



Положение во сне детей и взрослых с неврологическими нарушениями: систематический обзор литературы

ЦЕЛЬ: системы, относящиеся к положению во сне, часто входят в программы круглосуточного управления осанкой для детей и взрослых с неврологическими нарушениями. В поисках доказательств эффективности для детей с церебральным параличом организация Кокрейн обнаружила два рандомизированных контролируемых испытания. Этот обзор направлен на оценку более широкого спектра исследований, включающие разнообразные диагнозы и возраста испытуемых, чтобы предоставить терапевтам качественные доказательства, лежащие в основе практики.

МЕТОДОЛОГИЯ: поиск всех рецензируемых исследований, в которых оценивалось использование систем положения во время сна, был проведен в базах данных MEDLINE, EMBASE, CINAHL, Cochrane Library, BNI, HMC, PEDro, OTSeeker и реестрах клинических испытаний. Также, исследователи связались с организациями, поддерживающими людей с инвалидностью, производителями и коллегами по всему миру. Заголовки статей были проверены на актуальность двумя рецензентами. Данные извлекались с помощью специальных количественных или качественных форм одним рецензентом, а после проверялись вторым рецензентом.

РЕЗУЛЬТАТЫ: Всего в данную работу было включено 14 исследований; все исследования являются маленькими и большинство из них имеет низкое качество. Исследователи не могли сделать выводы о пользе системы, основываясь на полученных данных, однако они также не обнаружили никакого вреда.

ЧТО ЭТО ЗНАЧИТ: Количество фактических данных, подтверждающих практику, остается небольшим и, в основном, низкого качества. Терапевты должны проявлять осторожность, рассказывая о преимуществах лечения семьям.

Автономная некоммерческая
организация «Все дети могут»



Sleep positioning systems for children and adults with a neurodisability: A systematic review

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Abstract

Introduction: Sleep positioning systems are often prescribed as part of a 24-hour postural management programme for children and adults with neurodisabilities. In a search for evidence of effectiveness for children with cerebral palsy a recent Cochrane review found two randomised controlled trials. This review aims to appraise a broader set of studies including any neurological diagnosis and users of all ages to inform therapists about the quality of the evidence underlying practice.

Method: A comprehensive search for all peer-reviewed studies that evaluated the use of sleep positioning systems was conducted in MEDLINE, EMBASE, CINAHL, Cochrane Library databases, BNI, HMIC, PEDro, OTSeeker and clinical trials registries. Disability organisations, manufacturers and colleagues worldwide were also contacted. Titles were screened for relevance by two reviewers. Data were extracted into bespoke quantitative or qualitative forms by one reviewer and checked by a second. Findings were analysed into simple themes.

Results: A total of 14 studies were eligible for inclusion; all were small and most were of low quality. Inferences of benefits cannot be made from the literature but also no harm was found.

Conclusions: The body of evidence supporting practice remains small and mostly of low quality. Therapists should remain cautious when presenting the benefits to families.

Keywords

Postural management, positioning, sleep systems, cerebral palsy, neurodisability, sleep

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Introduction

Postural management is an approach used by occupational therapists and physiotherapists for children and adults with neurodisability. Neurodisability is defined by expert consensus as being ‘a group of congenital or acquired long-term conditions that are attributed to impairment of the brain and/or neuromuscular system and create functional limitations. A specific diagnosis may not be identified. Conditions may vary over time, occur alone or in combination, and include a broad range of severity and complexity. The impact may include difficulties with movement, cognition, hearing and vision, communication, emotion and behaviour.’ (Morris et al., 2013).

Spasticity and/or weakness are common characteristics of these conditions and cause the adoption of postures that are unstable and asymmetrical. These asymmetrical postures may in turn lead to pain, progressive loss of function and fixed changes in body shape including hip dislocation and spinal curvature (Graham, 2004; NICE, 2012; Soo et al., 2006). Postural management programmes use equipment over 24 hours to support sitting, standing and lying (Gericke, 2006). Postural support in lying is provided primarily at night with whole body systems. There are several manufacturers of these systems which consist of one or

more component parts held in position by a base layer or sheet (Polak and Clift, 2007). They all have similar therapeutic aims of increasing comfort, reducing asymmetry and improving sleep. Some provide support only in supine lying or prone while others can also be used for supported side lying (Polak and Clift, 2007). While there are differences between systems the generic description is used here as this study is not comparing between types. There are several terms used to describe the equipment that provides postural

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support in lying; night-time postural management equipment (NTPME), sleep systems, sleep positioning systems. We have chosen the latter.

