

# Hip and Spine Surgery is of Questionable Value in Spina Bifida

## An Evidence-based Review

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### Abstract

**Background** Although many children with spina bifida and associated scoliosis or dislocated hips undergo spine or hip surgery, the benefits are uncertain.

**Questions/purposes** The purpose was to perform an evidence-based review on the benefits and risks of surgery for dislocated hips and scoliosis in spina bifida.

**Methods** I performed a Medline® and Embase® search from 1950 to 2009 for Level I to Level III studies investigating the benefits and risks of surgery for scoliosis and hip dislocation in patients with spina bifida. When available, I extracted types of surgery, complication rates, functional outcomes of seating, walking, and overall physical function. All treatment recommendations received a Grade of Recommendation: Grade A (consistent Level I studies); Grade B (consistent Level II and III studies); Grade C (consistent level IV and V studies); or Grade I (insufficient or contradictory studies).

**Results** Combined anterior and posterior surgery had lower rates of nonunion for scoliosis. Although there may be some benefit in seating, overall physical function measured in a different and nonstandardized fashion was not much changed and major complication rates, including nonunion and infections for scoliosis surgery, exceed 50% in several studies. For dislocated hips, the impact on

walking ability appears related to contracture (not dislocation). Surgery for hip dislocation did not improve walking ability. The literature provides no guidance on the best treatment for unilateral dislocation.

**Conclusions** The benefits of scoliosis surgery are uncertain (Grade I). Spine surgery, if performed, should be anterior and posterior (Grade B). An all-pedicle approach for scoliosis surgery may be effective (Level I). Hip reduction surgery did not improve walking (Grade B) but may be appropriate in low-level unilateral dislocation (Level I).

### Introduction

Spina bifida is a complex congenital abnormality of peripheral nerves, the spinal cord, brain, and brain stem. In addition to the typical flaccid paralysis of the lower extremities, these children often have a mixed neurologic pattern, which may include some elements of spasticity, upper extremity dyscoordination, and abnormalities in bulbar function. Anatomically, they also have deficient posterior spinal elements, absence of posterior spinal musculature, and congenital deformities of the spine with acquired deformities and contractures of the lower limb. For the orthopaedic surgeon, the primary focus involves maintaining, restoring, or preventing anticipated future decline in physical function.

The WHO International Classification of Functioning provides a useful framework for evaluating the needs of children with spina bifida [19]. In this framework, spina bifida, the disease, leads to abnormalities in structure or function. These abnormalities in structure or function in turn cause restrictions in activities such as running, walking, or sitting. These activity restrictions interfere with role

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function such as play or attending school. Although many other factors may affect this pathway [28], this framework allows orthopaedic surgeons to understand the progression from disease to role limitations. Patients' concerns usually relate to activity or role limitation. The challenge for the orthopaedic surgeon is to attribute these concerns to impairment in structure or function that can be addressed by surgical treatment and thereby in turn improve children's functional abilities. Although the focus for orthopaedic surgeons is on physical function, the positive effects that improved physical function can have on mental health, social function, and overall physical fitness in children and young adolescents cannot be underestimated.

Two common orthopaedic abnormalities in children with spina bifida are dislocated hips and scoliosis. Hip dislocation occurring at birth or throughout childhood can affect up to 30% of children [8]. Although the pathoetiology is unclear, dislocation is probably related to some combination of abnormal sensation, motor function, and joint development as well as contractures in muscles and soft tissues. Although it may seem intuitive that relocation of the dislocated hip is appropriate, surgery can be complicated by a fracture, infection, pain, and stiffness with a high rate of redislocation. Scoliosis, affecting at least 30% of children [1], also can have onset in young children with the potential to cause pelvic obliquity, spinal imbalance, seating difficulties, and in the long-term pulmonary dysfunction [26]. However, surgery is a major undertaking with high complication rates, including infection, pressure sores, nonunion, failure of instrumentation, and even death. Thus, it is vital to understand the best form of treatment for children with a dislocated hip and scoliosis and spina bifida.

