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Cardio-Respiratory Sleep Studies at Home: Experience in Research and Clinical Cohorts

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1. Home
2. Domiciliary
3. Obstructive sleep apnoea
4. Sleep-disordered breathing
5. Cardiorespiratory polygraphy
6. Screening

Abbreviations

AHI	Apnoea/hypopnoea Index
CI	Confidence Interval
CRPG	Cardiorespiratory polygraphy
EEG	Electro-encephalogram
NIV	Non-invasive ventilation

ABSTRACT

Objective: To evaluate the success rates of home cardiorespiratory polygraphy in children under investigation for sleep-disordered breathing and parent perspectives on equipment use at home.

Design: Prospective observational study

Setting: Sheffield, Evelina London and Southampton Children's Hospitals.

Patients: Data are reported for 194 research participants with Down syndrome, aged 0.5-5.9 years across the three centres and 61 clinical patients aged 0.4-19.5 years from one centre, all of whom had home cardiorespiratory polygraphy including respiratory movements, nasal pressure, pulse oximetry, position and motion.

Main outcome measures: Percentage of home cardiorespiratory studies successfully acquiring ≥ 4 hours of artefact-free data at the first attempt. Parental report of ease of use of equipment and preparedness to repeat home diagnostics in the future.

Results: 143/194 (74%; 95%CI [67%, 79%]) of research participants and 50/61 (82%; 95%CI [71%, 90%]) of clinical patients had successful home cardiorespiratory polygraphy at the first attempt. Some children required multiple attempts to achieve a successful study. Overall this equated to 1.3 studies per research participant and 1.2 studies per clinical child. The median artefact-free sleep time for successful research studies was 515 minutes (range 261-673) and for clinical studies 442 minutes (range 291-583). 84% of research and 87% of clinical parents expressed willingness to repeat home cardiorespiratory polygraphy in the future. 67% of research parents found the equipment 'easy or okay' to use, while 64% of clinical parents reported it as 'easy' or 'very easy'.

Conclusions: Home cardiorespiratory polygraphy offers an acceptable approach to the assessment of sleep-disordered breathing in children.

What is already known on this topic

1. Home based cardiorespiratory polygraphy (sleep studies) have been proposed as a viable alternative to in-hospital studies but there are limited reports in children

What this study adds

1. Parents report that home sleep studies are an acceptable alternative to in-hospital studies in children with diverse comorbidities
2. On average 76% of children had successful home CRPG studies on the first attempt and 87% after one or more repeat studies
3. While the standard for a successful study was more than 4 hours of artefact-free data, overall 83% of studies acquired more than 6 hours

INTRODUCTION

Sleep-disordered breathing, an umbrella term for conditions that result in disturbed ventilation during sleep¹, can broadly be classified as being either obstructive or central in nature. Obstructive sleep apnoea is characterised by intermittent collapse of the upper airway, and central sleep apnoea by repetitive complete cessation of respiratory effort, during sleep.

Obstructive sleep apnoea, the commonest cause of sleep-disordered breathing, peaks during the pre-school years in association with adenotonsillar hypertrophy and again during adolescence with obesity. Demand on diagnostic services has arisen as a result of increased understanding that a wide range of paediatric conditions predispose to sleep-disordered breathing².

The international gold standard investigation for sleep-disordered breathing is polysomnography which combines cardiorespiratory and neurophysiological sensors³. Polysomnography provides the most accurate estimate of hypopnoea (partial reduction in airflow) as these events are only scored when associated with either oxygen desaturation or an EEG (electro-encephalogram) arousal⁴. However, polysomnography is not universally available and requires considerable technical expertise to set up and interpret. Cardiorespiratory polygraphy (CRPG), which excludes neurophysiological measures, provides a recognised alternative in adults^{5,6,7} and, is increasingly reported in children^{8,9,10,11}. Paediatric CRPG has adequate sensitivity (90.9%) and specificity (94.1%) for the diagnosis of clinically significant obstructive sleep apnoea¹². While the European Respiratory Society Taskforce identifies polysomnography as the preferred diagnostic method, it recognises CRPG as an alternative where resources are limited¹³. As noted by a leader in the field 'the times they are a changing'¹⁴.