Many therapists are prescribing this equipment routinely in the United Kingdom (UK) and elsewhere. Service users are increasingly demanding it although provision by service providers is patchy (Department for Education and Skills, 2007). Consideration of the use of postural support at night is recommended by the National Institute for Health and Care Excellence for children and young people with non-progressive brain disorders (National Institute for Health and Care Excellence, 2012), although no evidence of effectiveness was provided. The use of sleep positioning systems also has critics. Children with neurodevelopmental disabilities are known to have increased difficulties with sleep (Jan et al., 2008) and some have suggested the equipment may interfere with sleep (Gough, 2009). Pountney et al. (2009) found children and families were less motivated to use sleep positioning systems than day time support in sitting and standing.

There is a lack of robust evidence for the effectiveness of sleep positioning systems. Our recent Cochrane review of sleep positioning systems to reduce hip migration specifically in children with cerebral palsy found only two low-quality randomised cross-over trials that met the inclusion criteria in respect of secondary objectives relating to sleep quality and pain (Blake et al., 2015). A recent review searching for evidence of effectiveness of postural management for people with intellectual disabilities and severely impaired motor function reported that the distinct lack of evidence for efficacy of sleep positioning systems should be of urgent concern (Robertson et al., 2016).

A systematic review of a wide range of literature, including those with lower levels of evidence, will be useful in this field of practice in which there is a paucity of randomised controlled trials. This type of 'configurating review' (Gough et al., 2012) enables synthesis of a broad range of literature taking account not only of the quality of a study but also the relevance to the review.

Objectives

This review sought to identify and summarise qualitative and quantitative literature reporting on studies of children and adults with a neurodisability using sleep positioning systems.

Primary research questions

1. What benefits for users of sleep positioning systems are reported in the literature?
2. What risks or adverse reactions are reported?

Secondary research questions

1. Can/do users and carers adhere to the prescribed therapeutic regimens?

2. Do parents/carers need specific training in the use of sleep positioning systems?

Method

Design and search strategy

The review was undertaken following the general principles published by the National Health Service (NHS) Centre for Reviews and Dissemination (CRD, 2008) and was registered with PROSPERO on 3 June 2015 under the title 'Postural support in lying for children and adults with a diagnosis of neurodisability' with the registration number CRD42015022099 (<http://www.crd.york.ac.uk/PROSPERO/>). The PRISMA checklist (<http://prisma-statement.org/PRISMAStatement/Checklist.aspx>) was used throughout the process to assist in adhering to best practice in the conducting of the systematic review.

A comprehensive search strategy using MeSH and free-text terms was developed by an information specialist (MR) in consultation with the review team. Terms for interventions (postural support equipment) were combined with terms for sleeping or lying. The search was developed for MEDLINE and adapted as appropriate for the other databases. The full electronic search strategy for Ovid MEDLINE can be seen in Appendix.

Searches were initially carried out in May 2015. An updated search was carried out in February 2018 to identify relevant studies published since May 2015. The following databases were searched: MEDLINE and EMBASE (via OVIDSp); CINAHL via EBSCOhost; CENTRAL, CDSR and DARE via the Cochrane Library; BNI via ProQuest; HMIC via OVIDSp; PEDro via <http://www.pedro.org.au/> and OTSeeker via <http://www.otseeker.com/>. Searches were also run on Conference Proceedings Citation Index – Science (CPCI-S) and Conference Proceedings Citation Index – Social Sciences & Humanities (CPCI-SSH) via Web of Science, and WorldCat (dissertations & theses). Previous reviews and the bibliographies of included studies were scrutinised for relevant articles. We checked for any ongoing research through the Current Controlled Trials Register, the MRC Clinical Trials Register and PROSPERO.

We anticipated that non-academic literature may be an important source of information for this topic. We searched the websites and/or contacted relevant organisations (Posture and Mobility Group, The Foundation for Assistive Technology, SCOPE, cerebralpalsy.org.uk, Cerebra, Postural Care Skills, Pamis, Postural Care Action Group, Mencap Postural Care Campaign) to ask for copies of any relevant reports. Colleagues working in the field of night-time postural management around the world were contacted at the Cerebral Palsy Alliance in NSW, the Centre for Cerebral Palsy in Perth and CanChild at McMaster University. We used these connections to snowball the contact list of people and organisations that may have publications.

We contacted the following manufacturers of sleep positioning systems for relevant reports: Active Design Ltd (Chailey Lying Support), Jenx (Dreama), James Leckey Design (Sleepform), JCM (Moonlite), Helping Hand (Symmetrisleep), Smirthwaite (Snooooooze), Simple Stuff Works, Posture Care Ltd (Hugga), Medifab, Megalong Positioning Services, Cuneiform (Matchett system).

Inclusion and exclusion criteria

Types of studies: Quantitative and qualitative studies that had some external scrutiny or formal peer review of the final report were eligible for inclusion. Conference abstracts published in journals were eligible as we assumed these would have been peer reviewed before acceptance for presentation. Attempts were made to contact the authors of these abstracts to ask whether full papers had been published or full data were available. Case studies produced by manufacturing companies were only included if they could demonstrate that external scrutiny was applied to the final report and not just to the protocol or funding application.

