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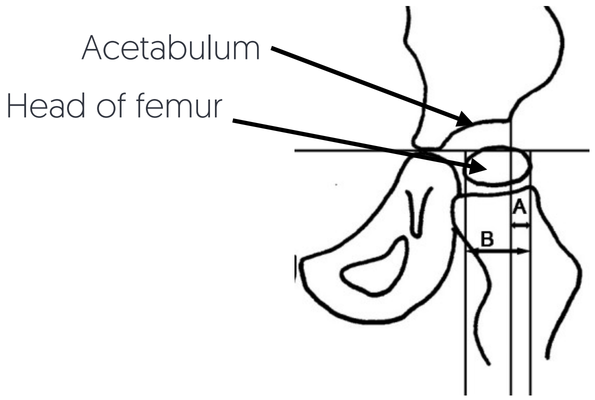
What is the evidence for the effect of hip abduction in standing on hip integrity in children with cerebral palsy?

The clinical importance of maintaining hip integrity

Children with cerebral palsy (CP) are no more likely to be born with hip problems than any other children (Pope, 2007), yet hip dislocation is a real problem for children with CP. An estimated 15% - 20% of the whole CP population is affected (Hagglund, et al., 2014), but this rises to 70%-90% for children at GMFCS levels IV-V (Soo, et al., 2006). In addition to non- ambulation, children at particular risk are those aged 2-5 years with spastic or dyskinetic CP (Hagglund, et al., 2014). Hip dislocation can result in pain, difficulties with positioning and functional abilities, contractures, skin breakdown, difficulties with personal hygiene and other orthopaedic complications (Soo, et al., 2006; Spiegel & Flynn, 2006; Hagglund, et al., 2007; Dalen, et al., 2010; Willoughby, et al., 2012; Paleg, et al., 2013). Maintaining hip integrity for children with CP is therefore a key multidisciplinary aim amongst postural management teams.

Figure 1: Calculating Migration Prcentage



Hip dislocation

Hip dislocation is described as “the gradual, lateral displacement of the femoral head from under the acetabulum, and is measured by a migration percentage (MP)” (Mayson, 2011).

The MP is determined radiologically (by X-rays) and is calculated as MP = A/B x 100 (Hagglund et al, 2014), Figure 1.

(Adapted from http://www.biomedcentral.com/content/figures/1471-2474-8-101-1-l.jpg)

Static standing frames and hip integrity

Standing frames are commonly used as part of postural management programmes to provide a number of physiological and psychosocial benefits for children with cerebral palsy, including maintaining hip integrity (Pin, 2007). The research evidence for this passive stretching practice is limited in general (Gough, 2009), and while there is more support for the benefits of increased bone density and muscle stretching (Pin, 2007), the role of static standing frames in the maintenance of hip integrity remains less clearly demonstrated.

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A review by Paleg, et al., (2013) reported six papers relating supported standing programmes specifically to hip integrity, four of which were considered to support improvement. The authors concluded that there was "fair evidence, possibly effective" and gave the outcome a "yellow light" (p.241) to indicate the level of clinical relevance – i.e. "minimal evidence; proceed with caution" (p.233).

There is certainly a widely held clinical view that children with disabilities who stand at the typical developmental age of 12-16 months are considered more likely to form the femoral head and acetabulum of the hip joint (for example, Labandz, 2011; Labandz, 2010; Dobrich, 2010; Rosen, 2010; Silberstein, 2008). In addition, a MacKeith multidisciplinary consensus statement on 24-hour postural management for children with CP recommended that the most severely affected children (GMFCS levels IV or V) should be introduced to standing programmes at 12 months of age (Gericke, 2006).

Abducted standing frames and hip integrity

More recently a single, but heavily cited publication (Martinsson & Himmelmann, 2011), has fuelled a clinical trend toward standing in 'maximum tolerated abduction' of up to 600 of bilateral hip abduction and 00 extension as a means of specifically developing and maintaining hip integrity in children with CP (see Figure 2).

