Effects of Surgical Adductor Releases for Hip Subluxation in Cerebral Palsy: An AACPDM Evidence Report

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N. Susan Stott M.D, Ph.D., FRACS
Louis Piedrahita MD

Approved by

AACPDM Treatment Outcomes Committee Review Panel: American Academy for Cerebral Palsy and Developmental Medicine, 6300 North River Road, Suite 727, Rosemont, IL 60068-4226, USA.

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Greg Liptak MD		

Correspondence to first author: Dr Susan Stott, Department of Surgery, Faculty of Medicine and Health Science, The University of Auckland, Private Bag 92019, Auckland, New Zealand. E-mail address: s.stott@auckland.ac.nz.

Objective

The objective of the American Academy for Cerebral Palsy and Developmental Medicine (AACPDM) evidence reports is to provide the biomedical research and clinical practice communities with the current state of evidence about various interventions for the management of developmental disabilities. The AACPDM evidence reports aggregate all that has been published about outcomes of an intervention for a medical condition, gauge the credibility (i.e. strength of the internal validity) of that evidence and identify gaps in out scientific knowledge.

The AACPDM reviews are not evidence-based "practice guidelines". As yet, the bodies of evidence in developmental disabilities are neither robust nor comprehensive enough to allow confident generalization to groups of people at large, a prerequisite for evidence-based practice guidelines. Moreover, absence of evidence of effectiveness in an evidence report should not be construed as proof that a treatment is not effective; rather, it may reflect areas in which more meaningful research is needed. In the meanwhile, clinicians must be circumspect about their treatment recommendations, relying on current "best evidence" to inform individual choice[a].

[a]Best evidence is represented by the study (or studies) in the evidence report that most closely approximates the patient characteristics of interest to the clinician, that used a therapeutic regime most like the one the clinician can provide, that investigated outcomes greatest concern to this patient, and that provides the most credible or internally valid results.

Disclosure

Every effort has been made to assure that AACPDM Evidence Reports are free from any real or perceived bias. The Academy's editorial review panel is a multidisciplinary group comprised of the current members of the AACPDM Treatment Outcomes Committee who serve a five year rotating term. This Committee may invite up to two additional reviewers to encourage substantive input by knowledgeable proponents of all points of view. Potential conflicts of interest by authors and reviewers have been disclosed and are documented in the AACPDM Database of Evidence Reports. The Treatment Outcomes Committee is charged and overseen by the AACPDM Board of Directors with the task of developing evidence reports and operates under an approved methodology of systematic review of the scientific literature and under approved procedures (1,2). The Board grants final review and sanction for each report.

Consensus Process

The review authors organize intervention outcomes in a predefined manner and answer predefined questions to describe scientific evidence. Members of the editorial review panel give their input and resolve any differing opinions to reach agreement about statements made therein on behalf of the Academy. Nevertheless, the data in an AACPDM Evidence Report can be interpreted differently, depending on people's perspectives. Please consider the conclusions presented carefully.

Adductor Release for Prevention of Hip Subluxation in Cerebral Palsy

HIP SUBLUXATION AND DISLOCATION:

Hip subluxation and dislocation is a common feature of cerebral palsy. The hip joint is normal at birth but the spastic muscle imbalance and lack of weight bearing leads to the development of progressive structural changes around the hip joint (3,4). These include excessive femoral anteversion, posterolateral acetabular dysplasia and flexion-adduction contractures (5-11). These deformities contribute to a shift in the mechanical axis away from the center of the femoral head to the lesser trochanter, leading to posterolateral migration of the femoral head and subsequent hip subluxation and dislocation (3,8).

There is general consensus that the incidence of hip dysplasia increases as the level of neurological involvement in cerebral palsy increases (3,4). The consequences of hip subluxation in early childhood are far-reaching, with progression to hip dislocation in 10 to 59 percent of patients (7,12-16). This occurs on average by the age of 7 to 8 years (7). Reports of the natural history suggest that, if left untreated, hip dislocations may be the source of significant pain and disability in approximately 50% of patients (13,17,18). Other potential consequences of a hip dislocation include difficulty with toileting and perineal hygiene and the development of pelvic obliquity causing problems with sitting (7,13). Scoliosis is also common in children with cerebral palsy who have dislocated hips, however the temporal and structural relationship between the two deformities is not clear (3,14,19). In some patients, hip dislocation occurs before pelvic obliquity and scoliosis while other patients develop a scoliosis and pelvic obliquity which then can lead to hip dislocation (14,16,20).

TREATMENT OPTIONS FOR HIP SUBLUXATION

The term "hip at risk" was first coined by Pollock and Sharrard in 1958 (21), who noted that a limitation of hip abduction with an associated flexion deformity, were indicators of hip instability. This led to widespread screening of patients with cerebral palsy and a recommendation for referral if hip abduction in extension was less than 30 degrees. It is generally believed that conservative measures have little success in preventing hip subluxation. Obturator nerve blocks (22) and intramuscular alcohol (23) have been used to reduce adductor spasticity, but there are no reports of use of these agents to prevent hip subluxation. There is, as yet, no data on the effect of intra-thecal baclofen on spastic hip subluxation, however, a recent trial of botulinum toxin injections into the adductors together with abduction bracing has produced some encouraging preliminary results(24). At the present time, however, surgical management remains the mainstay of treatment with options including either early soft tissue release or bony surgery such as femoral osteotomy, or, in more severe cases, open reduction of the hip and pelvic reconstruction (25,26).

ADDUCTOR SURGERY

Tachdjian and Minear 1956 (27) were among the first to suggest that hip subluxation and dislocation may be a preventable condition with soft tissue releases of the adductor muscles,. This surgical prophylaxis was based on the concept of muscle imbalance around the hip joint, with relative weak abductors and extensors and strong adductor and flexors (7,27-30). Early authors favored an open adductor release as opposed to the subcutaneous type and routinely performed a tenotomy of the adductor longus and adductor brevis with resection of at least one inch of the anterior obturator nerve and its branches (31). If the gracilis muscle was tight it was released as well and, if contracture persisted, a section of the most anterior fibers of the adductor magnus was performed. Although anterior obturator neurectomy and adductor myotomy was the recommended approach for many years, (30-32) its use is now more limited due to concerns about overweakening of the adductors with the development of possible abduction contractures (33). Bilateral instead of unilateral procedures are recommended because of the possible development of contralateral hip subluxation and windswept deformity(34).

In 1969, Stephenson and Donovan described their results with adductor transfer in spastic hip subluxations(35). Many transfers were tried with the intention of improving function by removing a deforming force and redirecting it to the relatively weaker extensor muscle group(36-38). Goldner 1981 and Reimers 1984 (38,39) challenged this hypothesis using the simpler adductor tenotomy procedure with significant less complications and similar functional results. Adductor transfers are now an uncommon procedure for patients who have cerebral palsy. However, adductor release and all the variants, continues to be a popular procedure to prevent and treat hip subluxation secondary to spasticity (4,33).

METHOD OF REVIEW

Inclusion Criteria

This review includes studies of patients with cerebral palsy in which the intervention was: 1) Specified as adductor releases. 2) Could be identified as adductor releases (based on the description of the surgical procedure). 3) Was said to be adductor releases but combined with other soft tissue releases around the hip joint. 4) Specified as adductor release and compared to other type of intervention. Studies that used bony surgery in all patients in addition to the soft tissue release were excluded from the review. Studies reported in languages other than English were also excluded.

Literature Search

The literature search included MEDLINE (1966 to the present), HealthSTAR (1975 to December 2001), CINAHL (1982 to September 2002), Best Evidence (1991 to 2001), EBM Reviews (3rd Quarter 2002) and The Cochrane Library. The electronic search terms were: "Spasticity", "Adductor" and "Hip" and "Cerebral Palsy." The list of articles obtained was analyzed and their reference list was carefully scrutinized for the inclusion of other potential articles. Seventy-eight citations were examined. Fifty one articles were excluded for one or more of the following reasons: 1) The article is a review or is a descriptive article and contain no specific database of a group of patients with cerebral palsy and hip subluxation treated with some form of adductor release. 2) The article describes adductor tenotomy as an intervention but applied to a group of patients with a diagnosis other than cerebral palsy. 3) The article is not in English. Twenty seven citations were included in this review.

CLASSIFICATION OF THE RESULTS

All reported results of adductor surgery were classified on the basis of 1) dimensions of disablement (i.e., what kind of evidence there is) and 2) levels of evidence (i.e., how strong the evidence is).[b]

Dimensions of Disablement is a concept and a classification system that facilitates the measurement, management, and research of rehabilitation outcomes and mimimizes the barriers between medical and social models of rehabilitation (Table I). It describes the effects of disablement (and interventions) in five dimensions: cellular and molecular physiology; body parts and systems; human activities; fulfillment of gender and societal roles; and in the dimension outside the individual including the family circumstances, prevalent societal attitudes, social policies, architectural barriers**.** The dimensions of disablement used in these reviews reflect the terminology used in two classification systems, the 1997 revision draft of the 1980 World Health Organization International Classification of Impairment, Disability and Handicap (ICIDH-2)(40) and the National Center for Medical Rehabilitation Research (NCMRR) model(41). The World Health Organization has recently

[b] The rationale and specific guidelines for classifying the treatment outcomes are available on the Academy's Internet web site at www.aacpdm.org in the document titled "AACPDM methodology for Developing Evidence Tables and Reviewing Treatment Outcomes Research".

published the final draft of their revisions, International Classification of Functioning, Disability and Health (ICF)(42). The Treatment Outcomes Committee intends to revise the levels of disablement currently used in the review methodology to reflect the terms of the new ICF.