The introduction of miniaturised devices means that CRPG is now feasible in the home environment^{15,16} and the scope to evaluate more 'efficient ways of diagnosing sleep-disordered breathing^{17,18} is paramount. We report lessons learned from research and clinical experience of home CRPG in 255 children with diverse comorbidities.

MATERIALS AND METHODS

Study Subjects

Setting

Research: Participants were recruited at Sheffield, Evelina London and Southampton Children's Hospitals.

Clinical: Data from a new home CRPG service in Southampton Children's Hospital

Eligibility criteria

Research: Children had a diagnosis of Down syndrome , were aged six months to six years, had not had a CRPG in the last 3 months and were not receiving home oxygen or NIV (non-invasive ventilation).

Clinical: Children were referred to the clinical service with suspected sleep-disordered breathing, or for a ventilation titration study. Families chose either in-patient or home CRPG. Exclusions included families not conversant in English or children requiring other hospital-based investigations contemporaneously.

Recruitment / Selection

Research: Children were recruited between 2013 and 2015 through community and hospital routes to reduce selection bias¹⁹. Families were actively encouraged to attempt home CRPG as the preferred setting if deemed appropriate by the clinician. However, if parents

expressed a preference for an in-hospital study then the CRPG was carried out on identical equipment in the sleep laboratory.

Clinical: Eligible families referred to the clinical service self-selected either in-patient or home CRPG between 2015 and 2016. Only clinical patients who underwent home CRPG are reported here.

Ethics and consent

The research study was approved by the UK National Research Ethics Committee (reference-13/SC/0106). Parents provided written consent on behalf of their child. The clinical patients were offered home CRPG as a new clinical pathway and their anonymous data reported in accordance with UK Department of Health guidance for research ethics²⁰ as part of a service evaluation.

Methods

Demographics and medical history

Research: Data were recorded on age, gender, and socio-economic status (parents' age at leaving full-time education) and age-appropriate sleep questionnaires^{21,22} reported whether the child had restless sleep.

Clinical: Data on age, gender, underlying comorbidity and past experience of CRPG were recorded.

Home cardiorespiratory polygraphy

For all children, sleep-disordered breathing was assessed using the SOMNOtouch device (SOMNOmedics, Germany) comprising: chest and abdominal respiratory inductance plethysmography; pulse oximetry (Bluepoint) yielding saturations (SpO₂), plethysmography and pulse rate; nasal pressure flow with integral snore sensor; body position sensor; and

actimetry. In addition, for the clinical cohort routine contemporaneous pulse oximetry (Masimo Inc., USA) and transcutaneous carbon dioxide (Sen Tec, Switzerland) monitoring was undertaken and a subgroup also had video monitoring. For study failures, families were given the opportunity to repeat the CRPG.

CRPG equipment training

Families attended the hospital on a single occasion and were taught how to set up and use the CRPG equipment. Written and photographic instructions were provided. The abdominal and thoracic bands were measured on the child to minimise parental adjustments later. Parents set up their own children that evening at home. Telephone advice was offered until 23:00hrs. The CRPG device was programmed to auto record at a predetermined start time or was started manually. The equipment was returned by next day courier to the hospital (research) or by the parent (clinical) for data download and analysis.

Sleep log

Parents recorded the time their child settled in bed, fell asleep and woke in the morning alongside timing and duration of night wakings.

Quality standards and analysis

Cardiorespiratory polygraphy

A detailed scoring procedure is published¹⁹. Studies were manually scored by experienced clinical physiologists (RK, JG) using Domino Light software (SOMNOmedics, Germany). Sleep logs, actimetry, heart rate and breathing pattern were used to classify sleep and wake for each 30-second epoch. Respiratory events were scored per standard paediatric scoring criteria for adapted sensors⁴.

