen, Richardson 2006).
Influenced confidence that the resources available are adequate (Stokes 2010).
Do you feel that things are manageable or within your control, that things can be handled or taken care of? (Mattiuzzi 2007).
Do you feel you have the skills or ability, the support, help or the resources necessary to take care of things (Mattiuzzi 2007)?

Meaningfulness

Wish to be motivated to cope (Antonovsky 1996).
The motivational component (Lindstrom, Eriksson 2006).
Aims worth striving for (Hintermair 2004).
Participation in shaping outcomes (Londal 2010).
Make sense, worth the investment (Flensborg-Madsen, Ventegodt, Merrick 2005).
Motivation.
The world is conceived to make emotional sense.
Problems are worthy of commitment.
Challenges are accepted (Sivberg 2002).
Increased motivation to engage in challenges faced (Stokes 2010).
Do you feel that things are interesting or fascinating, a source of pleasure or satisfaction? (Mattiuzzi 2007).
Do you feel that things are really worth it, that there is good reason or purpose to care about what happens? (Mattiuzzi 2007).

Appendix 9. Diary System User's Guide

Coventry University CCFAR Centre for Children and Families Applied Research South Warwickshire NHS NHS Foundation Trust WellChild the national charity for sick children

Diary System Users Guide.

Thank you for taking part in this research study.

The aim of this Coventry University study is to learn from families skilled in the care of their children’s long-term ventilation needs. The study aims to collect what works for you as a family. The diary/scrapbook entries can be anything that you think is appropriate; it does not need to directly relate to long-term ventilation issues. What we are trying to collect is what families think is important in their day-to-day lives.

You will have a choice of using an electronic diary or a paper version. If possible, I would be grateful if you could use the electronic version (however if this is not practical for you then please feel free to use the paper version.)

Both versions of the diary ask three questions:

The best things that have happened today?

Things that made life/care easier?

Things that made life/care clearer or easier to understand?

The idea of using the diary is to collect some real-time thoughts and feelings about what is going right for your child and family, or what makes life easier to cope with, or what makes life/care easier to understand. At the end of the month the researcher will come and talk to you about what is in the diaries/ scrapbook but also to get the wider context about the three questions which can include your experiences from diagnosis.

The diaries/scrapbooks are family documents, so we are happy for any member of the family to contribute to them. If you wish to put in photographs or if the children wish to put in drawings in, please do so. Could you please date your drawing on the back with the day of the week and which week it was and your participant number. You can either insert them into the electronic or paper diary or you can put them in the scrapbook that will be provided. The team will discuss them at the end of the month when one of the researchers comes to collect them. If there is a bigger souvenir you wish to discuss, pop it in the box and we will discuss it at the final interview

The diaries and scrapbooks and box will remain your property and once the research team have transcribed them they will be returned to you.

We are really keen to catch thoughts and events as they occur to you, so if you don't have a computer or the diary handy just jot your thoughts on a piece of paper with the date written on it and then you can stick it into the scrapbook. If you have your computer or Smartphone on send me an email to david.widdas@nhs.net

We appreciate you may not have something to write draw or say about each of the questions every day but if you can contribute something related to them each day the team will be very grateful.

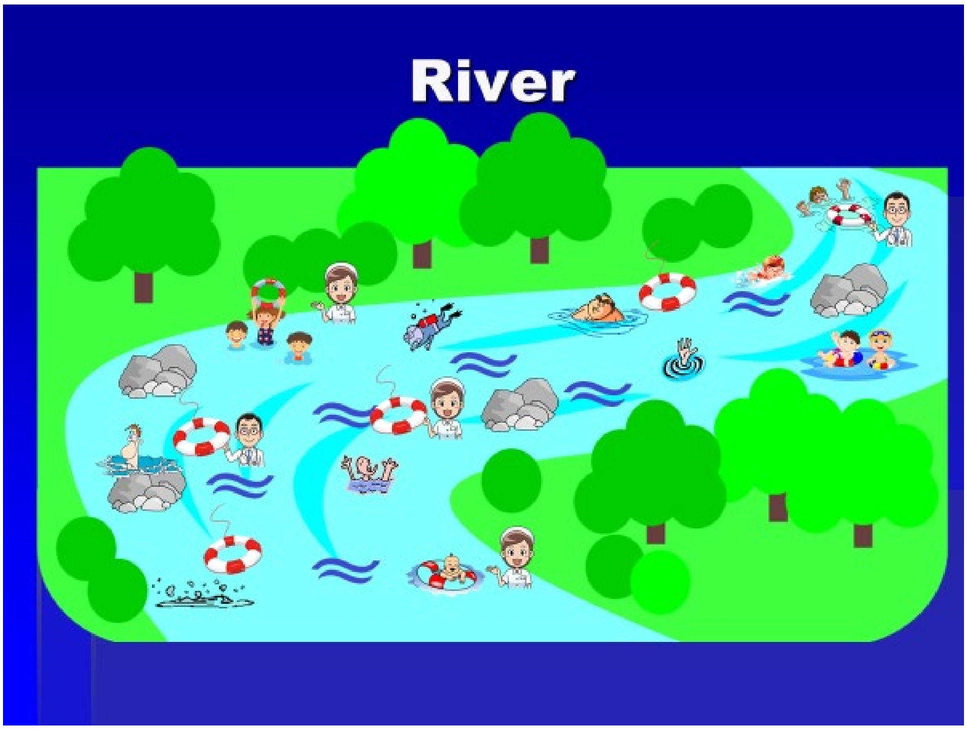
If you do choose to e-mail me information NHS.net is a secure e-mail system more information is available from www.connectingforhealth.nhs.uk/systemsandservices/nhsmail/using.