Figure 2: Abducted standing

(http://blog.easystand.com/content/uploads/2015/05/Zing-MPS-Leg-Abduction.jpg)

The widespread promotion of the Martinsson and Himmelmann (2011) study and marketing of abduction standers by some manufacturers has created uncertainty amongst clinicians about the validity of their existing practice of standing without maximum abduction. While professional reflective practice is to be encouraged, it should be accompanied by a critical evaluation of all the available published evidence. This has led to a need for a review of the evidence about hip abduction and extension in standing frames. This document therefore aims to address this need and asks the question, "What is the evidence for the effect of hip abduction in standing on hip integrity in children with cerebral palsy?"

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Method

A review of the published literature was conducted on 10th December 2015. Databases searched were Ovid MEDLINE, Allied and complimentary MEdicine Database (AMED), Excerpta Medica dataBASE (Embase), Cumulative Index to Nursing and Allied Health Literature (CINAHL Plus), and Evidence Based Medicine (EBM) Reviews.

Search terms used were abduct* AND stand* AND cerebral palsy AND hip dislocation. 'Hip dislocation' was excluded from the AMED and CINAHL+ searches, as combining all four search terms yielded a zero return. This was not the case for the other databases.

Retrieved papers were limited to those published between 2000 and 2015 with abstracts, focused on children aged 0-18 years, and published in the English language.

Papers were included if they were experimental studies which investigated children with cerebral palsy who used an abducted standing device (legs positioned more than shoulder width apart).

Papers were excluded if they: reviewed existing research; addressed measurement of abduction or gait patterns; exclusively investigated other forms of treatment for hip spasticity (medical or surgical procedures) or conditions other than cerebral palsy; included abduction in other postures (such as sitting and lying) or postural management programmes; or were clinical commentaries.

Hand searches of reference lists were also conducted.

Results

Table 1: Summary and results of search strategy

Only three papers were located which matched the search criteria (see Tables 1 and 2).

	Abduct*	Stand*	Cerebral Palsy	Hip Dislocation	Hits	Limits	Review abstract/ apply criteria
MEDLINE	13281	915011	20219	5016	5	3	0
AMED	1504	25586	2630	-	10	2	0
Embase	18131	1667110	30021	6362	9	6	0
CINAHL +	3814	1441	8121	-	8	7	2
EBM Reviews	1003	103022	1850	35	1	1	0
				Hand searches of reference lists		9	1
						Total	**3**

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Table 2: Summary of relevant studies retrieved

	Author(s) and Publication	Study outline	Outcomes
1	Macias-Merlo et al (2015)a Standing programs to promote hip flexibility in children with spastic diplegic cerebral palsy Pediatric Physical Therapy 2015; 27: 243–249	Retrospective cohort of 13 children, GMFCS III with spastic diplegia; Standing from 12-14 months to 5 years in custom-fabricated stand-er in approx. 300 abduction for each leg; 70-90 minutes per day, split into 2 sessions; All children walked with mobility aids between 30-36 months; Outcome measures = goniometry ROM	Baseline = 420 Outcome = 430 GMFCS level remained at III Authors conclude that it is possible to maintain hip abduction ROM with a daily standing programme, although causal relationship cannot be assumed.
2	Macias-Merlo et al (2015)b Effects of the standing program with hip abduction on hip acetabular development in children with spastic diplegia cerebral palsy Disability and Rehabilitation DOI: 10.3109/09638288.2015.1100221	Retrospective cohort of 26 children, GMFCS III with spastic diplegia: 13 in study group; 13 in control group Study Group: Standing from 12-14 months to 5 years in custom- fabricated stander in approx. 300 abduction for each leg; home visits and weekly PT ses-sion; 70-90 minutes per day, split into 2 sessions; Control Group: No stander; 3 x weekly PT session; Children from both groups walked with mobility aids between 30-36 months; Outcome measures = comparison of study group and control group hip MP via radiography.	All children in study group had MPs of both hips <33% 8 children in control group had 1 hip with MP >33% Authors conclude that abducted standing maintains hip symmetry and MP in stable range.
2	Martinsson & Himmelmann (2011) Effect of weight-bearing in abduction and extension on hip stability in children with cerebral palsy Pediatric Physical Therapy 2011; 23: 150-157	Case series design with experimental group: (surgery and abducted standing n=3) and (abducted standing n=11). Control group: (surgery and regular standing n=20) and (regular standing n=63). CP, GMFCS III-V (although most were IV-V), age 2-6 years; Standing for 30-90mins per day for 1 year in R82 Gazelle in 300 abduction for all but 1 child; Outcome measures = goniometry and MP.	Surgery and abducted standing (Study Group 1 n=3): mean reduction in MP of 20.8% Surgery and regular standing (Control Group 1 n=20): reduction in MP not reported as % Abducted standing (Study Group 2 n=11): mean reduction in MP of 8.6% Regular standing (Control Group 2 n=63): reduction in MP not reported as % Best results for 3 children in experimental post-surgery group. Authors conclude that standing for 1 hour per day may reduce MP after surgery.