Success rates of CRPG

Studies with ≥ 4 hrs of interpretable estimated sleep data and artefactfree respiratory parameter data^{15,16} were deemed successful.

Evaluation of Home Polygraphy

Research: Parent feedback was sought 3 months after the CRPG by structured telephone interview. Parents were asked to describe their experience of using CRPG equipment at home with response options: '*Easy*', '*OK*' or '*difficult*' and how they would feel about a home study in the future with the response options: '*happy to repeat*', '*uncertain*' or '*unhappy to repeat*'.

Clinical: Parents completed a next day service evaluation form reporting ease of use of the equipment on a 5-point Likert scale from '*very easy*' to '*very difficult*' and preference for future CRPG study location.

Statistical analysis

The primary outcome measure was the percentage of children that had a successful first home CRPG. The secondary outcome measure was the acceptability (ease of use and willingness to repeat at home in the future) of home CRPG to the caregivers. Data were analysed in SPSS v24 (IBM). Descriptive statistics are presented. Demographic differences between children who had a successful study at first attempt and those in whom the first attempt failed were explored with Chi squared or Fisher's exact test for categorical data, age was non-normally distributed amongst clinical and research children whose studies failed so group differences were explored with the Mann Whitney-U test. 95% confidence intervals (CI) around percentages achieving successful home CRPG were calculated using Confidence Interval Analysis.

RESULTS

Baseline Characteristics

Research: Of the 202 children with Down syndrome where families consented to participate in the research,¹⁹ 194 agreed to home CRPG. Median age (range) was 3.0yrs (0.5-5.9), 53.1% were male.

Clinical: This group comprised 61 patients typical of referrals to a tertiary respiratory diagnostic service. Median age (range) was 7.8yrs (0.4-19.5), 55.7% were male. The majority (77%) had comorbidities (Table 1).

Success Rates of CRPG

Research: 143/194 (74%; 95%CI [67%, 79%]) had successful home CRPG studies on the first attempt. There were no differences between centres: Southampton 58/81(72%); Sheffield 39/54(72%); London 46/59(78%). Of the 51/194 (26%) failures at first attempt, 31 were willing to have a second attempt at home, of which 25/31 (81%) were successful. Two families were willing to have a third home attempt and one was successful. In total $143+25+1=169/194$ children attempting home studies ultimately achieved a successful home CRPG study (87%; 95%CI [82%, 91%]), requiring an average of 1.3 attempts.

Clinical: A total of 50/61 (82%; 95%CI (71%, 90%)) children had successful home CRPG studies at the first attempt. There were no differences in success rates between typically developing (78%) and non-typically developing children (84%). Of the 11 unsuccessful studies, 5 were repeated as inpatient CRPG, in 3 cases a clinical decision was made based on oximetry data and 3 were successfully repeated at home. Overall therefore 53/61 (87%;

95%CI [76%, 93%]) children were successfully investigated in the home setting (requiring an average of 1.2 home attempts needed for each successful study).

Reasons for failed studies

Research: reasons included equipment failure (15%); sensors not tolerated (30%); sensors removed before 4hrs of artefact-free data were captured (43%) and no reason recorded for 13%. The equipment was new to the market and, with support from the manufacturer, technical problems were resolved early in the study.

Clinical: reasons included sensors not being tolerated (21%); or sensors removed before 4hrs of artefact-free data was captured (79%).

Characteristics of children where studies succeeded at first attempt.

Research: There were no differences in success rates based on age, gender, socio-economic status, or whether the children were usually restless sleepers.

Clinical: Success rates at first attempt did not differ by age, gender or experience of CRPG. Table 2a and Table 2b illustrate demographic data for the success and failure of the first attempt at CRPG.

Quality Control

Estimating sleep time

Research: Of the 169 successful home CPRG studies, most achieved well above the minimum 4 hours (240 minutes) of artefact-free data: median 515 (range 261–673) minutes; 87% achieved ≥ 6 hours of artefact-free estimated sleep time; 78% achieved ≥ 7 hours; 62% achieved ≥ 8 hours.