Population: Children over the age of 3 months and adults, with any diagnosis of neurodisability were included. Children under the age of 3 months were excluded since it is at this age that infant movements become more organised in the typically developing child.

Intervention: Any commercially available sleep positioning systems used at night with the aim of reducing hip and spine deformity and/or improving comfort, quality of life or sleep was included. We excluded seating and standing supports.

Outcomes: Any outcome which addresses the benefits or adverse effects of the intervention was included.

Settings: At home or in residential care settings were included. Neonatal wards were excluded.

Screening and study selection

Details of potentially relevant literature were uploaded to EndNote and duplicates were removed. The abstracts and titles were screened independently for relevance by two reviewers (from GH, JJ and TK), who classified each paper as potentially to include or exclude. Full text copies of potentially relevant studies were obtained. The retrieved articles were independently assessed for inclusion by two reviewers (from GH, JJ and TK), using the pre-specified inclusion/exclusion criteria. Discrepancies at each stage were resolved by discussion, with involvement of a third reviewer (CM or JTC) where necessary.

Data extraction

Two bespoke data extraction forms, one each for qualitative or quantitative studies, were designed using the tips and checklist in the Cochrane Handbook as a guide (Higgins et al., 2011). The data extracted included the setting of the study, participants' characteristics, inclusion/exclusion criteria, the intervention and the outcomes measured. These were piloted and refined. Data were extracted from included

studies by one reviewer (of GH, JJ, TK). Where a reviewer was an author of an included study, two other reviewers extracted the data (SB, CM). The veracity of extracted data was checked by another of the three reviewers and discrepancies resolved by discussion.

Risk of bias

Risk of bias was assessed at a study level using the Effective Public Health Practice Project toolkit for assessing quality in quantitative studies (available at <http://www.ehpp.ca/tools.html>), the recommendations of Wallace (Wallace, 2004) and the Critical Appraisal Skills Programme for assessing quality in qualitative studies (available at <http://www.casp-uk.net/casp-tools-checklists>). The latter recommended that questions were asked of each qualitative study to assess quality. These included clear statements of the aims and objectives, the theoretical or ideological perspective of the author (or funder), the study design, context, methods of analysis, any limitations and claims of generalisability. The combination of this quality assessment with Gough's Weight of Evidence Framework (Gough, 2007) asking questions about the relevance of the design, analysis and focus of the study to this review produced a rating of high, medium or low.

Data synthesis

Extracted data were tabulated and recurring outcomes were defined as themes. These are discussed narratively.

Results

The search results are presented in a PRISMA flow chart in Figure 1. Of the 2324 studies screened, 14 were included.

The quality of the included studies was generally rated as low with four being rated of medium quality and none of high quality. Table 1 sets out the characteristics and rated quality of the included studies. The studies are listed by relevance to this review using the Weight of Evidence Framework (Gough, 2007) and then by the quality rating for the individual study. The most relevant and highest quality evidence is therefore in the top rows of the table, with the least relevant in the bottom rows. Supplementary Table (online only see: <http://journals.sagepub.com/doi/suppl/10.1177/0308022618778254>) displays the outcomes, measures used and the data extracted from the included studies.

Benefits and risks

Pain and comfort. Overall six studies reported pain or comfort as outcomes (Aburto and Brown, 2015; Goldsmith, 2000; Humphreys et al., 2012; Innocente, 2014; Royden et al., 2013; Underhill et al., 2012).

There were indications that some children and young people had their pain alleviated with the use of a sleep positioning system (Aburto and Brown, 2015; Goldsmith, 2000;