Levels of evidence classifications and other quality-rating schemes are based on: 1) a hierarchy of research designs that range from the strongest to weakest according to ability to reduce bias, and/or 2) a means of assessing the scientific rigor of the conduct of the particular research study.[c] Generally speaking, Level I research designs contain the most scientifically rigorous methods, can produce the strongest evidence, and thus, can yield the most definitive results(43). Level II designs are less scientifically rigorous so can produce, at best, less convincing evidence; thus their results must be regarded as only tentative. Levels III and IV can produce still less persuasive evidence with results that can merely suggest causation. No conclusions regarding treatment effectiveness can be drawn from Level V evidence as it contains no before and after documentation or control of other variables that may account for the outcomes observed. However, even though a study may employ a Level I design, the actual conduct of the study may not implement all the controls for threats to validity under the design. Thus, evaluation of the conduct of the actual study is important.

The AACPDM levels of evidence classification (Table II) rates each study on the basis of its research design (Part A) plus a study quality indicator that reflects how well threats to validity appeared to be controlled within the parameters of the research design used (Part B). Unlike some other classifications, the AACPDM levels of evidence evaluation is limited to gauging only the internal validity of a study, i.e., its ability to attribute the observed outcomes to the intervention in that study. External validity, or the confidence with which a finding can be generalized, is more appropriately determined by individual users of the evidence reports. These users can focus on specific aspects of similarity between a patient of interest and the subjects reported in the study (e.g., their age; type and severity of cerebral palsy, conditions of treatment etc.).

Summary tables

Table III INTERVENTIONS AND PARTICIPANTS

Table III summarizes the interventions and participants in the 27 studies included in the review.

TABLE IV RESEARCH METHODS

Table IV summarizes the research methods used in the 27 studies. Levels of evidence were determined by the type of research design (Level I-V) plus a judgment about the degree to which the particular study controlled threats to internal validity within its design parameters (S for Strong, M for Moderate, W for Weak)[d]. Level V studies are not subjected to judgment about how well they controlled for threats to validity because there are no controls. Only two studies of adductor surgery in individuals with CP have included a control group. In both cases, the controls were historical and the indications for conservative management not well defined.

TABLE V (PARTS A): OUTCOMES, MEASURES, AND RESULTS

Table V, in two parts, summarizes the results from the 27 studies. Tables V: Part A and Part B show the coding of each result for the dimension of disability and level of evidence that it represented.

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[c] The concept of a 'quality determination' for articles used in systematic reviews is a matter of some debate. The science of critical appraisal of research, initially developed in internal medicine, is an on-going process. It is additionally difficult to apply this concept to research about disabling conditions in developing children. Despite the considerable challenge, there is agreement that teams developing systematic reviews can take certain steps to ensure that their approaches to grading the quality of research results meet current scientific standards.

[d] THE AACPDM methodology is based on current scientific standards for analyzing and weighting studies for bias and error and for judging study methods. Nevertheless, this type of critical appraisal is a new endeavor in medicine, in general and within the Academy, in particular. The AACPDM methodology will continue to evolve both with experience and as the science of critical appraisal improves. Therefore, the assigned level of evidence should be regarded as an estimate, and relative to other studies, rather than an absolute.

Some studies reported the results as group data, i.e, results that reflect the average effect of adductor surgery in a group when pre- and post-op status is compared. Other studies have also reported results according the uniformity of effect within the group, i.e., the change in status of individuals. Part A of the table includes 45 results that reflect comparisons of group averages; Part B includes 52 results that reflect uniformity of effect within the group.

Clinical importance or relevance (seldom explicit in studies) is reported when stated in the articles or when it was apparent to the reviewers. Statistical information is specified to the extent provided but has not been checked by an independent statistician. Each result is assigned the level of evidence derived from the study that produced it, (see Table IV), except for anecdotal evidence, which is coded as level V. When there is an absence of controlled observations, anecdotes can be useful to, at least, document that an outcome has been observed in association with the intervention. Anecdotal reports of outcomes can be useful in planning further studies.

Evidence Table

Table VI (PARTS A AND B): ORGANIZATION AND INTERPRETATION

The evidence table is displayed in two parts to accommodate these two different types of results. Interpretation can be made visually with this table or more in depth interpretation is possible by referring back to the summary tables.

Part A of the evidence table aggregates the 45 group average results produced by 12 of the studies. Each outcome is indicated by a superscript that is the citation number of each study that produced this result associated with a level of evidence (coded I-V for type of research design + S, M or W for strong, moderate or weak controls against threats to validity of the study). By rows, one can see which dimensions of disability have been targeted for investigation and which types and how often outcomes have been measured. For example, the range of hip abduction following hip adductor release has been investigated six times in six different studies (10,30,44-46) and all six studies have shown improvement compared to the average pre-op hip abduction measure. None of the studies have been investigated by statistics and a significant change has not been shown in any of the studies. The confidence that can be placed in this data is low. All but one of the studies are graded level IV+W indicating that the studies do not have a control group and are weakly controlled against internal threats to validity.

Part B aggregates the 52 uniformity of effect group results for 20 studies that reported effects for individuals in the group. Each outcome is indicated by the number of participants in the subgroup and is followed by a level of evidence and citation for the study that produced the result. For example, by rows one can see that the uniformity of increase in hip abduction range after adductor surgery has only been investigated in one study (47). In this study, 31 out of 43 hips gained hip abduction after adductor surgery. However, seven out of 43 hips lost hip abduction and another five hips had no long term change in hip abduction range. This data comes from a level IV+W study and the numbers studied are small. Thus the strength of evidence gathered from the uniformity of effect studies regarding changes in hip abduction following adductor surgery is very weak, despite the overall positive result.

GREATER ELABORATION OF THE EVIDENCE

One can delve into any data in the summary tables by using the superscript citations to refer back to the summary tables and thus elaborate the meaning of a data point. For example, in the row that summarizes the effect of adductor surgery on radiographic hip subluxation/dislocation, one entry (30) represents the strongest evidence that soft tissue hip adductor surgery can reduce the numbers of hips that are subluxated or dislocated at follow-up. Referral to Table III shows that this information comes from a study reported in 1975 that contained 69 children (30). Fifty-one patients in this study had spastic quadriplegia, 16 had spastic diplegia and 2 had hemiplegia. The intervention was open adductor surgery (134 hips) with additional anterior branch obturator neurectomy in 46/134 hips and iliopsoas lengthening in 3/134 hips. Table III lists the controls as 36 people who had been treated earlier than the operated group. A similar number of individuals in the control group had spastic quadriplegia, however the initial hip status in the control group is not defined. From table IV, this evidence can be further elaborated as follows. It came from a cohort study design with a historical control group and was coded as a level III study. The conduct of the study, however, was graded as weak and no statistics were performed. Final elaboration of the results (see Table V, Part A) shows that, compared to historical controls, the surgical group had less hips subluxated or dislocated at follow-up, improved range of hip abduction and less hip flexion contracture. Hip pain was also reduced in the surgical group but this was an anecdotal level V observation. Table V, Part B breaks down the results for the surgical group and shows that 67/115 hips had an improved result, 2/115 hips had a worse result and 46/115 hips were unchanged. A referenced footnote alerts the reader to the fact that the authors of the study could only obtain radiographs for 115 hips of the 134 hips treated surgically. All these aspects of the study need to be taken into account by the reader when interpreting the internal validity of the study.

CAUTION INTERPRETING RESULTS

Caution is advised in the interpretation of results that are not statistically significant (NS). Results may be NS because of lack of adequate power in the study sample and design. The power of a study is the probability that the study, given its design and sample size, can detect a true difference of a pre-determined magnitude (effect size). In the absence of a power calculation, there is always the possibility that a true difference existed between the two treatments being compared, but that there was inadequate power to detect a difference. However, if a power calculation is reported and the sample size needed to produce the power is obtained, then a NS result statistically supports the conclusion that there is no difference between the two treatments compared. Unfortunately no study in this review reported power calculations and sample sizes were generally small, drawing into question whether adequate power was achieved.

Medical complications table

Table VII aggregated the medical complications and adverse effects that have been reported.

ANALYSIS OF EVIDENCE ABOUT ADDUCTOR RELEASES

1. *What kind of evidence is there about effects on adductor surgery on aspects of impairment?*

    Hip subluxation: Table VI, Part A shows that five studies have documented the overall effect of hip adductor release on the number of hips subluxated or dislocated at final follow-up (7,30,48,49). Two studies showed improvement in hip subluxation (30,50), one study showed deterioration in hip subluxation (7) and another study showed no change (49). The final study in this group looked at the effect of unilateral adductor surgery noting an adverse effect on the contralateral hip (48).

    The uniformity of effect of adductor surgery on the number of hips that are subluxated or dislocated at follow-up has been investigated in eight studies (10,30,47,48,51-54). These eight studies showed an overall improvement in 168 out of 530 hips (32%), however, fifty-three hips were worse and 309 hips unchanged. As a corollary to this, five other studies reported that a total of 70 out of 324 hips undergoing adductor release required further surgery for hip subluxation and were deemed to be failures of the initial procedure (9,45,46,55,56). The definition of hip subluxation and dislocation has varied between studies, making direct comparisons of the data difficult to interpret. However the data does suggest that a significant percentage of hips that have soft tissue adductor release for subluxation will require further surgery, or will deteriorate following the initial surgical procedure.