Clinical: For the clinical data the median duration of artefact-free data was 442; (range 291–583) minutes; 83% achieved ≥ 6 hours of artefact-free estimated sleep time; 60% achieved ≥ 7 hours; 38% achieved ≥ 8 hours.

Acceptability Measures

Research: 165 of the 194 families (85%) were successfully contacted. 67% reported that the CRPG experience was ‘easy’ or ‘okay’ while 33% reported that they found the experience difficult. Nonetheless a majority (84%) stated they would be happy to repeat home CRPG in the future.

Clinical: Feedback from 45/61 families was provided the morning after CRPG before success of the study was determined. In the case of multiple studies only the feedback after the first study was evaluated. 29/45 (64%) found the equipment ‘easy’ or ‘very easy’ to use, 31% found it ‘okay’ and 4% difficult. Only 23 families responded to the question about future preferences. 20/23 (87%) stated they would prefer home CRPG in the future.

Tables 2a and 2b illustrate acceptability data for the success and failure of the first attempt at CRPG.

DISCUSSION

In this large sample of 255 children, 193 (76%) achieved a successful home multi-channel cardiorespiratory study at the first attempt and 222 (87%) were successful when including repeat attempts. Overall an average of 1.2-1.3 home studies per child was required to achieve adequate data. This is encouraging, particularly as the largest group were young children with Down syndrome who are often restless sleepers²³ and can be challenging to study. These data suggest that home CRPG is feasible and offers a realistic option for diagnostic testing in children.

Aside from national drivers to reduce bed occupancy and cost there is a key central argument for offering home CRPG, namely that home studies may achieve better sleep quality than in hospital^{9,24}. Knowledge about normal sleep architecture predicts that rapid eye movement sleep (when children are particularly vulnerable to sleep-disordered breathing) occurs predominantly towards the end of the night (Figure 1). Thus, a short night's sleep may under-estimate sleep-disordered breathing. Although a minimum criterion of 4 hours of interpretable signals is often quoted for reporting paediatric CRPG studies¹⁵, studies of this duration may miss significant sleep-disordered breathing at the end of the night. Our scoring criteria required this minimum period of sleep time to include only artefact-free data. The median estimated artefact-free sleep time was 8.6hrs in the Down syndrome research group and 7.4hrs in the clinical group and thus it is assumed that rapid eye movement sleep would have been captured during multiple sleep cycles.

Studies were unsuccessful for several reasons. Equipment failure was a feature of early studies as the SOMNOtouch device was newly available and staff were inexperienced in its use. Children sometimes did not tolerate sensors and removed them during the night; this was particularly a problem for nasal flow measures. With experience, staff instructed caregivers to check sensors during the night and replace sensors where possible. For clinicians introducing home-based CRPG services, we have provided practical recommendations based on our experience that may improve success rates [See Text box]. Sensor refusal is more difficult to predict and address. Caregivers were encouraged to attach sensors while the child was awake to allow acclimatisation. In some cases sensor attachment was only successful once a child was asleep. The future development of sensors that are embedded into clothing would overcome some of the issues that lead to data artefact.

Most families rated home CRPG easy or OK to use, and would choose to perform their next CRPG at home. Those who failed on the first attempt at home in the research group were

unsurprisingly more likely to report that the equipment was difficult to use (47% v 28%). While 76% of those responding were happy to repeat CRPG at home in the future, it should be noted that response rates were lower for parents when first attempt at CRPG failed. Further research could usefully clarify how families could be better prepared and supported to achieve successful studies.

In the clinical sample, experience of previous CRPG was not associated with successful acquisition. Most of the Down syndrome research children were CRPG naïve, however, they would have been familiar with hospital settings and their experience may not compare directly with otherwise healthy and typically developing children. Poels et al.²⁵ trained caregivers to set-up CRPG at home in 24 children assessed prior to adenotonsillectomy. These were likely to be typically developing children with no comorbidities and study success rate was low at 29%. There were 14 typically developing, otherwise healthy children in our clinical sample and success rate in this group on first attempt was 86%. While our numbers are too low to draw firm conclusions this does suggest that prior experience of CRPG or indeed the hospital environment is not a pre-requisite for successful outcomes of home studies.