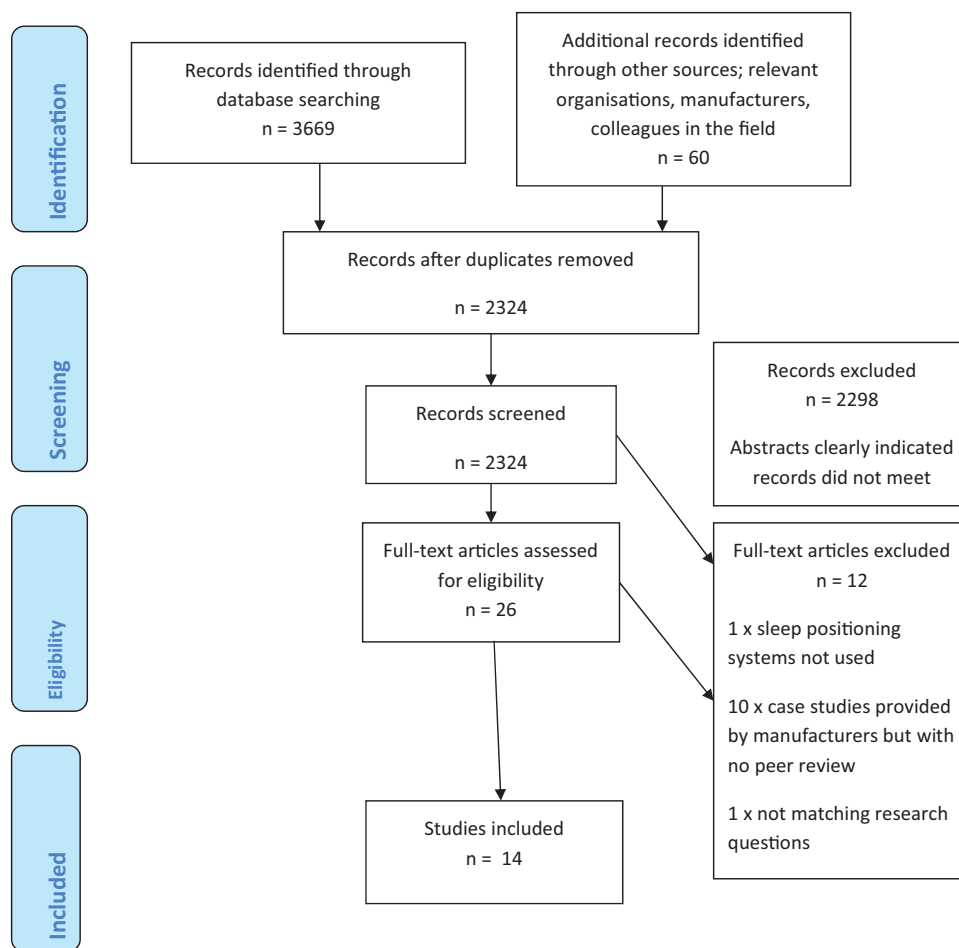


Figure 1. PRISMA flow chart (Moher et al., 2009).

Humphreys et al., 2012). However, the only study rated as medium rather than low quality, found no statistically significant difference in pain levels sleeping in or out of sleep positioning systems (Underhill et al., 2012). This study was conducted over 8 nights and the participants were well established users of sleep positioning systems. There was no evidence to suggest that use of a sleep positioning system would increase pain. One study (Goldsmith, 2000) found that some individuals faced problems of thermal comfort using their sleep positioning system. In most of the included studies, parents and carers used observational measures (Paediatric Pain Profile, the CP Child Questionnaire and a Likert scale) to report on the comfort of the user of the sleep positioning system. In one study (Humphreys et al., 2012) the children themselves were enabled to give their views using Talking Mat methodology. Most respondents asked about comfort reported improved comfort but some reported being undecided or less comfortable.

Posture and deformity. A total of six of the included studies considered posture, deformity, muscle contracture or joint range of movement (Aburto and Brown, 2015; Goldsmith, 2000; Hankinson and Morton, 2002; Innocente, 2014; Lawrence et al., 2007; Royden et al., 2013). The studies overall reported a slight trend towards stability or improvements to hip abduction as detailed below but

these may not be clinically significant, and beyond measurement error.

Hankinson and Morton (2002), in their small prospective pilot study, found a significant reduction (11%) in the rate of hip migration of the right hip and a non-significant reduction on the left, after 12 months of intervention with the Dreama. There was no statistically significant change in hip abduction but there was a change of 4 degrees per year of the right hips and 2 degrees per year of the left. In Royden et al.'s retrospective study (2013), 27 cases out of 58 had hip radiographs reported and of those 92% had remained stable or improved at 12 months. Hip abduction remained stable or improved in 18 out of 27 (66%). Using the Goldsmith indices (GIs) to measure body symmetry they report that 65% of 32 cases remained stable or improved. Aburto and Brown (2015) reported four out of four participants maintained the range of hip abduction.

The evidence above is all of low quality with low numbers of participants and three of the studies (Aburto and Brown, 2015; Lawrence et al., 2007; Royden et al., 2013) providing only a conference abstract with no further published data.

Quality of sleep. A total of seven of the included studies had sleep quality as an outcome (Aburto and Brown,

Table 1. Characteristics of included studies and strength of evidence.