Evidence-based medicine has been defined as the "conscientious, explicit, judicious use of current best evidence in making decisions about the care of individual patients" [22]. The practice of evidence-based medicine means integrating individual expertise with the best available external clinical evidence from systematic research while considering patient preference. Said another way, surgeons use the highest quality evidence and with parents and patients make the best treatment choices. Shared decision-making requires explicit discussion of the best evidence with children and families, including the advantages and disadvantages of different treatment options. The highest level of evidence comes from multiple randomized clinical trials. However, the dilemma for surgeons is that treatment recommendations must often be made in the absence of Level I studies, the effect of hip dislocation on walking ability and physical function.

The quality of research is based substantially on the research design. High-quality research minimizes the introduction of bias, defined as systematic deviation from

the truth. The truth here addresses the question of whether the treatment really and truly benefits the patient. With biased research, conclusions about the benefit, or lack of benefit, of treatment may be incorrect. For example, if patients are substantially dissimilar on important prognostic factors in different treatment groups, then the attribution of benefit of treatment may be the result of the probability of success based on the higher (or lower) likelihood of a good prognosis among patients.

Levels of evidence are a simple and explicit valuation of research founded on basic principles of design [32]. Levels of Evidence are from I to V and are based on each study type. For therapeutic studies, the basic principles of Levels of Evidence are that controlled studies are better than uncontrolled studies, prospective studies are better than retrospective studies, and randomized studies are better than nonrandomized studies. Although a full understanding requires a complete critical appraisal, levels of evidence do provide a rough estimate of study quality. The focus of this article is on therapeutic studies in which Level I evidence is from randomized clinical trials, Level II evidence is from prospective comparative studies, Level III evidence is from retrospective studies, Level IV evidence is from uncontrolled case series, and Level V evidence is from expert opinion [32]. For each treatment recommendation in this article, I have attached a grade of recommendation that summarizes the literature [31]. A Grade A recommendation, based on multiple consistent Level I studies, should definitely guide treatment decisions; Grade B, based on multiple consistent Level II and III studies, should probably guide treatment decisions; Grade C, recommendation based on consistent Level IV and V studies, may guide treatment decisions; and Grade I, based on inconsistent or insufficient evidence, provides no guidance.

In applying the principles of evidence-based medicine, surgeons must complete the five As: assess the clinical situation, ask the clinical question, acquire the appropriate information, appraise the appropriate information, and apply the evidence to the clinical situation [16]. In deciding how to treat an individual patient, evidence can be obtained from multiple sources, including prior training, individual surgical expertise, the opinions of respected colleagues, and the surgical literature. For the purposes of this article, attention was restricted to Level III studies based on the presumption that uncontrolled studies pose too many problems to determine treatment effectiveness.

The purpose of this review was to determine: (1) the effect of hip dislocation on walking ability and physical function; (2) the complication rate including redislocation of hip surgery; (3) the effect of scoliosis surgery on seating, ambulation, and physical function; and (4) the relationship between surgical approach and complications including infections and nonunion.

## Search Strategy and Criteria

A literature review was performed using Medline® and Embase® for the years 1950 to 2009. The search strategy was designed to identify Level I to Level III studies (Table 1). Only if a key issue was identified by a Level IV study were there studies noted, but these studies did not affect treatment recommendations. Inclusion criteria for the study included studies on spina bifida involving the hip or spine, English language, and in humans for children aged 0 to 18 years. The restriction to the English language was made given the difficulties of translation. The abstracts were reviewed by the author to determine eligibility, type of intervention, and clinical findings. Because there were no randomized clinical trials and because of the lack of uniformity among studies in determination of any aspect of outcome, a formal meta-analysis was not possible.

