Use of a lying hip abduction system in children with bilateral cerebral palsy: a pilot study

J Hankinson;
R E Morton*, Ronnie Mac Keith Child Development Centre, Derbyshire Children's Hospital, Derby, UK.

**Correspondence to second author at address above.*

Fourteen children aged 4 to 14 years with hip subluxation due to cerebral palsy (CP) were considered for a trial in a lying hip abduction system (Jenx Dreama, Jenx Limited, Sheffield, UK). Baseline data were recorded for 6 months, then assessments of the system were made for one year at 6 and 12 months. Assessments consisted of hip radiographs in the standard position, a parental questionnaire, and a sleep chart, which was completed by parents every Friday night during the trial. Three children could not enter the trial because of general sleep problems, and three could not complete it because they were unable to sleep with the system. One further child withdrew from the study just before the end because of unacceptable deterioration for which surgery was needed. The remaining seven children completed the trial and there was an overall improvement in rate of hip migration percentage on the right from 7% per annum in the baseline period to –4% with the system ($p<0.05$). On the left, changes were –3% and 0% respectively (*ns*). Average sleep at night changed from 9 to 9.4 hours (*ns*) and wakenings/night from 1 to 1.3 (*ns*). On the parental questionnaire, there were significant improvements noted with the system in position for seating and sleeping, ease of hip abduction for washing, and with pain reduction. This pilot study supports the use of this type of lying system but further studies are needed to establish the acceptability and efficacy of these systems, particularly in children aged 2 to 5 years when irreversible bony deformities of the hip tend to occur.

Hip dislocation in children with cerebral palsy (CP) is a major disability, which causes difficulties in sitting and positioning, maintenance of hygiene, and may lead to scoliosis (Kalen and Bleck 1985). It may also produce pain which can persist into adult life (Cooperman et al. 1987). Once it occurs, the dislocated hip is difficult to treat. It has been estimated that 0.68 per 1000 children have significant hip subluxation due to CP by the age of 5 years (Scrutton and Baird 1997). Recently there have been major advances in the identification of subluxing hips and in strategy to try and prevent against dislocation (Cornell 1995).

Although hip dislocation is most common in children with severe spastic quadriplegia who cannot walk at all (Cooke et al. 1989), it also occurs in those with diplegia, usually at a later age (Metaxiotis et al. 1989). Delayed locomotor milestones in both types of bilateral CP may be used as a marker for hip problems; for example, Scrutton (1989) found that inability to pull to stand by the age of 3 years is highly correlated with hip subluxation. He indicated that the main factors causing progressive subluxation appear to be hip adductor spasticity and shortening, together with lack of normal weight bearing (1989). Hip flexor tightness due to ileopsoas spasticity may also contribute (Bleck 1987). These factors lead to deformity of the hip joint (acetabular dysplasia), which is unable to contain the excessive lateral forces on the head of the femur. Once the hip is over 50% subluxed, progress to dislocation is rapid (Scrutton 1989).

To reduce hip subluxation, correct positioning is needed in sitting, standing, and lying. In non-ambulant children this consists of the following: (1) appropriate seating, keeping the hips abducted, which can best be achieved in children with severe spasticity by using a hip abduction brace; (2) use of a standing frame for around 1 hour a day; (3) use of a night hip abduction system. In a recent retrospective study, it was found that children receiving all three treatments maintained better hip integrity on X-ray than those using only a day hip brace and standing frame (Pountney et al. 2001).

Lying systems aim to provide hip abduction over long periods during sleep. Muscle stretching is provided and asymmetrical positions are discouraged as this often leads to unilateral hip dislocation. It remains unclear just how effective these devices are at relieving hip subluxation, and to date they have generally proved uncomfortable and consequently unpopular. We have investigated the use of a new lying system (Fig. 1), which consists of a modular mattress, altered according to specific body contours, and which incorporates padded supports to maintain correct positioning of the trunk, hips, and legs (Jenx Dreama, Jenx Limited, Sheffield, UK). We chose to evaluate the Dreama because it has certain advantages, including a rigid structure, that provides good positional control, and can be adjusted easily while the child is in it. It contains the legs in abduction while allowing some hip flexion, allows side lying, and enables individual contouring for back support in supine lying.

In this pilot prospective trial we evaluated the effect on hip stability on X-ray (acetabular index and hip migration percentage [MP]), sleep, and ease of positioning. We recruited mainly children with spastic quadriplegia, but included some participants with diplegia who were ambulant with aids, all of whom were thought to be at risk of hip dislocation. In order to use those with more established sleep routines, we chose children over the age of 4 years. Nevertheless, many of the

participants still had a history of poor sleep with frequent night awakening as is common with CP.