Studies in order of relevance to this review	Study design	No. of participants (full data)	Age range	Diagnoses	Type of sleep positioning systems used in study	Duration	Quality rating of study	Comments
Underhill and Pountney, 2012	RCT	11 (10)	5–15 yrs	Cerebral palsy GMFCS III–V	5 × Chailey lying Support 1 × Jenx Dreama 5 × Symmetrisleep	8 nights	Medium	Sample, context, outcomes closely match focus of review but small number of participants who were well established users of SPSSs and for fewer than recommended nights of actigraphy. Used a variety of types of SPSSs. Actigraphy important method of data collection on sleep quality.
Dawson et al., 2013	Within subject cross-over study	15 (13)	1–19 yrs	Severe motor disorders (9/13 with cerebral palsy)	Not stated	14 nights	Medium	Sample, context and outcomes closely match focus of review. Sample are potentially less complex medically and socially because only 59% of those eligible were approached. Context is child's own home sleeping environment.
Humphreys et al., 2012	Interviews and observational	7 families	2–6 yrs	Cerebral palsy GMFCS levels III–V	3 × Chailey Lying Support 2 × Dreama 1 × Symmetrisleep 2 × Sleepform	4–6 mths	Low	Collecting views of users of SPSSs, family and therapists. Sample, context, outcomes, closely match focus of review. Small number of participants and limited quotes to support themes. More quantitative methods would have contributed to strength of evidence.
Hill et al., 2009	RCT	11 (9)	5–16 yrs	Severe cerebral palsy	Not stated	2 nights	Medium	Sample are established sleep system users. Outcome measures used were appropriate. Setting in a sleep lab does not replicate the usual sleeping environment for the child. Risk of selection bias present as potential participants excluded by therapists.
Goldsmith, 2000	Interviews based on questionnaire	31 families	9 mths to 19 yrs	Not known	Symmetrisleep	12 mths	Low	Sample, context, outcomes relate closely to focus of study. Limited data. Questionnaire not included. Likert scale not best for finding views. Potential for selective outcome reporting from funding.
Hankinson and Morton, 2002	Pilot prospective cohort study	14 (7)	4–14 yrs	Bilateral cerebral palsy	Jenx Dreama	18 mths	Low	Sample, context and outcomes closely match focus of review but missing data, small numbers and potential bias from funding.
Mol et al., 2012	Cross-sectional survey	82 of which 19 using NTPME	6–15 yrs	Cerebral palsy GMFCS I–V	Not stated	N/A	Medium	Children in Flemish schools, looking at sleep, useful in an international context. Useful in the comparison of SPSS compared with other orthoses used at night and none. But difficulty in separating SPSSs and other orthoses. Parental personality and competence related to the experience of burden is potentially new.
Aburto and Brown, 2015	Pilot prospective cohort study	4	3–21 yrs	Not known	Symmetrisleep	6 mths	Low	Insufficient information from abstract only but sample, context and outcomes closely matching focus of review.
Moens et al., 2014	Focus groups, interviews	20 × PT / OTs 5 × carers	N/A	N/A	N/A	N/A	Low	Design and analysis are appropriate but limited by poster presentation only. Appropriate involvement of parents of children using sleep systems.
Royden et al., 2013	Retrospective cohort study	58	0.5–19.25 yrs	Cerebral palsy and non-cerebral palsy	Not stated	12 mths	Low	Insufficient information from conference abstract only. Sample, context and outcomes relate closely to this review but with much missing data. Other factors (e.g. intervention such as Botox, surgery) that could have contributed to changes in the outcome measures were not considered.

(continued)

Table 1. Continued.

Studies in order of relevance to this review	Study design	No. of participants (full data)	Age range	Diagnoses	Type of sleep positioning systems used in study	Duration	Quality rating of study	Comments
Polak and Clift, 2007	Postal survey	448 PTs	N/A	N/A	N/A	N/A	Low	Postal survey not most effective for gaining views of therapists (24% response rate). Unable to substantiate parents' views as they are expressed via the therapists' opinions.
Newman et al., 2006	Postal survey	Parents of 173 children	6–12 yrs	Cerebral palsy GMFCS I–V	N/A	N/A	Low	No data on SPSS. Subjective comment on why they make no appearance in difference to sleep quality (families stop using).
Innocente, 2014	Postal survey to NTPME users	16	Not given	Neurodisabilities	Not stated	N/A	Low	Setting in New Zealand. Very small numbers. No separate data specifically on SPSS though users are included.
Lawrence et al., 2009	Descriptive report	5	Mid 20s	Moderate and severe musculoskeletal conditions	Symmetrisleep	5 yrs	Low	Descriptive report with no data.

GMFCS: Gross Motor Function Classification System; N/A: not applicable; NTPME: night-time postural management equipment; OT: occupation therapist; PT: physiotherapist; RCT: randomised control trial; SPSS: sleep positioning system.

2015; Goldsmith, 2000; Hankinson and Morton, 2002; Hill et al., 2009; Humphreys et al., 2012; Mol et al., 2012; Underhill et al., 2012). Of these, two were randomised controlled trials and rated medium quality (Hill et al., 2009; Underhill et al., 2012); the others were rated low. The findings suggested that sleep disturbance is not increased by the use of a sleep positioning system and is reduced for some. Although not significant for sleep latency, sleep efficiency and actual sleep time, Underhill et al. (2012) found seven different behaviours which make sleep more disruptive when children were out of their sleep positioning systems however these participants were accustomed users. Overall, 4 of the 11 needed increased re-positioning and 3 out of 11 had a substantial increase in latency when out of their equipment. Hill et al. (2009) found no difference in sleep quality measures when children slept in or out of a sleep positioning system, each for one night in a laboratory. One study reported no statistical difference in sleep disturbance between children using and not using night orthoses (Mol et al., 2012) and another found no significant difference in the number of awakenings (Hankinson and Morton, 2002). There were two studies that reported reduced number of awakenings when using sleep positioning systems (Aburto and Brown, 2015; Humphreys et al., 2012). Another found 11/19 'slept better' using sleep positioning systems which suggests fewer awakenings but was not reported as such, (Goldsmith, 2000).