The most common radiographic outcome measures used to assess subluxation were the migration percentage (9), the center edge index (57) and the acetabular index. Two studies reported an overall improvement in Reimers migration percentage following adductor surgery (9,45) but another study found no difference in the migration percentage compared to an untreated control group (49). Overall, Reimers migration percentage improved in 241 of 467 hips (51%) undergoing bilateral hip adductor releases and deteriorated in 118 of 467 hips. 108 of the 467 hips were unchanged after bilateral hip releases. Two studies examined the results of unilateral adductor releases and the effect on the contralateral hip (34,55). Unilateral adductor release was associated with deterioration in the contralateral hip in 15 out of 26 hips,(34.55). Silver et al 1985 (55) found improvements in migration percentage in ten of 11 ipsilateral hips undergoing unilateral release but in only one of the 11 contralateral hips.

Three studies (10,44,47) reported an overall improvement in the center edge angle following adductor surgery. Overall, 63 of 126 hips (50%) treated by adductor surgery showed improvements in the center edge angle, while 31 hips deteriorated and 32 hips showed no change (11,37,53,58). Two studies reported overall improvements in the acetabular index following adductor surgery (10,45). The acetabular index also improved in 17 out of 47 hips but was unchanged or worse in 30 out of the 47 hips (37,53)

Overall these results suggest that radiographic improvements can be measured in about 50% of hips following soft tissue adductor release. However, it was not clear from the current studies if this radiographic improvement was of a large enough change to be clinically important. For example, in one study, significant changes in the acetabular index were reported with a p value < 0.01 however the average change was only 3 degrees (from 24 degrees preoperatively to 21 degrees postoperatively) (53).

Range of motion at hip. The post-operative range of hip abduction and the presence of hip flexion contractures were the most commonly assessed measures in this category. The overall effect on hip abduction was documented in six studies. All six studies reported that post-operative hip abduction was increased compared to the pre-operative range (10,30,37,44-46). Only one study provided more detail regarding the uniformity of the effect of adductor surgery on hip abduction range of motion (47). In this study, 31 out of 43 hips improved in abduction range while 12 hips were unchanged or deteriorated. Hip flexion contractures showed an overall decrease postoperatively in two studies (30,37) with documented improvement in 29 out of 43 hips in one level IV study (47). Other passive range of motion measures that were tested included the popliteal angle (45), extension of the hip (44) external rotation of the hip (45) and internal rotation of the hip (45). No change was found in hip extension, hip internal rotation or hip external rotation however an overall improvement was found in popliteal angle in a single study (45).

As the studies were predominantly retrospective, it was not possible to determine how these measures had been obtained. Those studies that did document range of motion did not outline the testing procedure or describe who carried out the measures. None of the studies discussed inter and intra-observer errors in goniometric measurements, which have been reported to be up to 20 degrees in the cerebral palsy population (59). Thus, although the evidence suggests that hip abduction range is improved by hip adductor release, the conduct of the studies is too weak to draw a firm conclusion.

Pelvic obliquity/Scoliosis: Only one study has reported the overall effect of adductor surgery on the development of scoliosis (46). This study found that scoliosis had worsened in their population of 30 children with spastic quadriplegia during an average of six years follow-up. Pelvic obliquity likewise deteriorated over the average six years of follow-up (46). In contrast, Feldkamp and Denker 1989 (44) reported improved pelvic symmetry following hip adductor releases but did not do any statistical analysis (44). Carr and Gage 1987 (34) reported that unilateral hip adductor release led to pelvic obliquity in 7/14 patients.

Hip Pain: Long term pain in the hip was investigated by only two studies (30,49). In a level III study, Bagg et al 1993 (49) found that patients who had soft tissue adductor releases were just as likely to have pain in the hip as patients who were untreated at long term follow-up. The presence of pain in their patient population correlated well with the presence of degenerative joint disease or hip dislocation on the hip radiographs. The only other study to look at the presence of pain reported anecdotally that pain was reduced in their patients that had soft tissue adductor releases compared to historical controls (30). As this data was not quantified in any way and represented an anecdotal observation it was graded level V.

2. WHAT EVIDENCE IS THERE ABOUT EFFECTS IN DIMENSIONS OF DISABLEMENT OTHER THAN IMPAIRMENT?

Pathophysiology: There is no evidence regarding effects on cellular or molecular structure or function

Functional Limitation / Activity

The Functional Limitation/Activity dimension is concerned with functional activities themselves, such as sitting, walking, dressing, playing or interaction with other people. Although many studies mentioned these as positive benefits of surgery (11,49,55), most studies did not report the effect/s on functional activities. In particular the effect on sitting stability and ability to transfer was considered by only four studies (37,47,48,53). Moreau et al 1995 (47) conducted a questionnaire to assess the effect on standing and sitting and used observation by physiotherapists and parents to grade ability to transfer post-operatively and to ambulate. 18 out of 22 parents responded to the questionnaire and reported improvements in standing in 11 patients, in sitting in 8 patients and in walking in 5 patients. Interestingly, the clinical observations regarding changes in sitting and ambulation were different to the parents' perceptions. Only one patient changed from 'propped' to 'independent sitting' postoperatively, all others remaining unchanged. Ambulation did improve by clinical observation in 12 patients, however, this was mostly in the sub-group of patients with the diagnosis of spastic diplegia. Significant rehabilitation was also carried out with potential ambulators "being kept in hospital until ambulation began". Root and Spero 1981 (1981) did not document how their data was obtained but commented that sitting balance was improved in 37% of patients postoperatively and standing balance improved in 56% of patients. Two other studies commented that sitting ability had improved in a small number of patients postoperatively but these were both anecdotal observations graded level V (48,53). Several other studies also made anecdotal level V observations of improved function in the areas of gait endurance (37), ability to ambulate (10,51,53) and reduction in postoperative brace use (37).

Disability/Participation

The effects of hip adductor surgery in participation in normal societal roles have not been investigated.

Societal Limitation/Context Factors: Two aspects of effects on the context of the child's life were investigated; ease of toileting the child and caring for perineal hygiene. Many authors commented that hip subluxation and dislocation leads to problems with perineal care and personal hygiene and that this is a reason for adductor surgery (11,45-47,49) Despite this assumption, there was only one study that looked at the effect of adductor releases on perineal hygiene and the ability to toilet the child. This study commented that improved ability to diaper the child was found for 8 of the 18 children (47). In 5 children, there was no change and in 5 the question was said to be not applicable. This data was obtained from questionnaire at either 2 or 5 years after surgery and not all parents answered the questionnaire.

3. WHAT LINKAGES EXIST FOR TREATMENT EFFECTS ACROSS THESE DIMENSIONS?

   No linkages can be determined given the inconsistent findings and/or general lack of treatment effect documented in any dimension.

4. ARE THERE SUBGROUPS IN WHOM ADDUCTOR RELEASE MAY BE MORE OR LESS EFFECTIVE?

   Though the information is not shown in the summary tables, studies analyzed potential factors expected to identify subgroups of children who might experience greater or lesser benefit from adductor surgery. These variables included age at entry to treatment, sex of the patient, type and severity of disability and degree of hip subluxation. No power analyses were conducted to see if the subgroups studied were indeed large enough to analyze statistically.

   Eleven studies analyzed age as a variable (9-11,37,46,52,55,56,58,60). Of these, eight studies could not demonstrate a difference in outcome related to the age at the time of surgery (9-11,37,46,52,55,56,58,60). Conversely, three studies found that younger children did better (9,60,61) with children under four years (9,61) or five years (60) having a better outcome. The sex of the patient made no difference in two studies (9,58), however (56) found that girls did slightly worse than boys with 62% failures in girls compared to 55% failures in boys.

   The type and severity of disability was addressed by three studies. Two studies (30,49) showed that hips of children with spastic diplegia had better outcomes than the hips of children with spastic quadriplegia. In contrast, a third study (45) found that the final hip migration percentage did not vary between ambulatory and non ambulatory patients with a p value>0.05.

   Radiographic measures of preoperative hip subluxation were significant predictors of outcome. Almost all studies found that a smaller preoperative migration percentage was associated with a more successful postoperative outcome defined by arrest or improvement in the migration percentage and avoidance of the need for further hip surgery (45,46,49,52,56,61). Migration percentages less than 30 to 40 % preoperatively were associated with successful outcomes for 75% to 90% of hips (45,52,61). Conversely hips with a migration percentage > 40 to 50% had a more uncertain outcome with 75 to 77% hips remaining subluxated or dislocated (49,52). Cottalorda et al 1998 (46) found that hips with a migration percentage of >40% did not improve radiographically following adductor surgery but that 35% of the hips stabilized.

   Looking at the data from a different perspective, if only hips with good outcomes are considered, the average preoperative migration percentage ranged from 30 to to 39% (45,55,56) compared to hips with poor outcomes, which had preoperative migration percentages of 42 to 58.9% (45,55,56). This difference was significant in two of the three studies (55,56). The center edge angle was also predictive of outcome in two studies, with hips that had center-edge angles of greater than 27 degrees (52) and 30 degrees (46) classified as having poor responses to adductor surgery.

   In the only level III study that looked at migration percentage, Bagg et al 1993 (49) showed that in nine control hips with migration percentages <50%, five had improved at the time of follow-up without surgical intervention. None of the control hips with migration percentage greater than 50% improved spontaneously suggesting that these two groups may have a different natural history. Thus the improvement or stabilisation of the migration percentage seen in most of the studies may reflect the variable natural history of the condition rather than the effect of the surgery (45,49).

