Author/s:
Toovey, R;Harvey, A;Johnson, M;Baker, L;Williams, K

Title:
Outcomes after scoliosis surgery for children with cerebral palsy: a systematic review

Date:
2017-07-01

Citation:
Toovey, R., Harvey, A., Johnson, M., Baker, L. & Williams, K. (2017). Outcomes after scoliosis surgery for children with cerebral palsy: a systematic review. Developmental Medicine and Child Neurology, 59 (7), pp.690-698. https://doi.org/10.1111/dmcn.13412.

Persistent Link:
https://hdl.handle.net/11343/292605

Received Date : 21-Oct-2016

Revised Date : 18-Jan-2017

Accepted Date : 19-Jan-2017

Article type : Systematic Review

[systematic review; 1 table; 1 appendix, 1 figure, 4 tables online]

Outcomes after scoliosis surgery for children with cerebral palsy: a systematic review

RACHEL TOOVEY[1]

ADRIENNE HARVEY[1,2,3]

MICHAEL JOHNSON[4]

LOUISE BAKER[2]

KATRINA WILLIAMS[1,2,3]

1 Developmental Disability and Rehabilitation Research, Murdoch Childrens Research Institute, Parkville; **2** Developmental Medicine, The Royal Children's Hospital, Melbourne; **3** Department of Paediatrics, University of Melbourne, Parkville; **4** Orthopaedic Surgery, The Royal Children's Hospital, Melbourne, Vic, Australia

Correspondence to Rachel Toovey at Developmental Disability and Rehabilitation Research, Murdoch Childrens Research Institute, 50 Flemington Rd, Parkville, Vic, Australia 3052. E-mail: rachel.toovey@mcri.edu.au

PUBLICATION DATA

This is the author manuscript accepted for publication and has undergone full peer review but has not been through the copyediting, typesetting, pagination and proofreading process, which may lead to differences between this version and the Version of Record. Please cite this article as doi: 10.1111/dmcn.13412

Accepted for publication 19 January 2017.
Published online 00 Month 2017.

ABBREVIATIONS

LOS Length of stay
QoL Quality of life

[Abstract]

AIM This study aims (1) to evaluate and synthesise the evidence for the postoperative outcomes after scoliosis surgery for children with cerebral palsy (CP), and (2) to identify preoperative risk factors for adverse outcomes after surgery.

METHOD Medline, EMBASE, CINAHL, and PubMed were searched for relevant literature. Included studies were assessed for risk of bias using the Cochrane Effective Practice and Organisation of Care tool. Quality of evidence for overall function, quality of life (QoL), gross motor function, caregiver outcomes, deformity correction, and postoperative complications were assessed using GRADE (Grades of Recommendation Assessment, Development and Evaluation).

RESULTS Fifty-one studies met inclusion criteria, including 35 case series designs. Risk of bias was high across all studies. On average good deformity correction was achieved, the trend appears positive for caregiver and QoL outcomes, but there was minimal to no change for gross motor or overall function. Inconsistent measurement limited synthesis. A mean overall complication rate of 38.1% (95% confidence interval 27.3% to 53.3%) was found. The quality of evidence was very low across all functional outcomes.

INTERPRETATION Limited high-quality evidence exists for outcomes after scoliosis surgery in children with CP, a procedure associated with a moderately high complication rate. The intervention appears indicated for deformity correction, but currently there is insufficient evidence to make recommendations for this surgery as a way to also improve functional outcomes, caregiver outcomes, and quality of life.

DOI:

Developmental Medicine & Child Neurology 2017, 59: 000−000

What this paper adds

- Scoliosis surgery in children with cerebral palsy has a moderately high complication rate (38%).
- Surgery is indicated for deformity correction but evidence is lacking for improved functional outcomes, caregiver outcomes, and quality of life.

