The effects of night positioning on sleep, postural deformity and pain in children and young people with cerebral palsy - an exploratory study.

Introduction

Cerebral palsy is an umbrella term to describe a number of disabilities where motor (movement) difficulties are caused by damage to the developing brain. Children and young people who are unable to walk are particularly vulnerable to musculo-skeletal deformity which contributes to pain, deformity and inability to carry out daily tasks, amongst other problems.

Sleep patterns in children with cerebral palsy are often affected as a result of their neurological impairment. Approximately 66% of children aged 2-16 with cerebral palsy have sleep problems, compared with less than 1% in non-disabled children.

A sleep system aims to provide support to a child when lying down in bed and is often used at night time. Clinicians use a sleep system to help manage a child’s posture and in particular reduce hip problems and the need for surgery. However, there is very little evidence of the effectiveness of sleep systems.

The aim of this programme of research is to find out if using a sleep system helps children with cerebral palsy by allowing them to be more comfortable in bed at night, reduce pain, improves sleep, prevents deformity and avoids the need for surgery. If this is the case, the provision of sleep systems, which are relatively inexpensive, could potentially offer significant saving to the NHS in addition to improving the quality of life of the children with cerebral palsy and their families.

This exploratory study is an essential first stage and will help in the development of a protocol for a larger study. It will help to identify whether the acceptability of and adherence to using sleep systems is an issue, the size of any effects and inform the calculation of an indicative sample size for a future randomised controlled trial.
Sleep patterns in children with cerebral palsy are affected as a result of their neurological impairment with approximately two thirds of children aged 2-16 experiencing sleep disturbance, compared with less than 1% in typical children (Khan et al 2002, Khan 1996). Poor quality of sleep ultimately affects their quality of life and also the quality of parents/carers lives (Wiggs 2001).

Although disturbed sleep quality has been attributed to a variety of causes including: obstructive sleep apnoea, discomfort or pain, hormone imbalance, digestive problems such as reflux and behavioural factors, there is limited published literature. Two small studies have investigated respiratory function (Kotagal et al 1994, Hill et al 2009) and suggested respiratory function is assessed prior to prescription of a sleep system.

Pain is a factor which becomes increasingly important in young people as postural deformity develops. Hip dislocation affects approximately 60% of children with bilateral cerebral palsy by the age of 5 years (Scrutton et al 2001) and often causes significant pain with a mean duration of 20 years (Schwartz et al 1999). The effects of such pain may impact significantly on the quality of day time activities as well as night time sleep quality.

Associations have been seen between asymmetrical lying and the subsequent direction of postural deformity including windswept deformity and the side of hip dislocation (Porter et al 2007, 2008) although this was not proof of cause and effect.

Night time positioning is part of a conservative approach to the management of posture, pain and hip dislocation. There is some evidence to suggest that night time positioning may help to decrease the level of hip dislocation and this might be maintained through to adolescence (Pountney 2002, 2009, Hankinson and Morton 2002).

A study by the main applicant indicated that the introduction of the use of 24 hour postural management (a system of continuous support in lying, sitting and standing) resulted in a decrease in the level of hip dislocation in children with bilateral cerebral palsy (Pountney et al 2002, 2009). However, it is not possible to tell from this research how effective the sleep system component of the overall package of 24 hour postural management is.

Hankinson and Morton (2002) reported a small mixed methods pilot study involving seven children with cerebral palsy who were positioned in supine with 20° of hip abduction using the Jenx Dreama lying system. X-ray, parental questionnaire and a sleep chart data was recorded at baseline and
again 6 and 12 months after the introduction of the sleep system. Reductions in migration percentages and in parental questionnaire scores were reported although the sample was very small.

Goldsmith (2000) conducted a feedback study of the experiences of families using the Symmetrisleep night-time positioning system over a one year. The results were inconclusive due to potential for bias and lack of clarity of method (Wynn & Wickam 2009).

Other authors have recommended the use of postural management including the use of lying supports (Farley 2003, Chia 2005, Pope 2007, Gibbs 2005, Wynn & Wickam 2009).

To date there have not been any substantial studies investigating the effect of night lying positioning using sleep systems on sleep quality, pain or hip deformity, and research is needed to clarify this (PASA 2009).