The literature review identified 379 citations. Of the 379 studies, 326 were not eligible because they were not directly relevant to the treatment of hip dislocation or scoliosis. Of the remaining 53 citations addressing treatment for hip dislocation and scoliosis (Table 2), 14 were Level III evidence and formed the basis of this review. None of the studies were randomized and therefore I assessed the elements of nonrandomized controlled studies from the Cochrane Communication Review Group J: Study Quality Guide (<http://www.latrobe.edu.au/cochrane/assets/downloads/StudyQualityGuide050307.pdf>). None of the studies used concealed allocation, blinding, or intention-to-treat analyses. All of the studies had baseline differences in prognostic variables but only Feiwell et al. [14] and Alman et al. [3] attempted to adjust for their differences using

**Table 1.** Search strategy: hip and spine surgery in children with spina bifida

Database: Ovid MEDLINE(R) 1950 to present with daily update

- 1 (Meningomyelocele/or (myelomeningocele or myelocele or myelomeningocele).ti,ab.) and (hip/or spine/or (hip\$ or spin\$).ti,ab.) (1455)
- 2 clinical trials/or clinical trials, phase i/or clinical trials, phase ii/or clinical trials, phase iii/or clinical trials, phase iv/or controlled clinical trials/or randomized controlled trials/or multicenter studies/or cross-over studies/or double-blind method/or meta-analysis/or random allocation/or single-blind method/or systematic review\$.ti,ab. or ((singl\$ or doubl\$ or tripl\$) adj (mask\$ or blind\$)).ti,ab. or (blind\$ or random\$).ti,ab. (761116)
- 3 case-control studies/or retrospective studies/or comparative study/or case series.tw. or exp treatment outcome/or exp Cohort Studies/(2658666)
- 4 2 or 3 (3092586)
- 5 1 and 4 (520)
- 6 limit 5 to (english language and humans) (472)
- 7 limit 6 to “all child (0 to 18 years)” (415)
- 8 remove duplicates with EndNote (378)

**Table 2.** Search strategy: hip and spine surgery in children with spina bifida

Surgical location	Levels of Evidence for two study questions					
	I	II	III	IV	V	Total
Hip	0	0	5	9	0	14
Spine	0	0	9	29	1	39
Total	0	0	14	38	1	53

multivariable techniques. Most studies assessed some elements of physical function but only studies by Alman et al. [3] and Wai et al. [28] used validated measures of physical function. In addition to the Level III studies addressing therapy for hip dislocation, there were 10 Level II prognostic studies that examined the effect of the patient characteristics including hip dislocation on ambulation.

Treatment recommendation received a grade of recommendation. Grade A is consistent Level I studies; Grade B is consistent Level II and III studies; Grade C is consistent Level of IV and V studies; and Grade I is insufficient or contradictory studies.

## Results

The specific studies identified in the search are discussed in Appendix 1. The first purpose was to determine the relationship between dislocation and walking ability and physical function. All studies found neurosegmental level was the primary determinant of walking ability. No study found any relationship between hip position and walking ability. One study reported marginal improvement in gait energetics in a small subset of study patients but no difference in physical function evaluated by Child Health Questionnaire and Rand Health Insurance functional score [3]. This later study was the only study to evaluate overall physical function and found no benefit of hip surgery for the dislocated hip.

The second question related to the complication rate of hip surgery for hip dislocation including redislocation. Although not all studies evaluated the rate of hip redislocation after surgery, the rate of dislocation varies from 30% [3] to 45% [7].

The third question asked what effects scoliosis surgery had on seating, walking ability, and physical function. Only two studies, both cross-sectional, evaluated the physical function of patients with and without surgery for scoliosis. Both of these studies [17, 28] concluded surgery had little beneficial effect on physical function. Mazur, in their uncontrolled before and after study, reported seating was improved in 70% but walking ability was decreased in 67% of patients.

The fourth purpose was to determine the relationship between surgical approach and complications including nonunion and infection. The correction and fusion were higher in patients receiving combined anterior and posterior fusion. The nonunion rate was 46% in the posterior only fusion group [20] compared with 14% to 23% in the combined anterior and posterior group [4, 20, 27]. No study provided comparative data on complications, including infection, comparing different surgical approaches. All pedicle posterior only approaches were described in one study but with insufficient numbers to make any conclusions [21]. No study, however, considered the long-term effect of untreated scoliosis and the potential role for surgery in preventing late decline in pulmonary function.

## Discussion

The role of hip surgery for children with spina bifida and dislocated hips and spine surgery for children with scoliosis and spina bifida is controversial. This controversy originates largely from literature that relies almost exclusively on uncontrolled case series. Although the studies are not well designed as discussed further subsequently, controlled studies provide little support for surgery in children with spina bifida with dislocated hips or scoliosis.