[main text]

Children with cerebral palsy (CP) have an increased risk of developing scoliosis.[1] The incidence varies but is generally accepted to be between 20% and 25%.[2] In a total population of 666 children with CP aged 4 years to 18 years in Sweden, 17% had mild scoliosis while 11% had moderate or severe scoliosis based on clinical examination, with the risk of developing scoliosis increasing with age and severity of motor impairment.[1] The low levels of evidence of studies examining risk factors for the emergence and progression of scoliosis in children with CP (Gross Motor Function Classification System [GMFCS] levels IV to V) make it difficult to draw firm conclusions.[3]

The definitive treatment for progressive scoliosis in children with CP is surgical intervention with the aim to halt curve progression, level the pelvis, and achieve good frontal and sagittal balance.[4] Most consider spinal fusion for curves that progress beyond 50° or those that have caused a deterioration in functional sitting[4]; however, there are no agreed criteria for recommending spine fusion surgery to parents/guardians and clinicians use widely divergent criteria for their surgical decision-making.[5]

For any proposed medical intervention, the diagnosis, course of the disease, goals of treatment, and risks and benefits must be carefully considered.[6] For children with CP who have scoliosis, other associated comorbidities may be present before surgery, including hip subluxation/dislocation, spasticity, pulmonary function, gastrointestinal issues and poor nutritional status, seizure disorders, and coagulopathy.[7] These may increase the complexity of management and potentially be determinants of the risks versus benefits of surgical correction.

Medical indications for scoliosis surgery in children with severe CP are conflicting, with variable benefits but a risk of substantial complications.[8] Overall complication rates ranging from 44% to 70% with a perioperative death rate of 0 to 7% have been reported.[7] Other complications include respiratory compromise and infection, wound infections, metalware complications, pseudo-arthrosis, urinary tract infections, gastrointestinal complications, severe bleeding, and spinal cord dysfunction.[5,7,9,10]

Facilitated by growing use of the World Health Organization International Classification of Functioning, Disability and Health as a common language, activity and participation outcomes are increasingly being recognised as important for both families and clinicians in decision-making.[11,12] However, there is limited evidence of the benefit versus risk of scoliosis surgery in children with CP with respect to these meaningful postoperative outcomes.

The primary aim of the review is to evaluate and synthesise the evidence for the clinical postoperative outcomes after scoliosis surgery for children with CP. The secondary aim is to identify preoperative risk factors for complications and/or poor outcomes after surgery.

METHOD

The protocol for this review was registered with the PROSPERO register of systematic reviews (http://www.crd.york.ac.uk/PROSPERO), registration number CRD42015024984.

Eligibility criteria

Published studies regarding spinal surgery for children with CP and scoliosis were included if they met the following criteria:

(1) They were categorised as level II to IV studies using the National Health and Medical Research Council (NHMRC) levels of evidence.[13] For intervention studies this included randomised and pseudorandomised controlled trials (level II, III-1), comparative studies with concurrent controls (III-2), comparative studies without comparative controls (III-3), and case series where *n* was greater than 10 (level IV). Level III to IV studies were included given the known lack of randomised controlled trials in surgical interventions and in this population. For prognostic studies this included prospective cohort studies (level II), retrospective cohort studies (level III-3) and case series where *n* was greater than 10 (level IV).

(2) More than 50% of the participants were children with CP aged 0 to 18 years.

(3) Involved all approaches and types of instrumentation used in spinal surgery for scoliosis. Studies involving other surgical procedures conducted within the same surgical episode (i.e. hip surgery) were excluded.

(4) Outcomes were reported separately for children with CP and included one or more of the following: overall function, gross motor function, quality of life, parent or carer outcomes, scoliosis curve, and pelvic obliquity correction and complications.

Studies were also included if they examined risk factors for poor postoperative outcomes.

Search methods for identification of studies

Relevant articles were identified by searching OVID MEDLINE, EMBASE, CINAHL, and PubMed, with all searches restricted to articles published in peer-reviewed journals in English from 1980 to October 2015. The following search strategy using OVID terms was used: (cerebral palsy AND (scoliosis / su [surgery] OR spinal fusion/)) OR (cerebral palsy AND scoliosis/ep). Review authors also searched reference lists of included studies and other narrative reviews.

Study selection

Study eligibility was assessed by two of the review authors (RT and AH), who independently reviewed titles and abstracts of each study. If the inclusion criteria were met, full-text studies were independently evaluated.

Data extraction

For included studies, data were extracted based on the Cochrane recommendations[14] by two authors (RT and AH) and included study details, study design, NHMRC level of evidence,[13] characteristics of the study sample, details of the surgical intervention and comparison group if applicable, outcomes measured, effects of the intervention, and author conclusions. In studies involving participants other than those with CP, only outcomes data reported separately for children with CP were extracted. The data extraction form is available from study authors on request.

Assessment of risk of bias in individual studies

Risk of bias was assessed for individual studies using the criteria for reviews conducted by the Cochrane Effective Practice and Organisation of Care group[14] because of the likely lack

of randomised studies. Each article was assessed as low, high, or unclear risk for six criteria (see epoc.cochrane.org/epoc-specific-resources-review-authors).