Respiratory function. A total of four studies considered respiratory function (Dawson et al., 2013; Goldsmith, 2000; Hill et al., 2009; Moens et al., 2014). The two studies of medium quality found no significant difference between respiratory function in a sleep positioning system or out; however individual participants did show differences with some showing improved respiratory measures while others were breathing less well when using a sleep positioning system (Dawson et al., 2013; Hill et al., 2009). Children with severe motor disorders and neurological conditions were found to have a great variability in their respiratory function and demonstrate significant 'silent' respiratory compromise during sleep either in or out of a sleep positioning system (Dawson et al., 2013; Hill et al., 2009). Recommendations from the Dawson et al. study were that screening for ventilatory function consisting of oxyhaemoglobin saturation and carbon dioxide measures should take place prior to the prescription of a sleep positioning system and because of the variability between nights that the screening should be over 3 consecutive nights and include at least one rapid eye movement sleep cycle when vulnerability to hypoventilation is at its greatest.

Pressure areas. Overall, two low-quality studies considered the issues of pressure (Innocente, 2014; Lawrence et al., 2007). Of these, one mentions decreased pressure over bony prominences in adults when using a sleep positioning system (Lawrence et al., 2007). The other does not distinguish adequately between users of full sleep

positioning systems and smaller items of postural equipment such as pillows (Innocente, 2014).

Quality of life for users and burden of care. Overall, four studies reported on quality of life (Aburto and Brown, 2015; Humphreys et al., 2012; Innocente, 2014; Royden et al., 2013). There were two further studies that considered burden of care (Hankinson and Morton, 2002; Mol et al., 2012). One study was medium quality (Mol et al., 2012) and others were rated low quality. There is weak evidence from these small studies that quality of life is improved in some children and adults using sleep positioning systems. Burden of care was reduced for carers of those subjects that tolerated a sleep positioning system (Hankinson and Morton, 2002; Mol et al., 2012). Carers with an increased burden of care reported negative effects on their daily functioning (Mol et al., 2012).

Risks and adverse events including risk of aspiration. In addition to 20% of participants in the included studies dropping out, three of the included studies report choking, vomiting and reflux as possible risks when using a sleep positioning system or when not using equipment that a user has become accustomed to (Goldsmith, 2000; Innocente, 2014; Underhill et al., 2012). Existing breathing difficulties prior to use of a sleep positioning system may account for these adverse events (Goldsmith, 2000) and as discussed earlier need to be part of assessment prior to prescription and choice of a sleep positioning system (Dawson et al., 2013). One medium quality study (Underhill et al., 2012) found a small number (3/11) of regular sleep positioning system users had increased regurgitation or gagging when not sleeping in their equipment.

Who is most likely to benefit? Overall, six of the included studies have children and young people with cerebral palsy as their population (Hankinson and Morton, 2002; Hill et al., 2009; Humphreys et al., 2012; Mol et al., 2012; Newman et al., 2006; Underhill et al., 2012). A further six studies (Aburto and Brown, 2015; Dawson et al., 2013; Goldsmith, 2000; Innocente, 2014; Lawrence et al., 2007; Royden et al., 2013) include other neurological conditions. Royden et al (2013) included children with other neurological disabilities. Subgroup analysis showed that improvement with use of a sleep positioning system was most significant in body symmetry (using the GIs as a measure) in children with cerebral palsy (81% versus 30%) however there are many incomplete data sets in this study which compromises robustness of the results. There were no studies targeting a specific condition other than cerebral palsy and only one included older age groups. There were no studies attempting to answer the question of who is most likely to benefit from using a sleep positioning system.

Adherence and training

Difficulties adapting to sleep positioning systems and adherence to prescribed regimens. A total of five of the included

studies reported on the difficulties encountered on adapting to sleeping in a sleep positioning system (Aburto and Brown, 2015; Goldsmith, 2000; Hankinson and Morton, 2002; Humphreys et al., 2012). In these studies, an aggregated 20% of participants (12/59) were unable to adapt. The reasons given were inability to adjust to the required position and difficulties sleeping in the equipment and others dropped out because of musculoskeletal problems requiring surgery. A total of 80% were still using their equipment at the end of the intervention period. This suggests that many children, young people and their families can adapt to using a sleep positioning system. However, complete adherence to prescribed hours of use were not given and children and families needed ongoing support when the equipment was introduced, often over a prolonged period of time.