Most reviews such as this have a number of limitations. First, as a result of the lack of randomized clinical trials, the strength of any grade of recommendations attached to any treatment recommendations for these patients is at best Grade B. Second, as a result of the lack of consistent criteria across studies, a formal meta-analysis to accumulate a larger sample size was not appropriate. Third, difficulties in interpreting retrospective studies are many. None of the studies were multicenter and therefore the number of patients in these single-center studies was relatively small. The power to perform meaningful statistical comparison was compromised by the low sample sizes. Fourth, the retrospective studies were inconsistent in reporting benefit. Benefit except in two studies was not reported using validated outcome measures. Fifth, complications such as infection and nonunion are variably reported and even when reported did not use explicit criteria. Any criteria that were described varied between studies making comparison of rates impossible. Sixth, duration of followup was variable. Finally, none of the trials were prospective or randomized compromising the ability to accurately and completely reflect surgical prognosis. Many of the elements of a well-designed uncontrolled study were also lacking, including concealed allocation, blinding, and intervention-to-treat analyses. Many of the studies had important differences in baseline prognostic variables but only two [3, 14] attempted to adjust for these differences.

Children with lesions above L4 have a low probability of walking in the community beyond adolescence. The literature provided no support for the surgical treatment of the dislocated hip in these children. In children at L4 level and below, the majority will walk in their home or community. Again, not one study reported a reduced hip improved walking ability.

In evaluating the role of surgery, a surgeon must also consider the risk of complications. Several studies documented substantial risks of surgery, including pressure sores and fractures with redislocation rates ranging from 30% to 45%. The literature provided no direct comparisons of different surgical procedures. The one subgroup of patients who theoretically may benefit from surgical treatment is children with a unilateral dislocation below L4 who have an associated decline in walking ability. Without treatment, these children may have a limb length discrepancy and a worsened Trendelenburg gait. However, whether surgery improves function in this one subgroup of patients is unknown from the literature.

We know very little about the long-term clinical course of untreated scoliosis. It is unknown whether these curves progress in adulthood, which might have implications for physical function and respiratory function. In the short term, spinal instrumentation and fusion may result in a small improvement in pulmonary function, but this again is of questionable clinical importance. Although scoliosis surgery can improve radiographic parameters, the overall effect on physical function was mixed. Although there may be some improvement in seating, several studies report walking ability may be compromised. The cross-sectional study of Wai et al. [28] found little relationship between any aspect of the spinal deformity and physical function or self-perception. Wai et al. [28] raised the possibility that sitting function may be improved simply by chair modification rather than scoliosis surgery. The ability of surgery to improve physical function and seating function in the short term and long term is an unresolved question. If improved seating is the primary rationale for surgery, then spinal balance is probably the key deformity to correct.

Scoliosis is an even more substantial surgical undertaking than hip surgery for the child and family. The risks associated with this procedure include infection rates from the identified studies in the range of 5% to 20% and nonunion rates up to 40%. The majority of studies focused on the surgical correction of spinal deformity. All comparative studies concluded that combined anterior and posterior instrumentation and fusion provided the best correction with the lowest nonunion rates. Although one study concluded anterior instrumentation did not improve correction [29], this remains an unresolved question. In addition, although one study found an all-pedicle posterior-only approach provided comparable deformity correction to

**Table 3.** Search strategy: hip and spine surgery in children with spina bifida

Recommendations on treatment	Grade of recommendation
The dislocated hip in all children (except unilateral dislocation in patients with lesions at or below L4) should NOT be treated surgically to reduce hip	B
The unilateral dislocated hip in children at or below L4 should be surgically reduced	I
The patient with spina bifida and scoliosis should NOT receive surgery	I
The surgical treatment for spina bifida scoliosis should be anterior fusion and posterior fusion and instrumentation	B
Anterior instrumentation should be added to anterior fusion for spina bifida scoliosis	I
All-pedicle posterior only approach appropriate for treatment of spina bifida scoliosis	I

anterior and posterior surgery [21], this is also an unresolved question.