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Discussion

1. Macias-Merlo et al, 2015a

   In the first study, Macias-Merlo, et al., (2015a) used goniometry to measure the hip abduction of 13 children (9 boys and 4 girls), all GMFCS level III with spastic diplegia, between 12-14 months, and again at 5 years old. Children stood using a custom fabricated stander for two 35-45 minute sessions, 5 days per week, and one 35 minute session on weekend days. The purpose of the study was to show whether standing in hip abduction could affect hip ROM, and the clinical implication is that maintenance of hip ROM leads to reduced contractures and improved mobility. Although only recently published, the work appears to be a subset of data from work which began in 1995, and which was presented as a poster by Macias (2005). This explains the retrospective nature of the study.

There are a number of concerns with this study, including the lack of control group, small numbers, and unreliability of goniometry, which are recognised by the authors. Selection bias is evident in that children were excluded if they were not cognitively able to cooperate with the programme, or their parents were perceived as unable to comply.

Home visits by a physiotherapist were carried out every 4-6 weeks to ensure compliance with the programme, but it is not clear if this represents a standard therapy procedure for all children in standing programmes, or an additional input as part of the research. The repeated goniometry measurements were taken by one physiotherapist, but it is not known if this therapist was the same as the one carrying out the home visits.

Most significantly however, the authors have made an assumption that sustained abducted stretch of tight adductor muscles is solely responsible for prevention of contractures and maintenance of hip ROM. As all children in the study walked with mobility aids (posterior walkers or crutches) between 30-36 months, and no ROM measurements were taken immediately prior to or after this time, the impact of mobility on hip ROM cannot be determined, nor discounted as a major confounding variable. The importance of weight- bearing for hip development is advocated by Willoughby, et al., (2012) who, in their randomised controlled trial, found that botulinum toxin and SWASH abduction bracing did not reduce the need for, or complexity of, surgery in children with bilateral spastic CP. They concluded that surveillance and surgery enable hip integrity, and suggest that weight-bearing and functional movement may have a role in preventing secondary changes to the hip.

The study by Macias-Merlo et al concluded that hip ROM could be maintained, and contractures prevented over a four year period, but the authors appear to suggest that the quality of gait of the children also improved, as they comment on the lack of scissoring observed in the children at 5 years old. Including formal assessments of quality of gait with appropriate outcome measures would have made this comment more valid and less observational. However, the absence of a control group in any case, makes such an inference impossible to substantiate.

If readers are led to infer improved mobility, they may also conclude that maintaining hip ROM leads to improved hip integrity.

Other researchers have found ROM to be a poor indicator of hips at risk from dislocation (Hagglund et al, 2007), while GMFCS, age, initial MP and head-shaft angle (HSA) have recently been found to be significant predictors of developing a MP greater than 40% within five years of the first x-ray (Hermanson, et al., 2015).

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While any intervention which preserves dynamic weight bearing is desirable, as children with spastic diplegia are considered to be at intermediate risk of hip dislocation (Hagglund et al, 2007), a direct connection to between abducted standing and maintenance of hip integrity cannot be made, nor can a causal relationship between abducted standing and hip ROM be assumed, as acknowledged by the authors.