Measures of treatment effect

REDCap data capture tool[15] and Stata v14.0 (www.stata.com; StataCorp., College Station, Texas) were used for data management and analysis. Outcomes data for individual studies were extracted and treated as whole group data, even in the studies where two surgical techniques were compared because of the heterogeneity in techniques. Variation in measurement meant only trends could be reported for functional outcomes, gross motor function, quality of life, and parent or caregiver outcomes. Outcome measurement was more uniform for curve and pelvic obliquity correction and complications. Summary statistics were used to describe mean short- and longer-term deformity correction and length of stay outcomes. Random effects logistic regression was used to determine overall complication rate and the odds of experiencing at least one complication from studies that measured complication rate as the proportion of participants who experienced at least one complication (minor and/or major). Studies reporting associations or correlations of preoperative factors with poor postoperative outcomes were presented in table form.

Assessment of the quality of evidence and recommendations across studies

The GRADE (Grades of Recommendation Assessment, Development and Evaluation) system was used to assess the quality of the evidence. It is an evidence-grading tool, endorsed by the World Health Organization and widely used, including in Cochrane reviews,[16] which rates the quality of the evidence for outcomes of interest across the studies and overall as well as the strength of recommendation for use of the intervention.

RESULTS

Characteristics of included studies

Fifty-one studies were included from a total of 224 titles and abstracts screened. Figure S1 (online supporting information) shows the PRISMA flow diagram[14] of included studies and Table SI (online supporting information) shows characteristics of included studies. Only one study was prospective,[17] and the majority were case series (*n*=35) or cohort studies (*n*=9). Inclusion criteria were generally described, yet in 30 studies exclusion criteria were not stated. Forty-two studies included only children with CP, whereas nine studies included children with mixed neuromuscular conditions (*n*=7) or adolescent idiopathic scoliosis (*n*=1)

or both (*n*=1). Descriptive information about participants was often missing. Only nine studies used the GMFCS to describe the sample and in these studies over 75% of the sample were classified as GMFCS level IV to V.[4,17–24] Twelve studies included posterior approaches to surgery only, 33 studies included posterior approaches or combined/staged anterio-posterior approaches, while approach was not reported, or was not defined for participants with CP, in six studies. Length of follow-up varied greatly among the studies and all except for five studies[18,25–28] did not state measurement time points or they were not standardised. Only six studies reported any missing data or loss to follow-up.[17,20,26,27,29–31]

Risk of bias assessment

There was an overall high risk of bias within and across included studies as shown in Table SI.

Outcomes

For each outcome, measurement time points were inconsistent, poorly defined, or not reported between and within the studies. Table SII (online supporting information) shows which specific functional outcomes of interest were reported and how they were measured.

Overall function

Twelve studies reported outcomes related to overall function[20,24,27–29,32–38] but with function measured differently between studies. All studies primarily involved parent report through questionnaires or interviews and included items about overall health and function, pain, social and communication, and upper limb function. Only two studies[27,32] used validated questionnaires (POSNA questionnaire, CP-CHILD, respectively) to measure overall function pre and postoperatively. In these two studies, the trend was towards no change in overall function but reported improvement in the functional areas of pain or comfort postoperatively. The remaining studies were limited by a lack of preoperative comparison, and retrospective assessment by parents without baseline data with a high likelihood of recall bias. Disparity in outcome measures used meant that it was not possible to analyse overall effect.

Quality of life

Three studies reported quality of life (QoL)[28,32,38]; however, QoL was defined and measured differently in these studies. One study used a validated disease-specific tool (CP-CHILD) to measure health-related QoL.[32] All studies involved parent (proxy) report. Two of the three

studies[32,38] compared preoperative and postoperative QoL, but measurement time-points were inconsistent or missing. Significant improvements were seen in health-related QoL as measured by the CP-CHILD at minimum 2-year follow-up in the domains of comfort and emotions, health, and overall quality of life, as well as in the total score.[32] Seventy-one per cent of parents completing the non-validated carer questionnaire[38] also reported improvements in QoL; however, both studies lacked a control group. The effect could not be examined in the remaining study[28] given lack of preoperative report of QoL.