Method

PARTICIPANTS

Participants were children aged 4 to 14 years with bilateral spastic CP, who had been unable to stand with support by the age of 3 years and had not had surgery or botulinum toxin to the hip adductors in the previous 12 months. All had tight hip adductors (abduction less than 30 degrees in supine with extended hips and knees) and/or significant hip subluxation with migration over 20%. No children had surgery or botulinum toxin to the lower limbs during the study or less than 6 months before testing.

An initial 14 children met these criteria but three had to be excluded from the start because of sleeping problems: two slept only on their sides and would not sleep supine, and one was a particularly poor sleeper. This left a total of 11 children for the study: eight with quadriplegia, of whom six were unable to weight-bear and two were able to weight-bear but unable to take steps, and three with diplegia who could walk short distances in a walking frame. The latter had significant scissoring while walking because of hip adductor tightness (see Table I).

PROCEDURE

The children were assessed at the beginning and end of an initial baseline period lasting 6 months. They were then provided with a Jenx Dreama in which they slept mainly supine with hips abducted 20 degrees bilaterally and assessed at 6 and 12

Table I: Participant details and trial outcome

Participant number	*Age (y)*	*Diagnosis*	*Hip migration R/L %*	*Acetabular index R/L°*	*Hip abd'n R/L°*	*Baseline hip migration R/L % per y*	*Hip migration with system R/L % per y*	*Finish trial*	*Comment*
1	4	Quadriplegia	27/20	20/20	40/40	12/0	0/0	Yes	Kept bed after trial
2	11	Quadriplegia	50/26	10.5/25	20/20	4/6	–2/–3	Yes	Rejected bed after trial
3	13	Quadriplegia	19/18	20/20	20/25	–2/–3	4/0	Yes	Kept bed after trial
4	8	Quadriplegia	47/40	25/20	35/45	12/0	0/0	Yes	Kept bed after trial
5	10	Quadriplegia	40/14	25/15	30/30	10/–8	–11/7	Yes	Kept bed after trial
6	6	Diplegia	20/20	16/18	25/35	41/0	–3/–6	Yes	Rejected bed after trial
7	5	Diplegia	28/28	15/12	15/20	–2/–12	2/2	Yes	Kept bed after trial
8	7	Diplegia	15/23	20/30	35/35	26/46	–	No	Late exit of study for surgery
9	10	Quadriplegia	39/39	26/24	30/30	6/8	–	No	Unable to sleep; exit 3 mo
10	14	Quadriplegia	25/27	25/25	10/15	–8/–6	–	No	Unable to sleep; exit 2 mo
11	9	Quadriplegia	15/15	10/10	15/20	0/0	4/8	No	Unable to sleep; exit 1 mo
12	14	Quadriplegia							No trial; poor sleeper
13	12	Quadriplegia							No trial; slept on side
14	6	Quadriplegia							No trial; slept on side