2. Macias-Merlo et al, 2015b
   In this second study by Macias-Merlo, et al (2015b), the authors used radiography to measure the hip migration percentage (MP) at 5 years old of 13 children (9 boys and 4 girls) using a custom fabricated stander, and 13 children (8 boys and 5 girls) not in a standing programme. All children were GMFCS level III with spastic diplegia. Those in the study group were enrolled at 12-14 months of age and stood for two 35-45 minute sessions, 5 days per week, and one 35 minute session on weekend days, as well as receiving home visits and a weekly physiotherapy session. The control group did not receive a stander or home visits, but received physiotherapy 3 times per week. The purpose of this study was to show whether a programme of abducted standing could improve hip MP of children with CP compared to those not receiving abducted standing intervention. This retrospective study appears to be a separate analysis of the same group of children reported on in the previous research, and which formed the poster presentation mentioned earlier (Macias, 2005).

Similar to the previously discussed published work by this author (Macias-Merlo, et al., 2015a), there are concerns regarding the small sample size and selection bias of study group participants. Nonetheless, the inclusion of a control group and use of radiographic outcome measures strengthen this 2015b study. A senior radiographer evaluated all x-rays, although measurement error is not recorded. While concurrent therapies are recorded for both study and control groups, the difference in standing intervention (the control group did not stand at all), home visits and physical therapy between the two groups introduces additional intervention variables which mean like-for-like comparison of single components of the treatment regimens cannot be made.

The results show that all children in the standing group had MP of less than 33% at 5 years old, while 8 children in the control group had one hip with MP of more than 33%. However, baseline MP is not recorded for any of the children, nor is MP established at the start of ambulation, significantly weakening the validity of the results. Macias-Merlo and colleagues acknowledge the need to identify the role of mobility in preventing hip deterioration in further research. The allocation of children to study and control groups was not randomised, with children with learning difficulties excluded from the study group. It is a possibility that these children may have been placed in the control group, and may have been less ambulant, or less motivated to be ambulant. An activity level record would have helped to illuminate differences between study and control groups.

The authors observed the walking of children from both study groups and noted (as in the previous research) that the study group children did not scissor their feet at 5 years old.

Unfortunately a typographical print error in the publication means the comparative gait quality of the control group is not clear. However, as with the previous work, gait quality is not part of the study protocol, and appropriate outcome measures are not employed.

The authors' use of 33% as a cut-off point for hip stability is not consistent with the 30% figure found by this review (Spiegel & Flynn, 2006; Hagglund, et al., 2014). While a lower MP cut-off does not change the study group results, it may have increased the numbers of children in the control group whose MP was considered to be in the 'at risk' subluxation category, further supporting the outcomes. The statistically significant asymmetry of MP of the left hip noted in the control group was an interesting finding for which there was no attempt to rationalise. Given the small numbers in the study, this finding may be down to coincidence. Baseline measurement of MP may have also helped to explain this finding.

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The outcomes of this study do appear to show that the children in the study group had better hip integrity and symmetry than their control group counterparts. However, due to the lack of baseline measures, the unknown influence of mobility on hip integrity, small study numbers, selection bias, difference in interventions and lack of randomisation, these results cannot be wholly attributed to the abducted standing programme. It is possible that multi- factorial influences are at play, of which abducted standing may be a part.

3. Martinsson and Himmelmann, 2011
   In the third study, Martinsson and Himmelmann (2011) used radiographic measurement of MP and goniometry to measure hip abduction and hip/knee extension of two study groups (14 children) and two control groups (83 children). The study groups were post-hip surgery and abducted standing (SG1 = 3 children) and abducted standing only (SG2 = 11 children); the control groups were post-hip surgery and standing (CG1 = 20 children) and standing only (CG2 = 63 children). All children were non-ambulant, GMFCS III-V, aged 2-6 years with spastic, dyskinetic or mixed CP, who stood for up to 1.5 hours daily for one year. The purpose of the study was to determine the effect of abducted standing on hip integrity post-surgery, and as a preventative measure. The clinical implication is that abducted standing may prevent the need for, and/or enhance the outcomes of, surgery.

Again, there are a number of methodological and reporting concerns with this study. While the use of control groups is positive, the imbalance of numbers between study participants and controls (3 vs 20 and 11 vs 63) means direct comparison may not be reliable as the likelihood of identifying unfavourable outcomes increases with sample size. Another confounder may have been that, with 8-10 hrs per week, stretching in the study groups was exceeding the standard of care that may have been provided to the control groups. In addition, the study groups received additional intervention (visits and telephone calls) which could have resulted in a Hawthorne effect in the study group families.