We worked with the manufacturer to adapt the CRPG equipment for paediatric home usage, specifically, leads from the thoraco-abdominal bands and nasal prongs were customised to be shorter to avoid risk of entanglement. This, plus the addition of an integrated video recorder for some clinical studies which aided scoring of studies may, in part, have explained the higher success rates in the clinical group.

Limitations and recommendations for future work

Participating families either self-selected as research participants or chose home CRPG in the single-centre clinical setting and are likely to be more motivated than a non-selective cohort. We do not have reliable comparative clinical data on families who chose in-patient

hospital studies to indicate how representative the clinical group was. Selection bias limits the generalisability of our findings. However, in clinical settings family preference is likely to be an important predictor of success and arguably our clinical cohort are representative of typical families who would make this choice. Future studies using a patient preference design could identify clinical and socio-demographic characteristics predictive of successful CRPG studies. In addition, future multi-centre studies should routinely test a wider range of important paediatric signals such as carbon dioxide and video in the home setting.

Our data did not attempt to capture costs. Indeed, no prior studies have examined the costs of paediatric home CRPG¹⁷. Cost analysis data from Spanish adult studies have reported considerable cost savings using home CRPG^{7,26}. If replicated in paediatric settings, home CRPG could offer significant cost efficiencies. Finally, and importantly, future research should capture child and caregivers experiences and views about home CRPG including their preferences for preparation and training on study set-up from skilled staff.

Conclusion

Home based paediatric services have been a health service aspiration for over half a century²⁷ and offer the advantage of timely, effective care that minimises disruption for the family²⁸. Home CRPG shows promise as an alternative to routine diagnostic in-patient studies. Better experience for families, convenience and potential cost savings could reduce the burden on families and health services alike. Children with chronic conditions, who need repeated CRPG studies through life, may be particularly suited to this approach. Future research into both costs and quality aspects of this exponentially growing health service diagnostic test are urgently needed in paediatric sleep centres across the UK.

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Table 1: Associated health and developmental comorbidities of the clinical cohort (n=61)

	Frequency
Typically developing with suspected sleep-disordered breathing	14
Typically developing with respiratory comorbidities (e.g. alveolitis)	5
Typically developing with other comorbidities (e.g. sickle cell disease)	4
Down syndrome	9
Cerebral palsy	8
Co-morbidities associated with hypotonia	6
Obesity	2
Craniofacial anomalies	3
Other neurodevelopmental / neurological comorbidities	10

Table 2a: Success and acceptability of CRPG at first attempt by demographic characteristics in the research cohort

Measurement	Successful first attempt	Failed first attempt	Significance level
Research Cohort	n=143	n=51	
Gender (% male)	50	61	P=0.466
Median age in years (range)	2.60 (0.5-5.9)	3.33 (0.6-5.9)	P= 0.951
% children with one parent with further education >18 yrs	64.7	57.1	P=0.451
% reported 'usually restless sleep*'	65.7	63.8	P=0.628
Research Cohort – Ease of Use of Equipment	n=127	n=38	
Easy	30	7	P=0.004
OK	61	13	
Difficult	36	18	
Research Cohort – Would the family repeat home CRPG?	n=122	n=37	
Happy to repeat	105	28	P=0.680
Not certain	12	2	
Not happy to repeat	5	7	