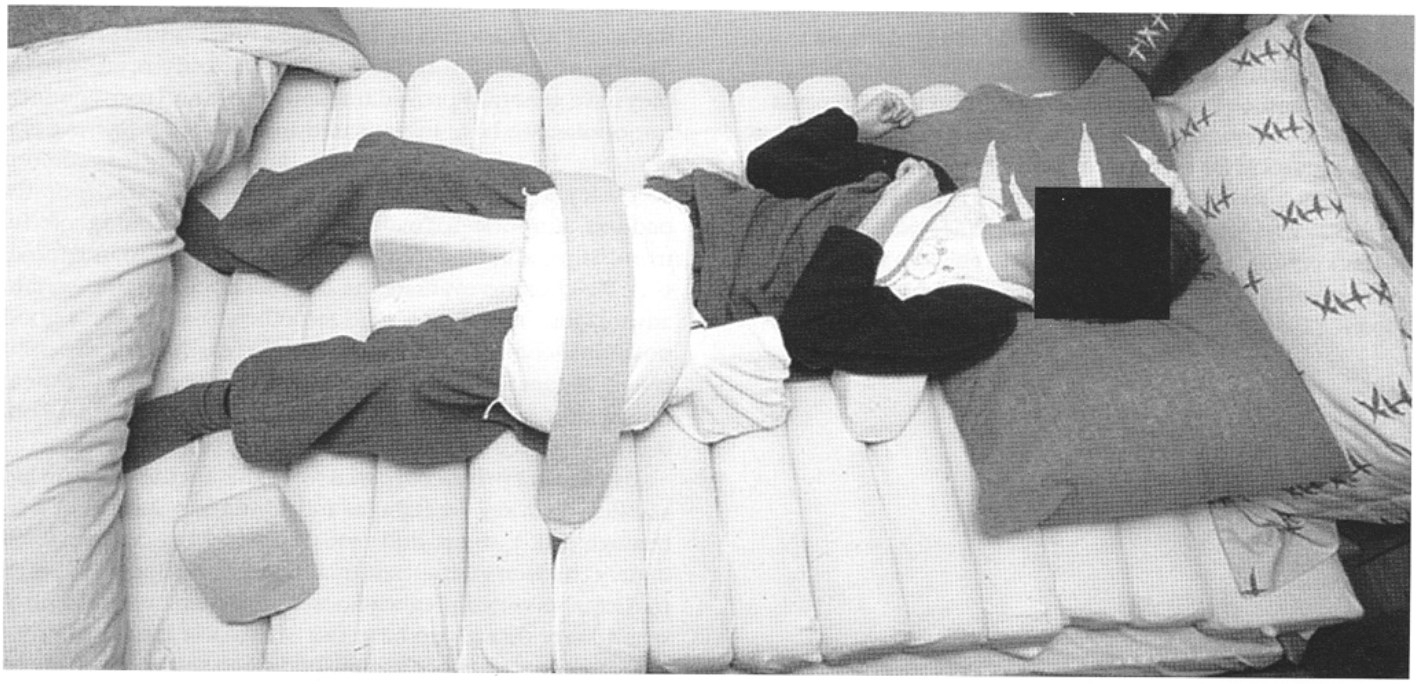

Figure 1: *The Jenx Dreama lying hip abduction system*

months. During the 18 months of the study, other measures to treat hip subluxation were kept the same, including time receiving physiotherapy, seating provision including use of hip brace, and use of a standing frame. Contacts made with the study coordinator were recorded, both in terms of home visits and telephone calls. Participants and carers were invited to keep the lying orthosis if they wished at the end of the study. This study was approved by the Southern Derbyshire Ethics Committee.

Assessments

First, hip radiographs were taken in the standard position with neutral hip adduction/abduction and compensated hip flexion for flexion contractures to eliminate lordosis (Scrutton and Baird 1997). Hip MP and acetabular angles were measured for each side. The rates of change in migration percentage per year were calculated. For the baseline, this was double the change measured over the 6-month baseline period. For the intervention period, the difference was taken between the hip MP at the beginning of this period, and 12 months later at the end. If, therefore, the change in hip migration was positive, subluxation had increased; if negative, it had reduced.

Second, maximal hip abduction was measured with a goniometer in the supine position with the hips and knees extended.

Third, carers were asked to complete a simple questionnaire asking about ease of positioning of the hips during the sitting, sleeping, and cleaning. This was done at the end of the 6-month baseline and the 1 year intervention period (Table II)

Fourth, carers were asked to complete a sleep diary every Friday night during the 18 months of the study. This consisted of a series of 24 boxes, one for each hour of the day, which were filled in for each hour the child was asleep.

Finally, for the three children with diplegia who could walk, three-dimensional gait analysis (Elite, Milan, Italy) was carried out at the end of the baseline 6 months and again after using the lying system for 12 months. A minimum three passes were analyzed for each assessment.

Study drop-outs

A further three children dropped out at 1, 2, and 3 months after starting the system because they could not sleep properly in it. One (participant 10) was instead treated with botulinum toxin to the hip adductors and another (participant 9) received adductor release surgery, both to good effect. The third (participant 11) did not receive alternative treatment and the hips significantly deteriorated (baseline MP change right [R]0%, left [L]0%, in next year R4%, L8%). One other girl with diplegia (participant 8) dropped out late after 11 months. She developed severe progressive hip subluxation during the trial; during the baseline period alone the MPs increased to R28% and L49% before the system was started. For the first 6 months she tolerated this and the MPs remained the same. During the second 6 months she became unable to tolerate the system and was later withdrawn from the study as with the MPs R31%, L58%. She received adductor tenotomy and derotation osteotomy on the left soon afterwards and is now returning to the use of the system.

This left seven children to complete the pilot study.

Table II: Comparison of changes in hips and sleep for baseline and trial periods

	Baseline period	*Period with orthosis*	*p*
Change in acetabular index	R 0	0	*ns*
	L 0	−2°/y	*ns*
Change in hip migration %	R 7%/y	−4%/y	$p<0.05$
	L −3%/y	0%/y	*ns*
Hip abduction	R 1° decrease/y	3% increase/y	*ns*
	L 1° increase/y	3% increase/y	*ns*
Sleep	9 h/night	9.4 h/night	*ns*
Night wakenings	1.3/night	1/night	*ns*

Results

Individual outcomes for the seven children who completed the pilot study, including changes in hip migration, are given in Table I. Group results for hip migration and the sleep charts are given in Table II. Results of the Carer Questionnaire are given in Appendix I.