The intervention adductor release was also quite heterogeneous across studies and several studies analyzed the effect of different surgical variations on the outcome. Studies varied as to whether the additional procedures made a difference. Additional surgical procedures such as anterior branch obturator neurectomy or ilio-psoas lengthening made no difference in four studies (11,46,52,56). In one study, (60) the additional psoas lengthening decreased the number of hips requiring further surgery to achieve stability from 64% to 28%. Descriptions of the surgical technique were not always reported, making comparisons across studies difficult. Overall, however, there was little evidence that the addition of an anterior branch obturator neurectomy could significantly improve the outcome of the procedure.

WHAT MEDICAL COMPLICATIONS AND ADVERSE EFFECTS HAVE BEEN DOCUMENTED?

Only ten of the 27 studies have documented complications, either systemic or localised. Relatively few complications have been reported. Wound problems, either infection or hematoma, have been reported in 19 patients. All of these resolved without complication. Abduction contracture has been reported in 15 patients, with one study having 12 patients with this complication (11). Almost all patients in the study by Houkom et al1986 (11) had an anterior branch obturator neurectomy followed by prolonged abduction bracing. Compliance with the bracing protocol was not documented but the high complication rate in this study suggests that this rigorous bracing protocol may be associated with this particular complication. More significant complications include avascular necrosis of the femoral head and heterotopic ossification around the hip joint. Manjarris and Mubarak 1984 (62) reported a case of bilateral avascular necrosis of the femoral head that followed bilateral adductor release and iliopsoas release at the lesser trochanter . Sharrard et al 1975 (30) also reported one case of coxa magna following adductor release, which may have been due to unrecognized avascular necrosis of the femoral head. Two case reports have also been published in which heterotopic ossification occurred following adductor release leading in one case to ankylosis of the hip (62,63).

6. WHAT IS THE STRENGTH OF THE EVIDENCE?

Credibility of a body of evidence depends on several factors. These include; the level of evidence or strength of the internal validity of the results; how extensively the population has been sampled (i.e., number of different studies and number of participants); the number of times that outcomes have been measured; and the consistency of results across the studies. A related and important issue is whether the magnitude of change is large enough to be clinically important.

The level of evidence gauges the extent to which the studies are more likely to inform than to mislead in regard to the likelihood that the observed changes are attributable to the interventions and not to extraneous factors. The body of evidence for adductor surgery is composed of very low levels of evidence. There are no Level I or II studies on the subject of hip adductor release for hip subluxation and the majority of studies were coded as level IV. The conduct of the studies was weak and failed to control for possible threats to validity. Common reasons for the judgment of weak internal validity in the studies were related to 1) the samples of children, 2) the intervention 3) the radiographic outcome measures and 4) lack of statistical analysis.

1) The weakest aspect of the studies is the considerable heterogeneity of the study groups. The studies included different individuals who varied considerably in their type of cerebral palsy and its severity, associated disabilities, degree of hip subluxation and age at treatment. Many of the studies included ambulatory children or children with spastic diplegia. These groups of children are thought to have a different risk of hip subluxation to those who are non-ambulatory with spastic quadriplegia. The age of the subjects at treatment also varied considerably, from one year to adulthood. Many studies included individuals with normal hip radiographs but limited hip abduction in their reports. These individuals may well have had a different natural history to those with documented hip subluxation on radiographs. The lack of control subjects also

compromised the studies. There is a lack of knowledge about the natural history of hip subluxation in cerebral palsy. In a study by Miller and Bagg (64), it was shown that not all subluxated hips progress to dislocation. In their study, hips with migration percentages of 30 to 60% have only a 25% chance of significant progression of subluxation. Hips with migration percentages of 60% or greater all progressed to dislocation regardless of the age of the patient. As this was a retrospective study, the exact percentages are open to question. However this study certainly shows that patients with mild subluxation may never go on to more significant dislocation. This throws into doubt the results of follow-up studies that define a stable migration percentage as a successful outcome.

2) Many individuals had had other soft tissue or bony surgery at the same time as their adductor release. The most common of these were anterior branch obturator neurectomy, psoas tenotomy and hamstring lengthening. Such heterogeneity in the intervention can obscure treatment effects when the group is analyzed as a whole. The postoperative management also varied between the studies. A variable length of abduction bracing was undertaken in seven studies (10,11,38,46,47,52,63). As compliance with postoperative bracing was not considered in these studies, it was not possible to assess whether this had influenced the outcome of the surgery.

3) The radiographic definitions of subluxation varied considerably between studies. As well, the hip radiographs were taken at undefined intervals in most studies and were not standardized with regard to position of the limbs. The hip migration percentage described by Reimers in 1980 (9) was the radiographic measure most commonly used to assess the degree of hip subluxation. Authors disagreed however in their definition of subluxation and what constituted a normal Reimer's migration percentage. Some authors defined a migration index of up to 33% as normal (9,52) while others chose 25% (45) or 30% (61). This definition is contrary to the available literature, which shows that migration percentage varies with age and that migration percentage in the young population is closer to 16% (65) The validity of the Reimers index as an index of subluxation has not been measured. Reimers estimated an error of ± 10% based on theoretical errors in goniometric measures, however, subsequent authors have taken a change of 10% as a significant change. Our own unpublished data suggests that changes in measures of up to 20% may occur even when the same observer measures the same radiograph in the same session (personal communication).

The validity of the migration percentage, that is, its ability to truly measure the degree of subluxation of the largely cartilaginous femoral head relative to the cartilaginous border of the acetabulum also had not been tested. This method is based on the width and location of the ossific nucleus of the femoral head in relation with the acetabulum. The ossification center of the femoral head is "notorious for its eccentricity of location relative to the true cartilaginous model of the femoral head and to its delay and variations in the rate and pattern of ossification", (11). Thus changes in the migration percentage could reflect changes in the ossification of the femoral head over time rather than true changes in femoral head coverage.

4). Statistical evaluation was not done in many cases. For those studies with statistics, statistical advice had not been sought. The sample sizes themselves were all relatively small calling into question the power of the studies to detect effects that did exist. Often statistics were applied to subgroups leading to analysis of only small numbers. Power calculations were not reported in any of the studies.

Overall, the weakest aspect of this body of evidence is the considerable heterogeneity of the population that has been studied and the lack of understanding of the natural history of hip subluxation in cerebral palsy. The strongest aspect of the evidence is the relatively large number of children and hips that have been studied with respect to radiographic measures of hip subluxation.

SUMMARY AND DIRECTIONS FOR FUTURE RESEARCH.

The primary goal of adductor release is to prevent hip subluxation in children with cerebral palsy from progressing to hip dislocation with subsequent significant morbidity. To what extent has the role of adductor surgery in 1) preventing hip subluxation and 2) improving the quality of life for children with cerebral palsy and their families been borne out by the available research?

This review of the current literature shows that radiographic hip subluxation was improved in 168 out of 530 hips following hip adductor release with a corresponding improvement in Reimers migration percentage in 241 of 467 hips. However, these results are weakened by the considerable number of concurrent surgical procedures, and by the variability of the patient population studied. A significant number of children still required further hip surgery or ended up with a dislocated hip despite adductor surgery. The number of children with poor long-term outcomes is probably underestimated by the current studies as the majority did not follow the results to skeletal maturity. The authors generally agreed that younger children had a better outcome from adductor release, as did children with normal hips but tight adductors. This raises the issue of the natural history of the untreated cerebral palsy hip (64). Not all subluxated hips in children who have cerebral palsy progress to dislocation (64). Thus without a control group, it is impossible to interpret whether a migration percentage that is unchanged on radiograph or a hip that remains subluxated but does not progress to dislocation, is a good result at follow-up or a result that would have occurred without treatment.

Prevention of hip dislocation has a number of potential benefits including reduced pain, improved perineal hygiene, increased ROM at hips, easier seating and prevention of pelvic obliquity/scoliosis. However the evidence that these benefits were obtained was lacking in most studies. Most of the data relating to these aspects of disability was presented as level V anecdotal evidence with the exception of one small study (47).

Overall, there are substantial gaps in the body of evidence that need to be addressed by future research before one can determine whether there is sufficient reason for choosing adductor release over another intervention for the treatment of hip subluxation in cerebral palsy. Given the low frequency and variable severity of cerebral palsy, multi-center studies would probably be needed to recruit adequate homogeneous samples. Control groups must also be included where ethically possible to allow discrimination between the natural history of the condition and the effects of the intervention. The reliability and validity of the available measures to document hip subluxation such as the migration percentage and the center edge angle were not well addressed in the current studies. Further studies of inter- and intra-observer reliability and correlation to gold standard assessments of femoral head coverage such as MRI and 3D CT imaging studies are required for these radiographic measures of hip subluxation.

In conclusion this body of published evidence must be regarded as preliminary at best. Its primary importance to date is in establishing the limited evidence in the literature to either refute or support soft tissue adductor surgery as an efficacious intervention in the prevention of spastic hip dislocation. Current studies have suggested that young children with mild hip subluxation may benefit from adductor release to prevent further hip subluxation. However, these results may merely reflect a more favorable natural history in this group of patients compared to those patients with a severely subluxated hip.

Table I: Dimensions of disability

Dimension	*Description*
Pathophysiology	Interruption or interference of normal physiology and developmental processes or structures
Impairment	Loss or abnormality of body structure or function
Functional Limitation/Activity	Restriction of ability to perform activities
Disability/Participation	Restricted participation in typical societal roles
Societal Limitations/Context factors	Barriers to full participation imposed by societal attitudes, architectural barriers, social policies and other external factors

Table II: AACPDM Levels of evidence. This designation is in two parts, indicating the highest level of evidence the research design provided (Level I-V, Part A) plus an evaluation of the conduct of the actual study (Strong, Moderate, Weak control of threats to internal validity, Part B).