The duration of regular standing in the control groups is not recorded, therefore poorer compliance to the recommended standing duration of 1-1.5 hours may be a confounding factor.

Concurrent therapies in the study groups are neither controlled nor restricted, while in the control groups, concurrent therapies were not recorded. In addition, four children received botulinum toxin injections in their hip adductors. The authors claim that this did not translate into increased ROM, but it may have enabled an abducted standing position to be sustained more easily.

Dr Andreas Kannenberg MD, PhD (Executive Medical Director, Otto Bock North America) reviewed the study's statistical analysis and observed that there was no direct statistical comparison of the mean improvement in the MP between the study and control groups, and only the mean values of improvement in the study groups were reported. The mean decrease in the MP produced by abducted standing in both study groups was 8.6% which is very close to the reported measurement error of ±8%. In addition, the reported correlation between surgery and MP is bigger (11.9%) than that of abducted standing.

Dr Kannenberg further noted that the 3 children in Study Group 1 started with very different hip migration percentages (40%, 60%, 85%), but had almost the same migration percentage (20-25%) at the end of the study. This strongly suggests that surgery may be the main contributor to the improvement rather than abducted standing.

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Given that 7 of the children in Control Group 1 (surgery and standing without abduction) showed as much or greater improvement in MP than that of the 3 children in Study Group 1, not only do the problems with representativity of very small groups become evident, but weight is added to the argument that surgery could be the most significant intervention, with the method of standing being less important (personal communication, 10th November 2015).

The positive influence of surgery in preserving hip integrity has been noted in other research, as discussed earlier (Willoughby et al, 2012). In addition, recent results from a 20-year hip surveillance programme in Sweden suggest that with regular monitoring, and early surgical intervention when indicated, the incidence of hip dislocation in children with CP can be dramatically reduced (Hagglund et al, 2014). Based on the outcomes of this study, hip integrity cannot be reliably attributed to standing in abduction.

Conclusion

While there is much contemporary focus on the benefits of abducted standing for hip stability of children with CP, there is a dearth of research which means it is not possible to draw firm conclusions at this stage. Although many of the difficulties of carrying out research with children with CP are acknowledged by the authors, the very small numbers of children, as well as methodological and reporting concerns mean that clinical restraint must be exercised.

However, this does not mean that passive stretch as part of postural management programmes should be abandoned – it simply means that the sub-group of children for whom it is effective is not fully defined, and therefore an individualised approach should be used (Gough, 2007).

This client-centred approach is consistent with the principles of evidence-based practice (Sackett, et al., 1996). It is likely that standing in abduction is beneficial for some children.

At the moment, the evidence to date suggests that abducted standing may have a support role in the development of hip integrity and symmetry for ambulant, cognitively intact children at GMFCS level III.

Children at GMFCS levels IV and V who have undergone surgical intervention may benefit from post-surgical standing, but it has not yet been determined whether abduction or regular standing is most effective. In addition, the most effective angle of abduction for MP improvement is not yet defined (Martinsson & Himmelmann, 2011).

The long-term outcomes for the children in these studies is not yet fully clear, and larger studies with randomisation, more stringent controls on duration and angle of standing, as well as restrictions on concurrent interventions are required to further clarify the potential benefits of abducted standing and the group(s) of children who may benefit the most from it. Until then, alongside a hip surveillance programme and timely surgical intervention, clinicians should continue to use their professional expertise to determine the most appropriate intervention for each child.

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Declaration of interest

- Leckey design and manufacture a range of standing frames, currently without 600 total bilateral hip abduction.
- VIDA, which is a member of the Leckey group, distributes the EasyStand Zing in the UK, a product with 600 total bilateral hip abduction.
- Dr Andreas Kannenberg is a Certified Family Practitioner and the Executive Medical Director for Otto Bock North America. Otto Bock distributes Leckey products within North America.
- Clare Canale is an Occupational Therapist and the Clinical Research Manager for Leckey.

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