The study has full ethical approval from Coventry University Ethical Committee.

Appendix 10. Children's Participatory Scenario PowerPoint Presentation

Children’s Play Room Research Activities

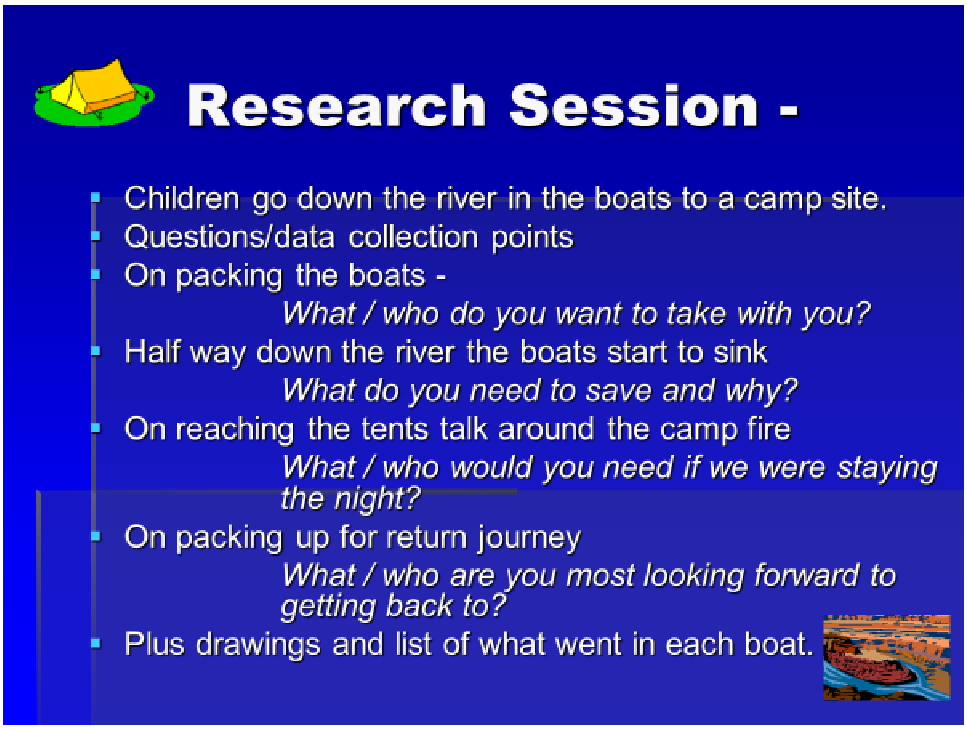
David Widdas
September 2014



River

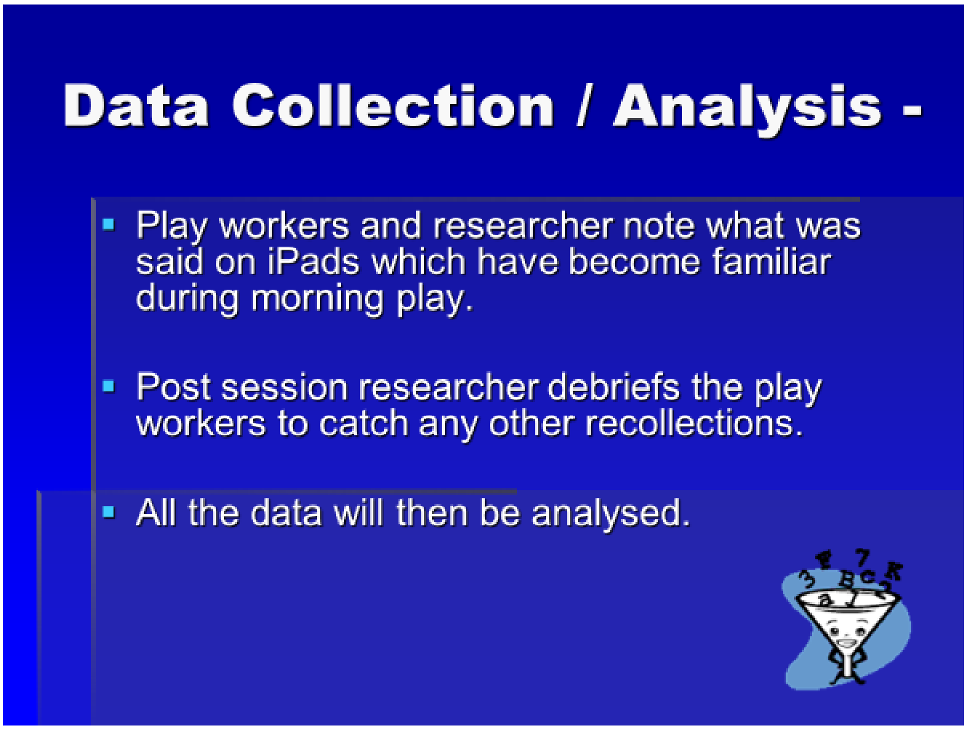
Children's Fun Session -

- Based on Anthronosky's river analogy.
- Morning general play to get settled plus making boats for the afternoon session.
- Lunch consent forms.



Research Session -

- Children go down the river in the boats to a camp site.
- Questions/data collection points
- On packing the boats -
  What / who do you want to take with you?
- Half way down the river the boats start to sink
  What do you need to save and why?
- On reaching the tents talk around the camp fire
  What / who would you need if we were staying the night?
- On packing up for return journey
  What / who are you most looking forward to getting back to?
- Plus drawings and list of what went in each boat.



Data Collection / Analysis -

- Play workers and researcher note what was said on iPads which have become familiar during morning play.
- Post session researcher debriefs the play workers to catch any other recollections.
- All the data will then be analysed.

Appendix 11. Warwickshire Children’s Community Nursing Assessment Tool

Appendix 12. Warwickshire Children's Community Nursing Delivery Model

Appendix 13. Quality Approaches used in Each Method (Flick 2009)

Quality Approaches used in Each Method (Flick 2009)

Methods – Arts Based	Quality Approaches Areas Reviewed
Diaries.	• Philosophical suitability. • In-depth review of the historical development of the method consideration of literature on review of method. • Design considerations including format electronic / written. • Review potential literacy issues. • Data security standards. • Formulation of questions. • Links to aim and research questions. • Formulation, understandability of questions-parental review and (appendix 8, Chapter 3 research diary and supervision notes). • Link to theoretical frameworks. • Launch approach. • User guide (Appendix 9) • Effectiveness review – Adult / Child • Lessons learnt. • Learning for next steps.