Part A: Type of Research Design

Level	***Non-empirical***	***Group Research***	***Outcomes Research***	***Single Subject Research***
I		Randomized controlled trial. All or none case series		N-of-1 randomized controlled trial
II		Nonrandomized controlled trial. Prospective cohort study with concurrent control group	Analytic research	ABABA design Alternating treatments Multiple baseline across subjects
III		Case control study Cohort study with historical control group		ABA design
IV		Before and after case series without control group		AB design
V	Descriptive case series/case reports Anecdote Expert opinion Theory based on physiology, bench, or animal research Common sense/first principles			

Part B. Conduct of Study

Conduct of the study is judged as Strong ('yes' score of 6 or 7), Moderate (score 5), or Weak (score $\leq$4)

1. Were inclusion and exclusion criteria of the study population well described and followed?
2. Was the intervention well described and was there adherence to the intervention assignment? (for 2-group designs, was the control exposure also well described?)
3. Were the measures used clearly described, valid and reliable for measuring the outcomes of interest?
4. Was the outcome assessor unaware of the intervention status of the participants (i.e. ws there blind assessment)?
5. Did the authors conduct and report appropriate statistical evaluation including power calculations?
6. Were dropout/loss to follow-up reported and less than 20%? For 2-group designs, was dropout balanced?
7. Considering the potential within the study design, were appropriate methods for controlling confounding variables and limiting potential biases used?

Table III: Summary of studies – interventions and participants

Author	Procedure	Additional Interventions	Post Op Bracing	Controls	Population	Preop hip status	Total (n)	Age at surgery (range)	Length of Follow-up
1967 Samilson[48]	•AR & extra-pelvic obt. neurectomy 142 hips •AR 33 hips Extra-pelvic obt. neurectomy only 2 hips Intra-pelvic obt. neurectomy 18 hips	Hamstring lengthening or Eggers transfer 64 hips Iliopsoas lengthening or section 22 hips Derotation femoral osteotomy 14 hips Miscellaneous 19 hips	Long leg casts with abduction bar 8 weeks (54 pts) Hip spica (34 pts) Unknown (10 pts) Traction (4 pts)	None	SQ 79 pts Mixed quad 20 pts Spastic paraplegia 3 pts Hemiplegia 3 pts	Located hips 116 Subluxated hips 47 Dislocated hips 41 Unknown 6	105 pts (210 hips)	13.5 yrs (8 mo-52 yrs)	5.1 yrs (4 mo-40 yrs)
1972 Samilson[7]	•AR+ON 112 pts		Not defined	None	Cerebral palsy	Not defined for this sub-group	112 pts (no of hips not defined)	Not defined	Not defined
1975 Sharrard[30]	Control (Gp I): 72 hips Surgery (Gp II): 134 hips •AR 85 hips •AR+ON 46 hips •AR+IP 3 hips		Plaster immobilisation x 4 weeks	SQ or mixed quad 26 pts SD 10 pts	SQ or mixed quad 51pts SD 16 pts Hemiplegia 2 pts	GP I: Not defined Gp II: Located hips 28 Dysplastic hips26 Subluxated hips57 Dislocated hips 4	Gp I: 36 pts (72 hips) Gp II: 69 pts (134 hips)	GP II: Av.age 4.5 yrs (1.2-12.7 yrs)	GP I: Av. age at flup 7.5 yrs (range 4 -I 6yrs) Gp II: Av. age at flup 8.5 yrs (range 3.5-18yrs)
1980 Reimers[9]	∂AR+/-ON+/-PT 72 hips	Hamstrings lengthening 24 pts Abductor release 4 pts Younts operation 3 pts Rectus elongation 5 pts Tendo Achilles lengthening 26 pts	Plaster cast in abduction 3 wks followed by abduction splinting	None	Cerebral palsy 36 pts	Median maximal MP 44% (range 15-100%)	36 pts (72 hips)	Not defined	6.7 yrs (1.3 to 18.8 yrs0
1981 Root [37]	∂AR 35 pts ∂AR+ON 17 pts	Hip flexor release 10 pts Hamstring release 19 pts Heel cord lengthening 14 pts Open hip reduction 2 pts Tendon transfer 4 pts	Hip spica 3 weeks	None	SQ 21 pts SD 3 pts Paraplegia 18 pts Athetoid 4 pts Athetoid/spastic 4 pts	Not specified	52 pts (102 hips)	2-5 yrs (6 pts) 6-10 yrs (24 pts) 11-15 yrs (12 pts) 15 yrs (12 pts)	5.4 yrs (1-10 yrs)

		Femoral osteotomy 8 pts			Hemiplegia 2 pts				
1984 Manjarris[62]	AR+PT 2 hips		Spica cast 3 weeks		SQ 1	Normal	1 pt (2 hips)	3 yrs	2 yrs
1984 Reimers[38]	∂AR 12 hips ∂AR+ON 14 hips ∂AR+PT 3 hips		Not discussed	None	SQ 11 pts SD 8 pts	Av MP 32% (range 9 to 40%)	19 pts (29 hips)	Av. 5 yrs 11 mo (2-18 yrs)	Av. 1 yr 8 mo (7 mo – 2 yrs 9 mo)
1984 Schulz[58]	❷AR+ON 22 hips	None	Not discussed	None	SQ 7 pts SD 4 pts	Av. MP 28% +/- 4%	11 pts (22 hips)	Av.5 yrs (2-12 yrs)	Av.5.3 yrs (1-16 yrs)
1984 Wheeler[10]	∂AR+ON 24 pts (16 bilat, 8 unilat) Intra-pelvic obturator neurectomy (1 pt)		Double hip spica in flexion and abduction for 6 weeks Abduction brace for night-time "many years"	None	All pts had cerebral palsy	Subluxated 29 hips Dislocated 3 hips Normal 9 hips	25 pts (41 hips)	Av. 5 yrs 9 mo (1-9 yrs)	Av. 3.7 yrs (2-10 yrs)
1985 Kalen[60]	Gp I: 20pts (•AR 16 hips, •AR+ON 23 hips) Gp II: 36 pts (•AR+IP 58 hips)	None	None specified	None	All pts had cerebral palsy	"At risk and subluxed hips"	75 pts (97 hips)	Gp I: Av. 5.2 yrs (2 – 17 yrs) Gp. II: Av. 4.8 yrs (1.5-12.8 yrs)	Gp I: Av 4.4yr (2-9.7 yrs) Gp II Av. 4.5yrs (1.5 - 12.8 yrs)
1985 Silver[55]	Gp. I: Bilat ∂AR+ON 39 pts Gp. II: Unilat ∂AR+ON 11 pts	Prox. Hamstring 8% Psoas tenotomy 26% Psoas & prox. hamstring 26%	Abduction casts x3 weeks then no splintage	None	Non-ambulatory with cerebral palsy 50pts	Gp. I: MP<50% 30 hips MP>50% 46 hips Gp. II not defined	50 pts (87 hips)	Not stated	45 mo (12-92 mo)
1986 Houkom[11]	∂AR+ON+PT 33 pts ∂AR+ON+PT+medial capsular release 2 pts ∂AR+ON 19 pts		Immobilisation in long leg cast with bar or hip spica 2-6 weeks. Some pts used abduction splint at night	None	ALL SQ OR SD - 6 additional athetosis - 3 additional rigidity	CE 0-10° 30 pts CE<0° 27 pts	57 pts (no of hips not defined)	Av. 4 yrs 4 mo (1-12 yrs)	2 – 8 yrs

	•Percut. AR 3 pts		long tem						
1987 Carr[34]	•AR 2 pts •AR+IP 7 pts •AR+IR+ON 3 pts •AR+ON 2 pts	None	"Various"	None	SQ 12 pts SD 1 pt Mixed quad 1 pt	Not defined	14 pts (14 hips)	9.3 yrs (1-15)	Minimum 1 yr Not defined further for this group
1989 Feldkamp[44]	•AR 12 pts Adductor transfer 28 pts	Medial hamstring lengthening 24 pts PT 20 pts	Not defined	None	All pts have cerebral palsy	28 hips CE<20^0 38 hips CE>20^0	40 pts (66 hips)	Not defined	Not defined
1991 Onimus[61]	∂AR+ON+IP (40 hips)	None	Abduction hip spica 2-3 weeks, then at night 2 weeks	None	SQ 24 pts	MP <30% 12 hips MP 30-49% 10 hips MP > 50% 18 hips	24 pts (40 hips)	Av.4 yrs (1.6 – 10 yrs)	3 yrs (1-7 yrs)
1991 Vizkelety[50]	∂AR 497 pts ∂AR+ON 33 pts	None	Abduction plaster 4 weeks	None	All had cerebral palsy	Hips without subluxation 35% Hips with subluxation 65%	530 pts (no of hips not stated)	Av 4.3 yrs	Av 9.2 yrs
1992 Lee[66]	•AR+PT+Hamstring release 1 pt. ∂AR+IP+ON+Hamstring release 1pt	None	None	None	SQ 2 pts	MP>80% I pt Other pt, hip position not defined	2 pts (4 hips)	10 and 6 yrs respectively	18 months and 3 months respectively
1992/93 Khalil[51]	∂AR (35 hips) ∂AR & "other procedures" (45 hips)		Long leg casts with abduction bar 4 weeks	None	SQ 17 Paraplegia 16 SD 8 Hemiplegia 2	Not defined	43 pts (80 hips)	5.8 yrs (2-18 yrs)	10 yrs (6-20 yrs)