This exploratory study and a subsequent larger scale RCT would show whether there is a benefit as a result of using a sleep system for night time positioning. If significant benefits in terms of postural deformity, and particularly hip dislocation, are demonstrated then it will be possible for funders to make an informed decision about the provision of such equipment. The cost of providing the equipment could then be weighed against the potential savings as a result of the reduced need for other expensive services including surgery, physiotherapy, occupational therapy and provision of orthoses. If there are benefits in terms of sleep quality and pain then this will also help to inform funders as to whether to provide sleep systems based on the potential to improve quality of life for the child and family.

**Purpose and design**  
This is an exploratory study to support the development of a full randomised controlled trial.

The primary research aim is to investigate the impact of night lying positioning equipment on the quality of sleep, pain and postural deformity.

The research objectives are to determine the effect of a sleep system on:

1. Sleep quality using actigraphy, oximetry, sleep diaries and Chailey sleep questionnaire.

2. Pain using the Paediatric Pain Profile and the Brief Inventory of Pain.
3. The development of musculoskeletal deformity using Reimer’s hip migration percentage, lower limb range of movement, Cobb angle and need for surgery.

This exploratory study will help to identify the size of effect including whether there is any change, whether it is a clinically significant change, and what are the pre and post differences. It will also help to identify the adherence to using sleep systems and factors affecting non-adherence. This will help to determine the choice of outcomes, appropriate duration and indicative sample size for a future randomised controlled trial. This line of investigation and the subsequent RCT will allow the hypothesis that “night time positioning using sleep systems has an effect on sleep quality, pain and development of postural deformity in children with cerebral palsy” to be tested properly.

This study builds on previous studies on sleep and postural management. The applicants are ideally placed with strong links with specialist centres, a network of community based paediatric services, and National multi-disciplinary interest groups including Posture and Mobility Group. Chailey Heritage Clinical Services & Oxford Brookes University have a track record in research on neurodisability including sleep and management of deformity.

The study has been reviewed by the NIHR Research for Patient Benefit panel and funding has been awarded.

**Methods**

The study will recruit 50 participants in 4 regions which are identified by a centre number.

**Inclusion Criteria:**

- Children not using a sleep system
- Children and young people with cerebral palsy and not walking independently.
- Aged between 3-16 years.
- Children at levels IV to V on the Gross Motor Function Classification System for cerebral palsy (Palisano et al 2008).

**Exclusion Criteria:**

- Children already using night time sleep systems
- Children under the age of 2 years because they tend not to have established sleep patterns.
Children over 16 years as they may move out of paediatric services before the end of the study.

Children with other conditions which may affect their musculoskeletal development or sleep quality.

Children and young people currently using a sleep system will not be able to join the study.

### Recruitment

Health professionals in paediatric services (including paediatric consultants, physiotherapists and occupational therapists) in the study areas will be asked to identify suitable children, based on the inclusion and exclusion criteria.

An invitation letter and information sheet will then be provided to the family by the identifying healthcare worker. Families will be asked to reply to the researchers indicating if they are interested in taking part in the study. A reply slip along with a stamped addressed envelope and contact telephone details for the researchers will be provided.

The local researcher will then make contact with the family that have expressed an interest in participating in the study and an appointment with the child and parent(s) will be arranged. At this appointment the research study will be discussed, any questions answered, and if and when appropriate informed written consent will be obtained. Where potential applicants are identified whose first language is not English, the NHS translation and interpretation services will be used.

Information sheets will be available in a variety of formats to ensure that children are able to understand the information provided. These will be in symbol format, two written and illustrated information sheets and verbally. Informed consent will be required by the parents and assent will be required from the child.

Initially recruitment will focus on users of health services in the South East, South West and Thames Valley where the applicants are based. It is intended that the study will be expanded to include additional areas.

### Allocation Using Minimisation

For each participant there will be a baseline assessment to collect information on current sleep quality, pain and musculoskeletal deformity. Following the baseline children will be allocated to the intervention or control group i.e. either using a sleep system or not. Allocation will be based on minimisation
as recommended by Altman & Bland (2005) for small studies. Age and hip migration at the baseline assessment will be used as the defining variables.