Training. Overall, two studies discussed carer training and one discussed carer competence. It was suggested that delivering training in postural care for parents and carers is likely to lead to higher levels of adherence (Goldsmith, 2000; Humphreys et al., 2012). Sense of competence is linked in one study with perceived ability to accomplish parenting tasks, including postural management (Mol et al., 2012). A total of three studies reported on the support needed for families while the users and carers get used to a sleep positioning system (Aburto and Brown, 2015; Goldsmith, 2000; Hankinson and Morton, 2002). The findings from these suggest that support for families and carers is likely to yield better results. They are all low-quality studies.

Discussion

The aim of this systematic review was to provide clinicians, families and service funders with evidence of the potential benefits and risks of using sleep positioning systems. We found some evidence that there are potential benefits in hip stability, improved sleep quality and an improved quality of life for users that can tolerate using a sleep positioning system but the quality of the evidence is very poor. No statistically or clinically significant differences were found in respiratory function in or out of a sleep positioning system. Many participants had difficulties in adapting to using a sleep positioning system and support from professionals over an extended period of time was often needed. The majority who tried using a sleep positioning system, however, did continue using it. No increased risks were identified in the literature other than those associated with having a severe neurodisability and for some the adverse events increased when taken out of a sleep positioning system they were accustomed to using. There is a question as to whether adverse effects might be under-reported as families may give up using the equipment if sleep disturbance is increased (Hankinson and Morton 2002; Newman et al., 2006) or do not initiate use if they have prior concerns of interfering with sleep (Polak and Clift, 2007).

The included studies were mostly of low quality largely because of the small numbers of participants, the lack of

methodological rigour, missing data, lack of information in the reporting or studies that were published only as conference abstracts. In the higher quality studies participants were already users of sleep positioning systems and this limits the applicability of the evidence to the general population of children and adults with neurodisability. One of these (Hill et al., 2009), a small randomised controlled trial, was conducted in a sleep laboratory rather than in the participants' homes which further reduces the generalisability of the findings. In some studies, it was reported that potential participants had been excluded by therapists but reasons for this were not given. In three studies there was the potential for risk of bias from the funding source (Goldsmith, 2000; Hankinson and Morton, 2002; Humphreys et al., 2012). In the light of the considerable withdrawal rate and the decision of many of those who were eligible not to participate, the findings must be considered with caution and best practice guidance cannot be provided at this stage.

One issue in practice is identifying who is likely to benefit from prescription of a sleep positioning system. Our findings relate very largely to children and young people, especially those with cerebral palsy. There are very limited data that includes older people; we found just one low-quality study, which was a postal survey, (Innocente, 2014).

The specific outcomes therapists are targeting when they prescribe sleep positioning systems is another practice issue. The outcomes and measures used in the studies vary. Our findings suggest outcomes in the areas of pain and comfort, posture and body shape, quality of sleep and quality of life. In order to improve the evidence base, agreement needs to be reached on the key outcomes to measure and how to measure them (Williamson et al., 2012).

Clinicians need to be aware of the difficulties users of sleep positioning systems may encounter and also the evidence of how these difficulties can be overcome. We found difficulties in adjusting to sleeping in the equipment because of the position required, getting used to the equipment itself and temperature regulation. These difficulties could be overcome in the majority of participants but often required prolonged support from services when the equipment was introduced.

The low quality of the evidence highlights the apparent difficulties of conducting robust research with this participant group. This situation is evidenced by Crombie et al. (2015) in an exploratory randomised controlled trial in night-time positioning in children with cerebral palsy in which the target for recruitment was 50 children and although 26 sites expressed interest only 3 sites participated, recruiting 22 children of whom 6 dropped out.

This systematic review had a broader scope of included studies than our Cochrane review (Blake et al., 2015). Efforts were made to find all relevant studies through searching the databases, using the list of excluded studies in the Cochrane review and by personal communication with colleagues, manufacturers and authors. The included studies all use different sleep positioning systems (as

detailed in Table 1). While the aim of this review was to review evidence of sleep positioning systems in general, variations in the study findings due to the different sleep positioning systems used in the studies cannot be ruled out.

Conclusion

A consensus statement in 2006 (Gericke, 2006) recommended that children with cerebral palsy in Gross Motor Function Classification System (GMFCS) levels IV and V should begin postural management in lying soon after birth and that more evidence for the effectiveness of the intervention needs to be obtained. Another review of the literature published up to 2007 (Wynn and Wickham, 2009) also found very limited evidence and recommended further research. Although 12 of our 14 included studies have been conducted since that date, evidence to support these recommendations remains weak and of low to medium quality.