The recommendations arising from this review are provided (Table 3). The conclusions are quite similar to the conclusions of Dias and Swaroop [13]. Although we have framed the questions differently, the conclusions that hip surgery for the dislocated hip has a minimal role and that the role for spinal instrumentation and fusion was unclear from the literature were similar. The benefits of spine surgery are uncertain (Grade I). Spine surgery, if performed, should be combined anterior and posterior (Grade B). An all-pedicle approach for scoliosis surgery may be effective (Level I). Hip reduction surgery does not improve function (Grade B) but may be appropriate in low-level unilateral dislocation (Level I).

## Appendix 1. Summary of key articles

### Hip Dislocation

Bazih and Gross [7] evaluated 74 patients with myelomeningocele older than 4 years including all neurosegmental levels. Of the 74 patients, 18 had had surgery. The subluxation and redislocation rate after surgery was 45%. The level of neurosegmental defect, not hip dislocation, was the most important factor in determining ambulation. The key comparison, in lower lumbar patients, found 10 of 13 who had surgery compared with 14 of 14 patients who had not had surgery were household ambulators or better. The authors concluded there was no overall beneficial effect of surgery on ambulation.

Crandall et al. [11] reviewed 100 patients with dislocation or hip dysplasia older than 10 years. Ambulatory

function at each neurosegmental level was not affected by whether the hips were located or dislocated. In the lower-level patients, two of eight nonambulators compared with five of 15 ambulators had hip dislocations. The authors concluded the functional level was related to the neurosegmental level and not whether the hips were located.

Feiwell et al. [14] studied 76 patients with spina bifida of all neurosegmental levels older than 5 years. Of the 76 patients, 41 had no operative treatment for the hip and 35 had been operated on one or more time to reduce the hip. The redislocation rate after surgery was 40%. Using multivariable analyses, the presence of a reduced hip was not related to ambulatory ability. The key comparison for patients with lower lumbar level found, of 16 ambulators, five (30%) had dislocated hips, whereas of the five patients who were nonambulators, one (20%) had a dislocated hip. The authors concluded hip dislocation was not related to ambulation.

Sherk and Ames [25] examined 36 children who had open reduction and iliopsoas transfer with other hip procedures for lumbar L1 to L5 level patients. Of the 36 children, 19 patients had a redislocation of one or both hips. The authors concluded the presence of located hips was unrelated to walking ability.

Alman et al. [3] reviewed 52 children with dislocated hips with L3 or L4 level lesions. Of the 52 patients, 30 patients had a variety of surgical procedures. The overall redislocation rate after surgery was 30%. The ambulatory level and functional ability scores (using Child Health Assessment Questionnaire and Rand Health Insurance Functional Score) of patients with and without surgery was similar. Restriction in motion and limb length discrepancy was greater in the surgical group. A small subgroup of 12 patients who had surgery had a slightly increased efficiency of gait. The authors concluded the benefits of surgery for surgical dislocation were at best marginal.

In addition to the therapeutic papers examining the benefits of hip surgeries, 10 Level II prognostic studies examined factors related to ambulatory ability in patients with spina bifida. The reported factors related to walking included cognitive ability [10], physiotherapy [10], compliant parents [10], neurosegmental level [6, 12, 15, 30], club foot deformity [12], hip and knee contractures [2, 6, 23], low back pain [6], lack of motivation [6], age [2, 23, 30], and scoliosis [23]. None of the studies reported a relationship between hip location and walking ability.

### Scoliosis

Unfortunately, none of the identified studies directly compared the function of patients with spina bifida and scoliosis with and without surgery. Only two

cross-sectional studies examined the relationship between function and the presence of scoliosis. Kahanovitz and Duncan [17] evaluated 39 patients with spina bifida and scoliosis with an average of 19.9 years' followup. Of the 39 patients, 15 had had surgery. Of these 15 patients, eight maintained, seven lost, and none improved their functional level. Although the presence of scoliosis and pelvic obliquity were related to poor seating, the authors concluded the benefits surgery were uncertain. Wai et al. [28] performed a cross-sectional study of 98 children with spina bifida and scoliosis. Using multivariable analyses, all aspects of spinal deformity were evaluated for their relationship with sitting balance, Jebsen Hand scale, Hoffer ambulation scale, Spina bifida Spine Questionnaire, Harter's Self Perception Scale, and the Activities Scale for Kids. Of all aspects of spinal deformity, only one, spinal balance, had any clinically significant relationship with only one aspect of function, seating. The authors concluded, in the short term, the potential benefits of surgery may be at best to improve only seating balance. Thus, the two studies that evaluated the relationship between surgery and function found no or minimal benefit.