The local researcher will inform the research assistant of the age and hip migration at baseline of each new participant. The research assistant will use MINIM software (recommended by Altman & Bland 2005) to perform the allocation using minimisation. By entering the age and hip migration information participants will be allocated to four categories:

- Age <= 9 years; hip migration <= 33%
- Age <= 9 years; hip migration > 33%
- Age > 9 years; hip migration <= 33%
- Age > 9 years; hip migration > 33%

The MINIM software will randomly allocate the first participant in each category. Subsequent allocations in each category will be performed to minimise imbalance in numbers allocated to intervention and control groups within that category. If at any time the number of participants in the intervention and control group are the same within a category, the next participant in that category will be allocated randomly.

Bland & Altman (2005) state that minimisation is a valid alternative to randomisation and has the added advantage, especially in a small trial such as this one, of ensuring that there will only be minor differences in numbers allocated to intervention and control groups within each category.

The guidelines contained in the updated CONSORT 2010 statement will be followed to ensure concealment and other principles of good practice.

Reference:

Intervention
Participants allocated to the intervention group will be provided with night time positioning using an appropriate lying sleep system for a period of two years. A choice of three different adjustable sleep systems will be offered (Chailey, Dreama and Symmetrisleep). All three systems provide similar postural support (with ability of positioning at 30 degrees of abduction at the hips) but are different aesthetically. The sleep system will be appropriately
set up and adjusted to suit the participant. The participant’s family or carers will be trained in the use of the sleep system. The child and sleep system will be monitored by the local therapist researcher.

Each child will be followed up every 6 months and the data on the outcome measures collected until to the end point of the study. All children who are recruited to the study will continue to have all their medical and therapy needs met by their local team. At the end of the study the children will be allowed to keep the equipment until they have grown out of it if they so wish. Children who had not used a sleep system and would like to use one, will be provided by their local trust. The local researchers will collect the data and send it to the chief investigator for analysis.

Any surgery carried out or planned during the intervention will be monitored and recorded.

**Outcome Measures**
The outcome measures include sleep quality, pain and the development of musculoskeletal deformity. The study will measure quality of sleep using the Chailey sleep questionnaire (Khan 1996, Khan & Underhill 2006, 2009), sleep diaries, overnight oxygen saturation levels using oximetry and actigraphy.

1. **Sleep Questionnaire** - The sleep questionnaire consists of 3 components: Sleep history; sleep behaviour; breathing quality during sleep and details of sleeping position. The sleep questionnaire has been tested for reliability and validity in children with and without cerebral palsy. Alternative questionnaire tools to measure sleep disturbance were considered (Owens 2000; Cook 1990). However, these two questionnaires contained several questions which are inappropriate for children with cerebral palsy and were not designed to pick up specific problems exhibited by children with cerebral palsy.

2. **Oximetry** - can be measured using a sensor on the child’s finger and will indicate percentage oxygen saturation. Saturations are graded as over 90% normal, mild 85-90%, moderate 80-85% and severe <80%. A drop of 5% under 90% would be significant. The baseline measure would involve collection of oximetry measurements and sleep measurements that the parents considered to be representative of a typical nights sleep for their child.

3. **Actigraphy** - this will be used to indicate periods when the child is awake and asleep. This involves the use of a simple device attached to
the wrist which detects movement via accelerometers. Although actually measuring movement it is generally accepted as a good indicator of when a child is asleep or awake (Underhill 2009). This information will be supported by the sleep diaries.

4. Pain - An indication of pain will be recorded using the Paediatric Pain Profile (Hunt et al 2004) and the Brief Inventory of Pain. The Paediatric Pain Profile (PPP) is a 20 item parent-reported behaviour rating scale for assessing pain in children with severe physical and learning impairments. The PPP rates pain on a five point scale (0-5) verbal rating scale (no to very severe pain) with a maximum score of 60. The Brief Inventory of Pain is similar but designed to be used with children who can self report.