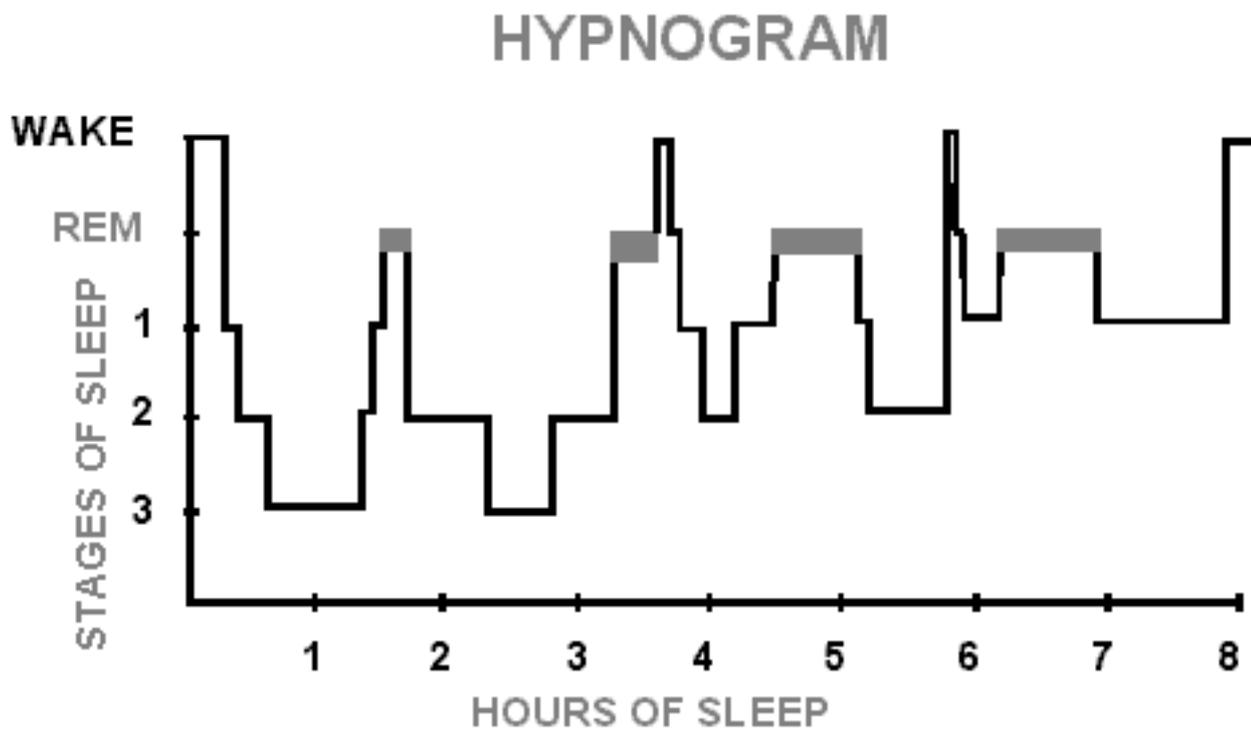
*Question taken from CSHQ [Owens 2000] or infant sleep questionnaire [Sadeh 2004] and missing for 7 children.

Table 2b: Success and acceptability of CRPG at first attempt by demographic characteristics in the clinical cohort

Clinical Cohort	n=50	n=11	
Gender (% male)	52	73	P=0.317 NS
Median age in year (range)	8.0 (0.4-17.5)	3.5 (0.5-19.4)	P=0.373 NS
Past experience of CRPG	19 (38%)	5 (45%)	P=0.738 NS
Clinical Cohort – Ease of Use of Equipment	n=39	n=6	

Very Easy	9	3	P=0.071 NS
Easy	15	2	
Okay	14	0	
Difficult	1	1	
Clinical Cohort – Would the family prefer home CRPG?	n=19	n=4	
Yes	16	4	P=1.000 NS
No	3	0	

Figure 1: Sleep Hypnogram



Text box: Technical tips for successful home studies

- Familiarise skilled staff in equipment use in the sleep laboratory setting to anticipate technical difficulties before home use
 - Select families who have had in-patient CRPG before or those with night-time carers as first subjects to trial equipment on
 - Create pictorial help guides / accessible online videos of set up procedures
 - Measure the child for their respiratory bands
 - Trial of flow leads as above
 - Plug all the sensors into the equipment so that they are securely in place before issuing to family
 - Give families choice of setting the device at a pre-agreed recording start time or pressing record at bedtime
 - Encourage parents / carers to do a couple of overnight checks of sensors
-