All 11 children who started the lying system required a mean of two visits (range 1 to 4), and four phone calls (range 1 to 17) from the therapist to help them become adjusted to the system.

Gait analysis was carried out on the three children with diplegia although one dropped out due to the need for surgery (participant 8). Of the other two, participant 6 showed marked improvement in scissor gait at the end of the study. His pelvic obliquity on the left during stance reduced from 15 degrees upward to neutral, with midline position of the leg in swing. The right side was neutral in stance both at the start and end of the trial. Participant 7 also improved her scissor gait considerably with the orthosis. Her pelvis showed bilateral obliquity upwards of 10 degrees in stance before the trial. This was primarily due to abductor weakness and remained at the end, but she was able to circumduct better with the swing leg on both sides due to a reduction in adductor tightness.

Five of the seven who completed the pilot study have decided to continue using the system: one did not continue with the orthosis because it was getting uncomfortable and one because he felt too old for it, although he still slept well in it.

Discussion

In this prospective pilot study we found that of 14 participants who could potentially benefit from the lying system, six could not use it because of sleeping difficulties. This included two who were initially eliminated because they preferred to lie on their sides, but we now find this can be accommodated by starting in this position and abducting only one leg, then gradually changing to the supine position. We feel this trial will encourage carers to persist more with the system in future, knowing it can be effective. With severe sleep problems, medications such as melatonin may improve tolerance during the crucial early stages.

However, seven children (i.e. half) did complete the study and maintained their previous amount of sleep as recorded by the sleep charts. They generally found the system comfortable, and the average number of wakenings per night was less, although this did not achieve statistical significance.

Most settled into it quickly, although a few needed up to four home visits and several phone calls to the carers for adjustments. Our overall impression was that the burden of care was less for those children who tolerated the system and six of seven carers were keen to continue with the system after the study.

In this study, we used the hip MP measured from a hip radiograph taken in the approved way (Scrutton and Baird 1997) as the main measure of hip subluxation. Variations of pelvic anteversion and hip rotation have minimal effects on this measurement but hip abduction may be more significant (Reimers and Bialik 1981). Measurement error on a single X-ray appears to be minimal, and Scrutton and coworkers (2001) have estimated that the standard error for interrater differences is 3.3% and for an experienced observer the intrarater differences can be under 2% (Scrutton et al. 2001). Even taking these possible errors into consideration the mean reduction of MP of 11% on one side during the intervention period can be considered significant.

Goldsmith and colleagues (2000) have tried a similar night orthosis in 32 children (Symetrikit Symetrisleep). Fifteen individuals tolerated this for 12 months and the rest for shorter periods with satisfactory positioning in all but four. Only three had rejected this system at the time of their report. Benefits to hip positioning were not given, but this appears to compare favourably with the lying system used in this study. We chose to use the Jenx Dreama for this pilot because of its practical benefits, although it is heavier than the Symetrisleep system, and less easily transferred to different places.

We have found that for those who can tolerate the lying system, there is a significant improvement in positioning for seating and toileting, as shown by the parental questionnaires. Rather surprisingly during the short period of the study, there was a significant improvement in the hip MP using the system on one side (right), with no significant change on the other (left). It is hoped that over a longer time greater hip stability could be achieved, although whether this will allow remoulding of the joint itself with an improvement in acetabular index remains to be seen.

There is good evidence that if the hip is centred before the age of 4 years, subsequent acetabular dysplasia and hip dislocation is less likely (Harris et al. 1975). Hips should be screened at the age of 30 months in those with bilateral CP: an MP of 30% or over at this age is a strong predictor of later hip problems (Scrutton and Baird 1997). Scrutton considers that those who have hip migration of over 14% and who cannot walk 10 steps unsupported at this time should be referred to an orthopaedic surgeon (2001). That is, the point at which conservative therapy could be started in order to avoid or postpone surgery, by maintaining the hip in a good position for as much of the day and night as possible. This pilot study supports the use of a lying system such as the Jenx Dreama to reduce hip subluxation. More research is needed to support these findings, particularly into the use of these systems in infants when irreversible bone deformity could potentially be avoided.

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Appendix I: Carer questionnaire and changes of scores with orthosis after 1 year

	Mean change in score ([a])
1. Is child easy to place symmetrically with hips abducted in seating?	+0.25 (0.01)
2. Does child settle easily to sleep?	+0.5 (0.01)
3. Does child remain in symmetrical position all night?	+1.8 (0.01)
4. Is stretching the hips apart for dressing and hygiene difficult?	–1.3 (0.05)
5. Do dressing and hygiene procedures cause pain?	–0.6 (0.05)

[a] Mann–Whitney alpha score.
Questionnaire given at end of baseline and intervention periods.
Each question scored on a 5-point Likert scale.