5. X-ray measures - Hip displacement will be measured using an x-ray with a child lying either on his or her back. It is a reliable method of measuring displacement. Current guidelines for hip surveillance for the population included in this study recommend radiographic assessments are carried out at least annually (MacKeith Consensus Statement Gericke 2006). Children who have had an x-ray within 3 months of the baseline data collection will not be required to have another x-ray until data point 3.

The progression of spinal deformity is linked to hip dislocation and we will request copies of the spinal x-rays either from local trusts if these are not available on PACS. Spinal curvature will be measured using the Cobb Angle preferably in sitting in their own equipment or held in place but this may not be always possible.

6. Lower limb range of movement will be reported using the Physical Assessment – Lower Limb.

The above measures will be carried out by the local researcher. Equipment, training and support will be provided. Radiographs will be anonymised and sent to the research team at Chailey Heritage Clinical Services so that blinded measurement of hip migration can be carried out.

Sample Size
It was not possible to calculate an accurate sample size as there is insufficient information currently available hence the reason for making this an exploratory study to inform the development of a randomised controlled trial.
Initial attempts at a sample size calculation were based on estimated changes in paediatric pain profile and oximetry. A power calculation using 0.95 at a significance level of 0.05 using the Paediatric Pain Profile would require 5 children to move pain score from a severe / moderate to mild (standard deviation of 13 and a delta difference value of 30). Using oximetry values a 0.95 power and 0.05 level of significance corresponds to 17 children using a standard deviation of 4 and a delta change of 5. It was recognised that within this group of children there is a likelihood that some will not tolerate the lying supports or have to withdraw due to other medical issues and therefore possible attrition would need to be taken into account. It was therefore estimated that a total of 50 children should be recruited to this exploratory study with 25 in the intervention group and 25 in the non intervention group.

**Data Analysis**

Local researchers will send data to the Chief Investigator and project team at Chailey Heritage Clinical Services for analysis. Independent t tests and analysis of covariance (ANCOVA) will be undertaken primarily to determine the sample size and inform the protocol for a subsequent RCT. Adherence and other issues associated with the use of the equipment will also be taken into account.

Data checking and exploratory data analysis will be conducted, including assessment of data distribution normality and of differences in baseline characteristics between the study arms. The primary analysis will be change in Paediatric Pain Profile and also hip migration percentage between baseline and immediate post-intervention assessments, using unpaired student’s t test, or Mann-Whitney U if the data are non-parametric. ANCOVA will also be used to take into account covariates. Analyses of intervention effects will all be made on an intent-to-treat basis, minimising Type 1 error and reflecting the pragmatic nature of this trial.

**Dissemination**

Findings will be disseminated at a number of levels to inform all the likely stakeholders including the children and young people with cerebral palsy and their families, paediatric healthcare professionals (including physiotherapists, occupational therapists, paediatric consultants) and their managers, in a number of different ways:

- Feedback to participants and their families in a suitable format and in an appropriate level of language and via the use of symbols and written documentation.
• Feedback on the progress and findings of the study via websites of appropriate organisations e.g. NHS, Universities, User groups and Professional Specialist Interest groups.

• Feedback to participating sites via in service training.

• Publication in peer-reviewed journals e.g. Developmental Medicine and Child Neurology, Child: Care Health & Development, APCP journal.

• Presentations at National and International conferences e.g. European Academy of Childhood Disability.

• Publicise the findings through professional networks such as the Association of Paediatric Chartered Physiotherapists, Posture and Mobility groups.

**Time Frame**
The total duration of the study will be for three years. Each participant will take part in the study for two years unless they need or choose to leave the study earlier. The sleep system will be retained by the family if they wish. Children who had not used the sleep system and would like to use a system will be provided by their local trust.

**Study Procedure**
• Ethical approval has been obtained. Research governance approval will be sought for the study in each location.

• Training and induction of the research physiotherapists and consultant paediatrician, within each centre, in the provision of sleep systems and in the use of the outcome measures.

• Recruitment of children into the study. Recording of demographic data, including age of child.

• Baseline assessment using all the outcome measures. The timing of the assessment is detailed in section 15.

• The end point for the children is at 24 months from their baseline measure. Children will be allowed to retain the sleep system if they so wish.

• Data analysis, write up and dissemination of results will take place between months 32 and 36. End point of the study.