1993 Bagg[49]	Gp. I Historical control, no Rx, 15 pts (18 hips) Gp. II: ❷AR, some percutaneous, 14 pts (21 hips)		Not specified	All had cerebral palsy	All had cerebral palsy	Gp. I: -MP<50% 9 -MP>50% 9 Gp. II: -MP<50% 9 -MP>50% 12	Gp. I: 15 pts (18 hips) Gp. II: 14 pts (21 hips)	Gp. II: Av. 10 yrs (range not specified)	Both groups Av. 19 yrs (8 – 30 yrs)
1994 Cobeljic[54]	Gp I: •AR (20 hips) Gp II: ❷AR+IP+ iliac crest resection, tenotomy RF, & elongation or transposition knee flexors (22 hips)	None	Not specified	None	All pts had cerebral palsy	Not specified	25 pts (42 hips)	Gp I: Av Age 4.5 yrs Gp II: Av age 7 yrs (3-13yrs)	Gp I: Av. 6.5 yrs Gp II: 5 yrs (1-12 yrs)
1995 Moreau[47]	❶AR 1pt (2 hips) ∂AR+PT 10 pt (20 hips) ❶AR+PT+ON 5 pts (10 hips) ∂AR+IP 6 pts (12 hips)	Bilat distal hamstring lengthening 1 pt	Long leg casts in abduction 7-14 days Night splinting with abduction brace 3 months	None	SQ 10 SD 12	20 hips normal 24 hips sublux 0 hips dislocated	22 pts (44 hips)	Av age 4 yrs (2 to 6 yrs)	Minimum 5 yrs
1997 Cornell[52]	❶AR+ON (25 hips) •Percutaneous AR (31 hips)	Nil	Long leg casts with abduction bar at 60° for 2 weeks. Then night abduction splint 6-8 hours out of 24 hours	None	SQ 24 pts, SD 5 pts Dystonic quad 5 pts Mixed quad 3 pts	MP<40% 30 hips MP>40 & <60% 14 hips MP>60% 12 hips Dislocated 8 hips	37 pts (56 hips)	Av. 5.6 yrs (2.1-15 yrs)	Av. 5.3 yrs (1.4-17yrs)
1997 Miller[45]	∂AR+IP 32 pts(72 hips) ∂AR+IP+ON 42 pts (75 hips)	Prox. Hamstring release (58 pts. Partial hamstring release 7 pts	Knee immobilised one month	None	SQ 57 PTS SD 16 PTS ST 2 pts	<25% = 29, >25%-39%=77, 40%-59%=32, >60%=9	74 pts (147 hips)	Av. 4.5 yrs (1.8 – 8 yrs)	39 mo (8-69 mos)
1997 Spruitt[53]	❷AR+IP 9 pts (13 hips) •AR+IP+ON 3pts (4 hips)	None	Not specified	None	SQ 9 SD 3	2 hips dislocated 7 hips subluxated 8 hips normal	12 pts (17 hips)	2 – 13.5 yrs (6 yrs)	Av. 4.05 yrs (2.5 – 8 yrs)

1998 Cottalorda[46]	❶ AR (31 hips) ❶ AR+ON (23 hips) ❷ Percutaneous AR (3 hips)	Nil	Spica cast 3 weeks Abduction bracing (night time) 3 mos	None	SQ 30	MP <20%=12 hips MP 20 – 40% 25 hips MP >40% 20 hips	30 pts (57 hips)	6 yr 11 mo (2.5-13)	Av.6 yr (2-20 yr)
1999 Ushmann[63]	•Unilat. AR (1 hip)	Bilateral hamstring lengthening 1 pt	Hip spica cast 7 wks. Hip abduction orthosis long term	None	SQ 1 pt	Hip adducted but located -1 hip	1pt (1 hip)	15 yrs	8 mo
2000 Turker[56]	❶AR 32 pts (64 hips) ∂AR+ON 13 pts(26 hips)	PT and distal hamstring releases 50% of pts	Not specified	None	SQ 45	Not stated	45 pts (90 hips)	Av age not defined (range 1.5-11 yrs)	Av. 8.1 yrs (Minimum 4 yrs)

Legends
Pts = patients
AR =adductor release
CE = center-edge angle
MP = migration percentage
RF = rectus femoris
ON =anterior branch obturator neurectomy
IP = intramuscular lengthening psoas
PT = psoas tenotomy at lesser trochanter
SQ = spastic quadriplegia
SD = spastic diplegia
ST = spastic triplegia
❶ = adductor release consisting of tenotomy adductor longus, gracilis and partial lengthening adductor brevis
❷ = adductor release, not otherwise defined

Table IV Summary of studies – research methods

Study	Research Design	Level of evidence and quality	Rx (n)	Control Rx (n)
1967 Samlinson[48]	Before and after case series	IV-W	105	None
1972 Samlinson[7]	Anecdotal case series	V	112	None
1975 Sharrard[30]	Cohort study with historical controls	III – W	69	36
1980 Reimers[9]	Before and after case series	IV-W	36	None
1981 Root [37]	Before and after case series	IV – W	52	None
1984 Manjarris[62]	Case report (n of 1)	V	1	None
1984 Wheeler[10]	Before and after case series	IV – W	25	None
1984 Reimers[38]	Descriptive case series	V – W	19	None
1984 Schulz [58]	Before and after case series	IV – W	11	None
1985 Kalen [60]	Before and after case series	IV – W	56	None
1984 Wheeler [10]	Before and after case series	IV - W	25	None
1985 Silver [55]	Before and after case series	IV – W	50	None
1986 Houkom [11]	Before and after case series	IV – W	57	None
1987 Carr [34]	Before and after case series	IV - W	14	None
1989 Feldkamp [44]	Before and after case series	IV - W	40	None
1991 Vizkelety [50]	Anecdotal case series	V	530	None
1991 Onimus [61]	Before and after case series	IV - W	24	None
1992-93 Khalil[51]	Before and after case series	IV - W	43	None
1992 Lee [66]	Descriptive case report	V	2	None
1993 Bagg [49]	Cohort study with historical controls	III – W	45	15
1994 Cobeljic [54]	Before and after case series	IV – W	25	None
1995 Moreau [47]	Before and after case series	IV – W	22`	None

1997 Miller [45]	Before and after case series No controls	IV – W	74	None
1997 Spruit [53]	Before and after case series No controls	IV – W	12	None
1997 Cornell [52]	Before and after case series No controls	IV – W	37	None
1998 Cottalorda [46]	Before and after case series No controls	IV – W	30	None
2000 Turker[56]	Before and after case series No controls	IV – W	45	None

Table V: Part A. Average of group comparison results. These results reflect comparisons, either of the average pre adductor surgery status of a group with its average post adductor surgery status, or comparison of the post adductor surgery results with control subjects (Sharrard et al., 1975; Bagg et al.,.1993)

Study	Outcome of Interest	Dim. of Disability	Measure	Results	Is change of clinical Importance ?	Stats	Level of Evidence
1967 Samilson[48]	Unilateral hip releases						
	Hip abduction deformity	I	Clinical observation	-	Yes		IV + W
	Subluxation /dislocation non-operated hip	I	Clinical observation	-	Yes		IV + W
1975 Sharrard[30]	No. of hips subluxated or dislocated at follow-up	I	Radiographic observation	+	Yes	-	III + W
	Hip flexion contracture	I	Goniometric measure	+	Yes		III + W
	Range of hip abduction	I	Goniometric measure	+	Yes		III +W
	Pain in hip	I	Anecdotal	+	Yes		V
1972 Samilson[7]	No of hips subluxated or dislocated at follow-up	I	Anecdotal	-	Yes		V
1980 Reimers[9]	Unilateral adductor releases						
	Migration percentage∂ (ipsilateral hip)	I	Goniometric measure on hip radiograph	+	Yes	P<0.001	IV + W
	Migration percentage (contralateral hip)	I	Goniometric measure on hip radiograph	U		P>0.1	IV + W
1981 Root[37]	Functional ability	FL/A	Minear scale•	U			V
	Sitting balance	FL/A	Anecdotal	+	Yes		V
	Standing balance	FL/A	Anecdotal	+	Yes		V
	Range of hip abduction	I	Goniometric measure	+	Yes		IV + W
	Hip flexion contracture	I	Goniometric measure	U			IV + W
	Scissor gait pattern	FL/A	Anecdotal	+	Small		V
	Reduction in brace use	FL/A	Anecdotal	+	Small		V
	Gait endurance	FL/A	Anecdotal	+	Small		V
1984 Wheeler[10]	Center-edge angle÷	I	Goniometric measure of hip radiograph	+	Yes		IV + W
	Acetabular index≠	I	Goniometric measure of hip radiograph	+	Small		IV + W
	Neck Shaft angle≡	I	Goniometric measure of hip radiograph	U			IV + W
	Range of hip abduction	I	Goniometric measure	+	Yes		IV + W
	Ability to ambulate	FL/A	Anecdotal	+	Small		V
1989 Feldkamp[44]	Motor function	FLA	Clinical observation	+	Small	NS	IV+W
	Passive hip extension	I	Thomas test	+	Small	NS	IV+W
	Hip flexion (standing)	FL/A	Photographic record	+	Small		IV+W
	Internal hip rotation (standing posture)	FL/A	Photographic record	+	No		IV+W
	Hip abduction	I	Goniometric measure on radiograph	+	Yes		IV+W
	Adduction asymmetry	I	Goniometric measure on radiograph		Small		IV+W
	Center-edge angle	I	Goniometric measure	+	Yes		IV+W

			on radiograph			
1991 Vizkelety[50]	No. of hips subluxated or dislocated on radiograph		Radiographic observation	+	Yes	V
1993 Bagg[49]	No. of hips dislocated at follow-up	I	Clinical and radiographic observation	U		III + W
	No. of hips with degenerative disease	I	Radiographic observation	U		III + W
	Migration percentage	I	Goniometric measure on hip radiograph	U		III + W
	Pain in hip at follow-up	I	Pain score 0-3	U		III + W
1995 Moreau[47]	Centre edge angle	I	Goniometric measure on radiograph	+	Yes	IV + W
1997 Miller[45]	Acetabular index	I	Goniometric measure on hip radiograph	+	Yes	IV + W
	Neck shaft angle	I	Goniometric measure on hip radiograph	U		IV + W
	Migration percentage	I	Goniometric measure on hip radiograph	+	Yes	IV + W
	Hip abduction	I	Goniometric measure	+	Yes	IV+W
	Hip internal rotation	I	Goniometric measure	U		IV+W
	Hip external rotation	I	Goniometric measure	U		IV+W
	Popliteal angle	I	Goniometric measure	+	Small	IV+W
1998 Cottalorda[46]	Pelvic obliquity	I	Clinical observation	-	Yes	IV + W
	Scoliosis	I	Radiographic observation	-	Yes	IV + W
	Range of hip abduction	I	Goniometric measure	+	Yes	IV + W