Gross motor function

Eight studies reported gross motor function outcomes[28,29,34,36,39–42]; however, it was defined and measured differently in all but two,[34,41] which used the Rancho Los Amigos Hospital Classification System.[43] In these two studies[34,41] 32% to 42% of the patients made gross motor function gains, moving up one level on a 5-point scale of gross motor function. However, follow-up was inconsistent and no comparison group was available. One study[36] used gait analysis whereas another used the World Health Organization functional level classification.[28] No change in ambulatory function was found on gait analysis[36] and minimal to no change was seen postoperatively in motor function using the World Health Organization functional levels classification system.[28] Similarly, no change in gross motor function was reported in the remaining four studies[29,39,40,42] where gross motor function was generally assessed subjectively and retrospectively with high likelihood of recall bias. Disparity in outcome measures meant that it was not possible to analyse overall effect.

Caregiver outcomes

Ten studies reported parent or caregiver outcomes,[20,23,27–29,32,35,38,39,44] which were defined and measured differently in all studies but consistently involved questionnaires or interview. Constructs measured included parent satisfaction,[27,28,32,35,38,44] ease of care,[20,23,29,34,39] and parent assessment of utility.[23] For studies that reported parent satisfaction[27,28,32,35,38,44] outcomes were generally positive while mixed ease of care outcomes were reported.[20,23,29,39] In the study [39] that assessed ease of care prospectively and compared outcomes with a control group who did not undergo surgery, no difference was found. Another study reported no decrease in care required postoperatively,[23] whereas the parents reported increased ease of care in the other two studies.[20,29] Outcomes were assessed retrospectively in all but one study.[39] Disparity in definitions of caregiver outcomes and measures used meant that it was not possible to analyse, estimate, or calculate overall effect.

Scoliosis curve correction

Thirty-two studies measured scoliosis curve correction,[9,18,20,21,23,24,27–30,32–34,36–38,40–42,44–56] with all studies using the Cobb angle.[57] Studies tended to measure whole group curve correction as mean percentage (%) correction or mean change in Cobb angle. Where reported, variability in the data was generally expressed in terms of range, rather than standard deviation. All studies reported a positive effect. Twenty studies reported shorter-term (typically 0 to 3mo) postoperative mean % curve correction.[9,18,20,21,28–34,40–42,44,45,47,52,54,55] The overall mean shorter-term postoperative curve correction was found to be 59%, with mean correction ranging from 36% to 77%. Fourteen studies reported longer-term (typically >2y) postoperative mean % curve correction.[9,23,24,28,33,37,38,44–47,49,50,56] The overall mean longer-term postoperative curve correction was found to be 61.4% mean curve correction, with means ranging from 55% to 78%. Mean whole group preoperative Cobb angles ranged from 65° to 82° and mean postoperative angles from 19° to 37°.

Pelvic obliquity correction

Twenty-six studies measured pelvic obliquity correction[18,20,21,23,24,28,29,32–34,36–38,41,42,44–46,48–51,54–56]; however, it was inconsistently defined and measured. Of the 10 studies that defined how pelvic obliquity was measured,[18,21,23,28,29,33,44,48,51,56] six defined it as the angle between the line across the level of the iliac crests and the line perpendicular to the line from the centre of T1 to S1,[23,29,33,44,51,56] whereas the other four studies used one of three other definitions. Studies tended to report group mean preoperative and postoperative pelvic obliquity angles or group mean percentage (%) pelvic obliquity correction.

All studies reported a positive effect. Twelve studies reported shorter-term mean pelvic obliquity % correction.[9,23,24,29,31–33,41,42,44,55,56] The overall mean shorter-term pelvic obliquity correction was found to be 65.5%, with means ranging from 48% to 83% correction. Seven studies reported longer-term pelvic obliquity % correction.[9,28,33,41,46,49,54] The overall mean longer-term pelvic obliquity correction was found to be 55%, with means ranging from 43% to 81%. Mean preoperative pelvic obliquity angles ranged from 9° to 25°, and mean postoperative angles from 4° to 10°.

Complications

Complications were reported in 45 studies.[4,9,10,17–24,26–29,31–34,36–42,44–56,58–63] Methods of reporting complications varied among studies. Nineteen studies reported whole group

complication rate, with 12 studies[9,20,27,32,38,39,45,50,51,54,56,62] measuring the complication rate as the percentage of patients who developed at least one complication (major and/or minor). The remaining studies reported number of complications per sample (with some children likely having more than one complication) or it was unclear. There was inconsistency with respect to reporting type of complications, for example major versus minor and the classification of complications, such as respiratory or neurological. Major versus minor classification was used in eight studies[18,19,27,28,48,55,62,64]; however, among these studies there were five different definitions of what constituted a major or minor complication. Types of complications are reported in Table SIII (online supporting information).