Future research is necessary to provide sufficient evidence on the benefits or lack of benefits of using a sleep positioning system and the characteristics of patients who can tolerate the equipment and adhere to treatment. For financial reasons also, it is important to understand the impact of sleep positioning systems on users and carers. The paucity of data demonstrates the importance of writing up and gaining peer review for studies that have been conducted and the contribution that clinicians can make to the evidence bank if they were to collect and publish data as part of their routine care plans and hip surveillance.

As recruitment to a large randomised trial may be difficult to achieve, we propose that seeking a consensus of expert opinion would be the way forward to strengthen the evidence from this review of literature. We plan to follow an example in the similarly under-researched field of early powered mobility (Livingstone and Paleg, 2014) and conduct a Delphi survey of experts who have significant experience of prescribing and using sleep positioning systems. We anticipate this survey, using multiple rounds to obtain consensus, would enable the writing of a practice considerations paper which would be an important clinical guide for therapists working in the field of postural care.

Key findings

- Although prescribed by occupational therapists and physiotherapists, evidence for sleep positioning systems is currently poor.
- Inferences of benefits cannot be made from the literature but also no harm was found.

What the study has added

This review demonstrates the low quality of evidence in the field of night-time positioning. A consensus of expert opinion would strengthen the evidence and enable a guide to practice.

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Research ethics

Ethics approval was not required for this study as it was a review of literature.

Consent

There being no participants, consent was not required.

Declaration of conflicting interests

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Contributorship

Ginny Humphreys conceptualised and designed the review, screened search results, extracted data, analysed findings and drafted the manuscript. Tanya King conceptualised and designed the review, screened search results, extracted data and analysed findings. Jo Jex conceptualised and designed the review, screened search results, extracted data and analysed findings. Morwenna Rogers designed the search strategy, undertook searches and revised the manuscript. Sharon Blake provided methodological support, extracted data, helped interpret findings and critically revised the manuscript. Jo Thompson-Coon provided methodological expertise in designing the study and critical review of the manuscript. Christopher Morris conceptualised and designed the review, helped interpret findings and contributed to writing and revising the manuscript.

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Appendix

SEARCH STRATEGIES

Database: Ovid MEDLINE(R) In-Process & Other Non-Indexed Citations and Ovid MEDLINE(R) <1946 to Present>

Search strategy:

- 1 Orthotic devices/ (5121)
- 2 Self-help devices/ (3594)

- 3 chailey.ti,ab. (20)
- 4 dream.ti,ab. (1)
- 5 sleepform.ti,ab. (0)
- 6 moonlite.ti,ab. (1)
- 7 symmetrisleep.ti,ab. (0)
- 8 snoooo*.ti,ab. (0)
- 9 simple stuff works.ti,ab. (0)
- 10 simplestuffworks.ti,ab. (0)
- 11 (sleep pod or sleep-pod).ti,ab. (0)
- 12 “active design”.ti,ab. (10)
- 13 jenx.ti,ab. (2)
- 14 leckey.ti,ab. (0)
- 15 JCM.ti,ab. (2588)
- 16 “helping hand”.ti,ab. (265)
- 17 smirthwaite.ti,ab. (0)
- 18 hugga.ti,ab. (0)
- 19 medifab.ti,ab. (0)
- 20 megalong.ti,ab. (0)
- 21 cuneiform.ti,ab. (957)
- 22 matchett.ti,ab. (6)
- 23 (postural adj3 support*).ti,ab. (240)
- 24 postural management.ti,ab. (30)
- 25 postural control.ti,ab. (3424)
- 26 (postural adj2 equipment).ti,ab. (7)
- 27 (position* adj2 equipment).ti,ab. (71)
- 28 (position* adj5 support*).ti,ab. (3033)
- 29 sleep* system*.ti,ab. (64)
- 30 (sleep* adj2 equipment).ti,ab. (8)
- 31 (night* adj2 equipment).ti,ab. (6)
- 32 (night* and (orthoses or orthosis)).ti,ab. (68)
- 33 (sleep* and (orthoses or orthosis)).ti,ab. (24)
- 34 (night* and (positioning or positioner*)).ti,ab. (113)
- 35 (sleep* and (positioning or positioner*)).ti,ab. (319)
- 36 or/1-35 (19638)
- 37 supine position/ (4786)
- 38 sleep*.ti,ab. (123216)
- 39 lying.ti,ab. (18305)
- 40 night*.ti,ab. (61938)
- 41 supine.ti,ab. (22215)
- 42 nocturnal.ti,ab. (28372)
- 43 (continuous and (postural or support)).ti,ab. (14563)
- 44 (“24 hour” or 24 h) and (postural or support)).ti,ab. (2233)
- 45 37 or 38 or 39 or 40 or 41 or 42 or 43 or 44 (236537)
- 46 36 and 45 (1062)
- 47 limit 46 to animals (86)
- 48 46 not 47 (976)