❶ Migration percentage described by Reimers (9)
❷ Minear scale of functional ability
❸ Center edge angle described by Massie and Howarth (57)
≠ Acetabular index
≡ Neck shaft angle
+ Overall group results better postoperatively
- Overall group results worse postoperatively
U Overall group results unchanged

Table V Part B: The uniformity of effect within a treatment group.

These results reflect how many of the group were improved with adductor surgery, how many were worse off after the surgery and how many were unchanged compared to the pre-operative status

Study	Dim. disability	Outcome	Measure	Improved result	Worse result or failure	Unchanged	Stats	Level of evidence
1967 Samilson[48]	I	Change from pre-op hip status	Radiographic and clinical observation	38/204 hips∂	19/204 hips∂	147/204 hips∂		IV + W
	FL/A	Sitting balance	Anecdotal	3/4 pts•	NK	NK		V
	FL/A	Gait improvement	Anecdotal	10/30 pts•	NK	NK		V
	SL/CF	Perineal hygiene	Anecdotal	46/69 pts•	NK	NK		V
1975 Sharrard[30]	I	No of hips subluxated or dislocated at followup	Radiographic observation	67/115 hips÷	2/115 hips÷	46/115 hips÷		III + W
1980 Reimers[9]	I	Migration percentage	Comparison of goniometric measures of primary and postop MP on hip radiographs	35/72 hips	26/72 hips	11/72 hips	P>0.01	IV + W
	I	Migration percentage	Comparison of goniometric measures of maximal MP and postop MP	51/72 hips	0/72 hips	21/hips	P<0.001	IV + W
	I	Hips requiring further bony surgery	Clinical observation		11/72 hips			IV + W
1981 Root[37]	I	Neck shaft angle	Goniometric measure of hip radiograph	4/30 hips≠	4/30 hips≠	22/30 hips≠		IV+W
	I	Centre edge angle	Goniometric measure of hip radiograph	4/30 hips ≠	4/30 hips≠	22/30 hips≠		IV+W
	I	Acetabular index	Goniometric measure of hip radiograph	6/30 hips≠	2/30 hips ≠	22/30 hips≠		IV+W
1984 Reimers[38]	I	Migration percentage	Goniometric measure of hip radiograph	17/29 hips	5/29 hips	7/29 hips	P<0.001	IV+W
1984 Schulz[58]	I	Migration percentage	Goniometric measure of hip radiograph	15/22 hips	5/22 hips	2/22 hips	NS	IV+W
	I	Centre edge angle	Goniometric measure of hip radiograph	18/22 hips	2/22 hips	2/22 hips	NS	IV+W
	I	Migration percentage index	Goniometric measure of hip radiograph	19/22 hips	0/22 hips	3/22 hips	P<0.01	IV+W
	I	Centre edge angle index	Goniometric measure of hip radiograph	20/22 hips	2/22 hips	0/22 hips	P<0.01	IV+W
1984 Wheeler[10]	I	No. of hips improved by radiographic assessment of subluxation and dislocation	Radiographic observation	21/41 hips	3/41 hips	17/41 hips		IV+W
1985 Kalen[60]		Gp I: AR						
	I	- No of hips requiring further surgery	Clinical observation		64% hips			IV + W
		Gp. II: AR+IP						IV + W
	I	- No of hips requiring	Clinical observation		28% hips			IV + W

		further surgery					
	I	- Migration percentage		15/40 hips≡	8/40 hips≡	17/40 hips≡	IV + W
1985 Silver[55]		<u>Bilateral releases</u>					
	I	Migration percentage	Goniometric measure of hip radiograph	48/76 hips	28/76 hips	0/76 hips	IV + W
	I	Hips dislocated at follow-up or further surgery required	Clinical and radiographic observation		15/76 hips		IV + W
		<u>Unilateral releases</u>					
	I	Migration percentage	Goniometric measure of hip radiograph	10/11 operated hips 1/11 non-operated hips	1/11 operated hips 9/11 non-operated hips	0/11 hips 1/11 non-operated hips	IV + W
	I	Hips dislocated at follow-up or further surgery required	Clinical and radiographic observation		1/11 operated hips 5/11 non-operated hips		IV + W
1986 Houkom[11]	I	Centre edge angle	Goniometric measure of hip radiograph	31/57 pts	19/57 pts	7/57 pts	IV+W
	FL/A	Ability to ambulate	Anecdotal	NK	1/57 pts	NK	IV+W
1987 Carr[34]	I	Unilateral releases					
	I	Migration percentage contra-lateral hip	Goniometric measures of hip	1/14 non operated hips≈	7/14 non operated hips≈	6/14non operated hips≈	IV + W
	I	Windswept deformity at pelvis	Clinical observation	NK	7/14 pts	NK	IV + W
1991 Onimus[61]	I	Migration percentage	Goniometric measure on hip radiograph	26/40 hips	14/40 hips	-	IV + W
1992-93 Khalil[51]	I	Hip position on radiograph	Radiographic observation	12/30…	1/30…	17/30…	IV + W
	FL/A	Ability to ambulate	Anecdotal	26/43 pts	1/43 pts	16/43 pts	V
1994 Cobeljic[54]	I	"Clinical and radiologic findings"	Three point scale derived from clinical and radiographic findings		20/20 hips		V
1995 Moreau[47]	I	Hips that are subluxated or dislocated at follow-up	Clinical and observations	19/44 hips	0/44 hips	25/44 hips	IV + W
	FL/A	Ability to ambulate	Parental questionnaire	6/11 pts		5/11 pts	IV + W
	FL/A	Ability to stand	Parental questionnaire	14/18 pts		4/18 pts	IV + W
	FL/A	Ability to ambulate	Hoffer classification of ambulation	12/22 pts	1/22 pts	9/22 pts	IV + W
	FL/A	Ability to sit	Clinical observation	1/22 pts		21/22 pts	IV + W
	FL/A	Ability to sit	Parental questionnaire	14/18 pts	N/A	4/18 pts	IV +W
	I	Hip flexion contracture	Goniometric measure	29/43 hips	10/43 hips	5/43 hips	IV + W
	I	Range of hip abduction	Goniometric measure	31/43 hips	7/43 hips	5/43 hips	IV + W

	SL/CF	Ability to diaper	Parental questionnaire	8/13 pts	0/13 pts	5/13 pts		
1997 Cornell[52]	I	Migration percentage	Goniometric measure on hip radiograph	27/59 hips	12/59 hips	20/59 hips		IV + W
	I	No. hips reduced, subluxated or dislocated⎸		7/59	8/59	44/59 hips		IV + W
1997 Miller[45]	I	No of pts requiring further surgery for subluxation	Clinical observation	N/A	8/74 pts	N/A		IV + W
1997 Spruit[53]	I	Centre edge angle	Goniometric measure of hip radiograph	10/17 hips	6/17 hips	1/17 hips	P=0.86	IV + W
	I	Acetabular index	Goniometric measure of hip radiograph	11/17 hips	3/17 hips	3/17 hips	P=0.01	IV + W
	I	Femoral head coverage	Goniometric measure of hip radiograph	10/17 hips	5/17 hips	2/17 hips	P=0.38	IV + W
	I	No. hips subluxated or dislocated	Clinical and radiographic observation	4/7 hips	0/17 hips	13/17		IV + W
	FL/A	Ability to sit	Anecdotal	3/12 pts	NK	NK		V
	FL/A	Ability to ambulate	Anecdotal	3/12 pts	NK	NK		V
1998 Cottalorda[46]	I	Migration percentage	Goniometric measure of hip radiograph	7/57 hips	20/57 hips	30/57 hips		IV + W
	I	No of patients needing further hip surgery		N/A	13/30 pts	N/A		IV + W
2000 Turker[56]	I	No of hips requiring further surgery or dislocated at followup		N/A-	31/90 hips	N/A		IV + W

∂ Numbers reported do not add up to 210 hips but instead 204 hips with 6 hips having an unknown outcome
• Selected subgroups of pts with different indications for sugery
÷ Only 115 of original 134 hip radiographs available for review
≠ 30 hips of original 102. All others excluded due to osteotomy at the time of or subsequent to add. Release
≡ Only 40 hip radiographs available, other 18 not available
≈ Improved >10% ↓MP, unchanged <10% change MP, worse >10%
… Pre-op radiograph only available for 30 out of 80 hips
⎸ Reduced MP<33%, subluxated MP 33-99%, dislocated MP 100%
N/A Not applicable
NK Not known