Overall complication rate

A random effects logistic regression model found patients had, on average, 1:3 odds of having at least one complication after surgery. Further, the mean proportion of patients who experienced at least one complication was 38.1% (95% confidence interval 27.3% to 53.3%), with the range of between-study proportions from 16% to 70%. However, heterogeneity was high. In some studies, the odds were in favour of not having a complication, whereas in other studies the odds were that a patient would have a complication after surgery.

Eighteen studies examined postoperative length of stay (LOS) in the paediatric intensive care unit.[9,18–21,23,24,28–31,33,41,44,45,47,48,55] In the 16 studies [9,18–20,23,24,28–31,33,41,45,47,48,55] that reported this as mean LOS in the paediatric intensive care unit, the overall mean was 4.4 days, with means ranging from 1.7 to 6.7 days.

Twenty-one studies examined overall hospital LOS.[9,18,20,21,23,24,28–31,33,39–41,44,45,47,48,51,55,60] Of the 18 studies that reported this as mean hospital LOS, the overall mean was 16.9 days, with mean hospital LOS ranging from 8.7 to 24.5 days.[9,20,23,24,28–31,33,39–41,45,47,48,51,55,60]

GRADE assessment of the quality of the evidence

Table I shows that the quality of evidence was very low for each postoperative outcome after spinal surgery for children with CP and scoliosis, aside from curve correction, for which the quality of evidence was low.

Preoperative risk factors for poor postoperative outcomes

Eighteen studies reported preoperative factors for poor postoperative complications[17–20,22,25,31,32,38,45,51,53,55,58,60,64–66]; however, only 11 of these reported statistical associations or

correlations separately for CP samples (Table SIV, online supporting information).[17,20,22,23,25,53,60,64–66] Comorbidities including gastrointestinal issues, respiratory conditions, seizures, malnutrition, intellectual disability, reduced speech ability, and prior surgical infection were identified as risk factors in addition to an increased weight, older age, higher preoperative platelet or white cell count, and the type or severity of preoperative spinal or pelvic deformity.

DISCUSSION

This systematic review examined the clinical postoperative outcomes after scoliosis surgery for children with CP and found that there are low levels of evidence for the improvement of curve and pelvic obliquity, a possible trend of improved functional outcomes but a high complication rate. Some risk factors for adverse outcomes were identified. The poor quality of the evidence resulting from study designs and inconsistencies in measurement and reporting preclude the ability to make clear recommendations that take into account important functional and participation outcomes of surgery, for the child and their family.

Most of the studies included were case series. There is debate as to whether Level 1 evidence is necessary or indicated in this population because of challenges with blinding, randomisation when patient preference might be to have surgery, and variability of surgical technique and experience.[8] However, there are avenues of data collection that could strengthen the level of evidence.

There are a number of limitations within each study and across the overall body of evidence. Many studies focused on deformity correction and complications with less emphasis on the effect of surgery on physical or respiratory function, participation, quality of life, positioning, pain, and caregiver outcomes. Those studies that did include some of these important patient and family outcomes were not consistent in their reporting. There was a lack of using established and validated outcome measures and many relied on retrospective recall by parents or caregivers. Of the studies that reported curve and pelvic obliquity correction and complication rates, there were inconsistencies in pelvic obliquity measurement and reporting of complications.

The secondary aim of the review was to examine risk factors for poorer outcomes after surgery. Although a number of factors were reported to be associated with wound infection, overall complications, or overall infection in particular, a range of variables were used as potential risk factors and outcomes were inconsistently defined. Further, risk factor analysis was not an objective of a number of studies, making them inappropriately powered.

Specific prognostic studies that consider standardised variables for standardised poor outcomes would be beneficial.