Interviews.	• Philosophical suitability. • Consideration of underpinning literature. • Consideration of approach, structures, unstructured or semi-structured chosen. • Issues around place. • Ethical considerations. • Positionality of researcher. • Issues of choice and control (user choice of diary entries to discuss). • Effectiveness review – Adult / Child. Lessons learnt. • Learning for next steps.
Focus Groups.	• Philosophical suitability. • Consideration of underpinning literature. • Consideration of approach. • Issues of power and representiveness. • Role in study – member checking. • Effectiveness review. • Lessons learnt.
Overall consideration of arts-based methods in children.	• Philosophical suitability. • Approaches – Arts informed, Arts based. • Power.

	• Nature of children's research.
Scrapbooks.	• In-depth history. • Underpinning Literature. • Design considerations. • Electronic – word, social media. • Health care use. • Copying of scrapbook – issues of format. • Launch approach. • Effectiveness for Children. • Review of Lack of success of Approach. • Lessons learnt. • Learning for next steps.
Memento boxes.	• Philosophical suitability. • Underpinning Literature. • Consideration of approaches. • Review of use in research. • Review of use in healthcare. • Ethics of approach. • Design considerations. • Suitability for children. • Collecting data transition. • Artefact to data transfer issues. • Launch approach. • Review of Lack of Success of Approach. • Lessons learnt. • Learning for next steps.
Participatory play-based scenarios.	• Philosophical suitability. • Exploration of literature around vignette research. • Relevance to 'real life'. • Approach of scenario – life like / unusual. • Creation of new approach considerations. • Launch considerations. • Skill set of researcher. • Data collection approach. • Interaction of research assistants. • Training of research assistants. • Review of method. • Review of data collection. • Overall review of approach. • Lessons learnt.