Table VI: Evidence table – Outcomes of Adductor Surgery for Cerebral Palsy

Part A: Average-of-group comparison results. These results reflect comparisons of the average pre-op status of a group with its post surgery status. Each outcome is indicated by a superscript that is the citation number of each study that produced this result associated with a level of evidence (coded I-V for type of research design and S, M, W for strong , moderate or weak controls to threats of validity in conducting the study). Each entry reflects whether the group-average outcome was better after adductor surgery compared to the pre-adductor surgery group

Outcomes classified by Dimensions. of Disability	Improved Results (Statistically Significant)	Improved Results (Not Statistically Evaluated)	Results worse at Follow-up	Results Unchanged or Not Significant
Pathophysiology				
Impairment				
Migration percentage	IV+W [9]	IV + W [45]		III+W [49]
Centre edge angle		IV+W [10,44,47]		
Acetabular Index		IV+W [10,45]		
Neck shaft angle				IV+W [10,45]
No. of hips subluxated or dislocated at follow-up		III+W [30] V [50]	V [7] IV + W [48]	III+W [49]
Range of hip abduction		III+W [30]IV+W [10,37,44-46]		
Hip flexion contracture		III+W [30], IV+W [37]		
Passive hip extension				IV+W [44]
Hip flexion (standing)		IV+W [44]		
Hip internal rotation		IV+W[44]		IV+W [45]
Hip external rotation				IV+W[45]
Popliteal angle		IV+W[45]		
Scoliosis			IV+W[46]	
Pelvic obliquity/asymmetry		IV+W [44]	IV+W[46]	
Degenerative joint disease				III+W [49]
Pain in hip		V [30]		III+W [49]
Functional Limitation/Activity				
Reduction in brace use		V[37]		
Scissor gait pattern		V [37]		
Functional ability		IV+W [44]		V[37]
Gait endurance		V [37]		
Ability to sit		IV+W [47] V+ W [53]		
Ability to stand		IV+W [47], V [37]		
Ability to ambulate		V[10]		
Disability/Participation				
Societal limitations/context factors				

Table VI: Evidence table – Outcomes of Adductor Surgery for Cerebral Palsy

Part B: Uniformity of results within a treated group. These results show how many participants improved, how many fared better before the adductor surgery, and/or how many were unchanged The number of participants within a treated group is followed by a level of evidence and the citation for the study that produced the result. Statistically significant results indicated by *.

Outcomes by Dimensions of Disability	**Improved Results**	**Results Worse at Follow-up**	**Results Unchanged or Not Significant**
Pathophysiology			
Impairment			
Migration percentage	17/29 hips IV+W (38)*	5/29 hips IV+W (38) *	7/29 hips IV+W (38) *
	35/72 hips∂ IV+W (9)	26/72 hips∂ IV+W (9)	11/72 hips∂ IV+W (9)
	51/72 hips• IV+W (9)*	0/72hips• IV+W (9)*	21/72 hips• IV+W (9)*
	15/22 hips IV+W (58)	5/22 hips IV+W (58)	2/22 hips IV+W (58)
	15/40 hips IV+W (60)	8/40 hips IV+W((60)	17/40 hips IV+W(60)
	48/76 hips IV+W(55)	28/76 hips IV+W (55)	0/76 hip IV+W (55)
	10/11 hips÷ IV+W (55)	1/11 hips÷ IV+W (55)	0/11 hips÷ IV+W (55)
	1/11 hips≠ IV+W (55)	9/11 hips≠ IV+W (55)	1/11 hips≠ IV+W (55)
	1/14 hips≠ IV+W (34)	7/14 hips≠ IV+W (34)	6/14 hips≠ IV+W (34)
	26/40 hips IV+W (61)	14/40 hips IV+W (61)	
	27/59 hips IV+W (52)	12/59 hips IV+W (52)	20/59 hips IV+W(52)
	7/57 hips IV+W (46)	20/57 hips IV+W (46)	30/57 hips IV+W (46)
Centre edge angle	4/30 hips IV+W (37)	4/30 hips IV+W (37)	22/30 hips IV+W (37)
	18/22 hips IV+W (58)	2/22 hips IV+W (58)	2/22 hips IV+W (58)
	31/57pts IV+W (11)	19/57 pts IV+W (11)	7/57pts IV+W (11)
	10/17 hips IV+W (53)	6/17 hips IV+W (53)	1/17 hips IV+W (53)
Acetabular Index	6/30 hips IV+W (37)	2/30 hips IV+W (37)	22/30 hips IV+W (37)
	11/17 hips IV+W* (53)	3/17 hips IV+W* (53)	3/17 hips IV+W* (53)
Neck shaft angle	4/30 hips IV+W (37)	4/30 hips IV+W (37)	22/30 hips IV+W (37)
Migration percentage index	19/22 hips IV+W * (58)	0/22 hips IV+W * (58)	3/22 hips IV+W* (58)
Center edge angle index	20/22 hips IV+W* (58)	2/22 hips IV+W* (58)	0/22 hips IV+W* (58)
Femoral head coverage	10/17 hips IV+W (53)	5/17 hips IV+W (53)	2/17 hips IV+W (53)
Hip status on radiograph(eg reduced, subluxated or dislocated at follow-up)	38/204 hips IV+W (48)	19/204 hips IV+W (48)	147/204 hips IV+W (48)
	67/115 hips III+W (30)	2/115 hips III+W (30)	46/115 hips III+W (30)
	21/41 hips IV+W (10)	3/41 hips IV+W (10)	17/41 hips IV+W (10)
	12/30 hips IV+W (51)	1/30 hips IV+W (51)	17/30 hips IV+W (51)
	19/44 hips IV+W (47)	0/44 hips IV+W (47)	25/44 hips IV+W (47)
	0/20 hips V (54)	20/20 hips V (54)	0/20 hips V (54)
	7/59 hips IV+W (52)	8/59 hips IV+W (52)	44/59 hips IV+W (52)
	4/17 hips IV+W (53)	0/17 hips IV+W (53)	13/17 hips IV+W (53)
No of hips/pts requiring further		11/72 hips IV+W (9)	

surgery			
		15/76 hips IV+W [55] 1/11 hips ÷ IV+W [55] 5/11 hips ≠ IV+W[55] 8/74 hips IV+W [45] 13/30 pts IV+W [46] 31/90 hips IV+W [56]	
Range of hip abduction	31/43 hips IV+W [47]	7/43 hips IV+W [47]	5/43 hips IV+W [47]
Hip flexion contracture	29/43 hips IV+W [47]	10/43 hips IV+W [47]	5/43 hips IV+W [47]
Pelvic obliquity/asymmetry	NK	7/14 pts÷ IV+W [34]	NK
Functional Limitation/Activity			
Gait improvement	10/30 pts V [48]	NK	NK
Ability to sit	3/4 pts V [48] 1/22 pts≡ IV+W [47] 14/18 pts≡ IV+W [47] 3/12 pts V [53]	NK 0/22 pts≡ IV+W [47] 0/18 pts≡ IV+W [47] NK	NK 21/22 pts≡ IV+W [47] 4/18 pts ≡ IV+W [47] NK
Ability to stand	14/18 pts IV+W [47]	0/18 pts IV+W [47]	4/18 pts IV+W [47]
Ability to ambulate	3/12 pts V [53] 26/43 pts V [51] NK 6/11 pts≡ IV+W [47] 12/22 pts≡ IV+W [47]	NK 1/43 pts V [51] 1/57 pts V [55] 0/11 pts≡IV+W [47] 1/22 pts≡ IV+W [47]	NK 16/43 pts V [51] NK 5/11 pts≡ IV+W [47] 9/22 pts≡ IV+W [47]
Disability/Participation			
Societal limitations/context factors			
Care giving: diapering	8/13 pts IV+W [47] 46/69 pts V [48]	0/13 pts IV+W [47] NK	5/13 pts IV+W [47] NK

¶ Primary migration percentage compared to postop migration percentage

• Maximal preop migration percentage compared to postop migration percentage

÷ Pre and post-op comparison for unilateral surgeries, ipsilateral hip

≠ Pre and post-op comparison for unilateral surgeries, contralateral hip

≡ Measured two different ways with different outcome measures

Table VII: Complications and Adverse Effects

Study	Type of Effect	No. of cases
1972 Samlinson[7] N=105	Wound infection / haematoma	14
	Spontaneous hip fusion	3
	Recurrent adduction deformity	39
	Hip abduction contracture	13
	Femoral fracture	2
	Contralateral hip dislocation	3
1975 Sharrard[30] N = 69	Wound haematoma / infection	3
	Hip abduction contracture	1
	Fracture following cast removal	2
	Coxa magna	1
1984 Manjarris[62] N=1	Bilateral avascular necrosis femoral heads	1
1985 Silver[55] N=50	Hip abduction contracture	1
1986 Houkom[11] N=57	Hip abduction contracture	12
	Hip hyperextension contracture	2
1992 Lee[66] N=2	Heterotopic ossification	2
1997 Cornell[52] N=37	Wound infection	1
	Wound haematoma	2
1985 Spruit[53] N=12	Hip abduction contracture	1
1998 Cottalorda[46] N=30	Wound haematoma	3
1999 Ushmann[63] N=1	Spontaneous ankylosis opposite hip	1

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