Studies have been published in this area since this review was conducted. Promisingly, these studies have a greater focus on QoL, activity, and participation outcomes,[67,68] as well as consideration of the importance of pelvic obliquity correction in this population.[69] Systematic means of determining risk of complications are also re-emerging with the use of a preoperative risk score and postoperative complications score in a recent study,[70] first used in 1999.[64] The Clavien-Dindo scale, an established tool for classifying surgical complications[71] has also been used in another recent study examining complications after neuromuscular scoliosis surgery.[72]

Although the results of this review must be interpreted with caution, it is important to acknowledge that lack of evidence for a procedure does not mean it should be dismissed.[8] Clinical experience suggests that the natural history of severe scoliosis in children with CP without surgical correction is progressive in terms of both deformity and functional outcomes including seating, pain, and skin issues. However, longitudinal studies documenting this are few and largely focussed on deformity outcomes,[73,74] whereas intervention studies with well controlled natural history groups[39] are limited. Given clinical awareness of the deteriorating curve and potential for subsequent loss of function in children and young people with CP, it has become mainstream care to offer surgical repair except in situations where surgical or anaesthetic risks are likely to result in death or worsening disability. This approach means that there is unlikely to be new evidence emerging about scoliosis progression and outcomes except in children and young people at the most severe end of the spectrum. However, the quality of evidence can still be improved in a way that will provide information that is important for decision-making. Consistency in reporting would significantly improve this. What is unclear is which children need surgery at what time for optimal outcomes where the benefit outweighs the risk.

For this population of children who often have complex comorbidities that put them at higher risk of complications during and after spinal surgery, it is essential that there is consistent and coordinated preoperative assessment and care-planning incorporating all those involved in their overall care. To accurately assess outcomes after surgery there is a need for prospective and controlled evaluation of spinal surgery for the treatment of scoliosis in patients with severe CP including measuring common objective data for patient-centred outcomes including complications, eliminating or limiting bias wherever possible, and consistency in inclusion and analysis.[8] International consensus on what this prospective data

collection should include is very important to ensure consistency across all centres and should be the focus of future work in this area across all outcomes.

This review is not without limitations. The broad inclusion criteria make specific conclusions difficult to draw. The use of aggregated group data rather than comparing types of surgery does not account for any differences in outcomes after the different surgical procedures included in the studies. There are also limitations in the tools available to assess risk of bias in non-randomised studies. Further, the considerable weight type of study design carries in the GRADE system for assessing the overall quality of evidence and determining the strength of recommendations means even though there is a large number of included studies, the retrospective case series design used in most studies means the evidence is initially rated very low, and has a high risk of bias, making it difficult to achieve a moderate or high level of evidence rating. However, the GRADE system is the most widely used system for assessing the quality of the evidence. It allows for comparison of the body of evidence for this operation to other interventions in this population.

Conclusion and recommendations

Considering the trade-offs among the benefits (curve and pelvic obliquity correction, trend towards positive QoL, and caregiver outcomes), the harms of the intervention (moderately high complication rate), the overall very low quality of the evidence, and the baseline risk of children with severe CP in developing progressive scoliosis, the overall strength of recommendation following this review is a weak positive: 'probably do it'. Surgery appears to be indicated for spinal and pelvic deformity correction, but it is not clear whether it is indicated for the remaining child and family outcomes in the domains of activity, participation, and quality of life.

This complex procedure requires careful preoperative planning and comprehensive longitudinal outcomes assessment to be able to accurately report postoperative outcomes. International consensus is required to ensure prospective and consistent methods of collecting these data. Evidence-based guidelines are needed to tailor surgery to individual children to optimise outcomes for each child after surgery.

ACKNOWLEDGEMENTS

This study was supported in kind by the NHMRC Centre of Research Excellence in Cerebral Palsy, the MCRI Developmental Disability and Rehabilitation Research Group and the RCH Developmental Medicine and Orthopaedic Surgery Departments. No funding was received to

undertake this systematic review. The authors have stated that they had no interests which might be perceived as posing a conflict or bias.

SUPPORTING INFORMATION

The following additional material may be found online:

Figure S1: PRISMA flow chart of included and excluded studies.

Appendix S1: Excluded studies and reasons for exclusion (date, outcomes not reported separately for CP, intraoperative, only report curve/pelvic obliquity, $n<10$, type of paper)

Table SI: Characteristics of included studies

Table SII: Outcomes reported and how they were measured (for functional outcomes only)

Table SIII: Types and rates of complications

Table SIV: Studies reporting significant associations between preoperative risk factors and postoperative outcomes

REFERENCES

1 Persson-Bunke M, Hagglund G, Lauge-Pedersen H, Wagner P, Westbom L. Scoliosis in a total population of children with cerebral palsy. *Spine* 2012; **37**: E708–E13.