The remainder of the studies focused on comparisons of different surgical techniques. Mazur et al. [18] evaluated 49 patients with spina bifida and scoliosis comparing anterior and posterior surgery with posterior and anterior surgery alone. Although sitting was improved in 70% of patients, the ability to ambulate was adversely affected in 67% of patients who had received anterior and posterior surgery. Although correction of deformity and pelvic obliquity were similar among the three methods, the rate of nonunion was 11% in the combined anterior and posterior group compared with 33% and 29% in the posterior and anterior fusion alone groups, respectively. The conclusion was that a combined anterior and posterior approach was most appropriate for children with spina bifida.

Parisini et al. [21] compared Harrington, Harrington-Luque, and all-pedicle screws posterior-only approaches with a two-stage anterior and posterior spinal fusion in 33 patients with spina bifida. The correction of deformity was greater in both the combined anterior and posterior fusion group and the all-pedicle posterior-only approach. The authors concluded the combined anterior and posterior procedure and all-pedicle single posterior approach using pedicle screws provided satisfactory correction.

Stella et al. [27] reviewed, at an average of 8 years' followup, the surgical treatment of 14 patients with scoliosis and spina bifida. Although the numbers are small, the conclusion of the authors was that combined anterior and posterior surgery with fusion to the sacrum provided the best results.

Stella et al. [27] evaluated 29 patients with spina bifida who were treated using spinal fusion techniques, seven by posterior arthrodesis, three by anterior arthrodesis, and 19 by the combined anterior and posterior approach. The correction of deformity was highest in the combined anterior and posterior instrumentation and fusion group with 14% nonunion. The authors concluded combined anterior and posterior instrumentation fusion gave the best results.

Ward et al. [29] compared anterior and posterior fusion with anterior or posterior procedures alone. Although a variety of techniques were used, better curve correction and lower nonunion rates were obtained with a combined anterior and posterior approach. The authors reported no advantage to adding anterior instrumentation to the anterior approach and no superiority of Luque or Harrington posterior instrumentation.

Banit et al. [4] reviewed 50 patients with at least 1-year followup of which six received Harrington rods and 47 received segmental fixation. The authors concluded the combined anterior and posterior surgery with segmental fixation had the lowest nonunion rate of 16%.

Osebold et al. [20] reported 40 patients who had spina bifida and scoliosis. The author reported combined anterior and posterior surgery had better correction of deformity and a nonunion rate of 23% compared with a rate of 46% in the posterior fusion-only group.

Only two studies, both case series (Level IV), addressed the issue of pulmonary function after spine fusion for patients with spina bifida and scoliosis. Banta and Park [5] evaluated 10 patients on average 10 months after staged anterior Dwyer and posterior Harrington instrumentation. Although peak flow increased in eight, forced vital capacity and forced expiratory volume in 1 second (FEV1) decreased in five patients. Carstens et al. [9] evaluated 10 patients 13 months after anterior and posterior instrumentation with fusion. The results were mixed, but eight had an increase in vital capacity and six had an increase in FEV1.

Although only a case series (Level IV), Schoenmakers et al. [24] performed the only study to examine the functional abilities before and after spinal fusion. Ten children with spina bifida were followed prospectively and evaluated with the Pediatric Evaluation of Disability Inventory on average 18 months after surgery. Although all patients had improvement in their spinal deformity, of four patients who had been able to ambulate before surgery, three had difficulty after surgery. Patients also experienced a post-surgical decline lasting for at least 6 months. By 18 months after the surgery, there had been a slight improvement in functional skills, specifically self-care, indicating less caregiver assistance. However, the overall complication rate was 80%.

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