2 Imrie MN, Yaszay B. Management of spinal deformity in cerebral palsy. *Orthop Clin North Am* 2010; **41**: 531–47.

3 Loeters MJ, Maathuis CG, Hadders-Algra M. Risk factors for emergence and progression of scoliosis in children with severe cerebral palsy. *Dev Med Child Neurol* 2010; **52**: 605–11.

4 Yaszay B, Scannell BP, Bomar JD, Sponseller PD, Shah SA, Asghar J, Samdani AF, Bastrom TP, Newton PO. Although inconvenient, baclofen pumps do not complicate scoliosis surgery in patients with cerebral palsy. *Spine* 2015; **40**: E504–E9.

5 Koop SE. Scoliosis in cerebral palsy. *Dev Med Child Neurol* 2009; **51**: 92–8.

6 Lo B. Resolving ethical dilemmas: a guide for clinicians. 5th ed. Philadephia: Lippincott Williams & Wilkins, 2013.

7 Sarwark J, Sarwahi, V. New strategies and decision making in the management of neuromuscular scoliosis. *Orthop Clin North Am* 2007; **38**: 485–96.

8 Whitaker AT, Sharkey M, Diab M. Spinal fusion for scoliosis in patients with globally involved cerebral palsy. An ethical assessment. *J Bone Joint Surg Am* 2015; **97**: 782–7.

9 Nectoux E, Giacomelli MC, Karger C, Herbaux B, Clavert JM. Complications of the luque-galveston scoliosis correction technique in paediatric cerebral palsy. *Orthop Traumatol Surg Res* 2010; **96**: 354–61.

10 Asher MA, Lai SM, Burton DC. Subsequent, unplanned spine surgery and life survival of patients operated for neuropathic spine deformity. *Spine* 2012; **37**: E51–9.

11 Rosenbaum P. Cerebral palsy: what parents and doctors want to know. *BMJ* 2003; **326**: 970.

12 Vargus-Adams JN, Martin LK. Measuring what matters in cerebral palsy: a breadth of important domains and outcome measures. *Arch Phys Med Rehabil* 2009; **90**: 2089–95.

13 National Health and Medical Research Council. NHMRC additional levels of evidence and grades for recommendations for developers of guidelines. Available at: www.nhmrc.gov.au/_files_nhmrc/file/guidelines/developers/nhmrc_levels_grades_evidence_120423.pdf (accessed 1 Oct 2016).

14 Higgins JPT, Green S. Cochrane Handbook for Systematic Review of Interventions. Available at: handbook.cochrane.org (accessed 1 Oct 2016).

15 Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap) – a metadata-driven methodology and workflow process for

providing translational research information support. . *J Biomed Inform* 2009; **42**: 377–81.

16 Guyatt GH, Oxman AD, Vist EG, Kunz R, Flack-Ytter Y, Alonso-Coello P, Schunemann HJ, for the GRADE working group. GRADE: an emerging consensus on rating quality of evidence and strength of recommendations. *BMJ* 2008; **336**: 924–26.

17 Sponseller PD, Jain A, Shah SA, Samdani A, Yaszay B, Newton PO, Thaxton LM, Bastrom TP, Marks MC. Deep wound infections after spinal fusion in children with cerebral palsy: A prospective cohort study. *Spine* 2013; **38**: 2023–7.

18 Beckmann K, Lange T, Gosheger G, Bovingloh AS, Borowski M, Bullmann V, Liljenqvist U, Schulte T. Surgical correction of scoliosis in patients with severe cerebral palsy. *Eur Spine J* 2015.

19 Hod-Feins R, Anekstein Y, Mirovsky Y, Barr J, Abu-Kishk I, Lahat E, Eshel G. Pediatric scoliosis surgery – the association between preoperative risk factors and postoperative complications with emphasis on cerebral palsy children. *Neuropediatrics* 2007; **38**: 239–43.

20 Karampalis C, Tsirikos AI. The surgical treatment of lordoscoliosis and hyperlordosis in patients with quadriplegic cerebral palsy. *Bone Joint J.* 2014; **96B**: 800–6.

21 Sitoula P, Holmes L, Jr., Sees J, Rogers K, Dabney K, Miller F. The long-term outcome of early spine fusion for scoliosis in children with cerebral palsy. *J Spinal Disord Tech* 2014; **29**: e406–e412.

22 Sponseller PD, Shah SA, Abel MF, Newton PO, Letko L, Marks M. Infection rate after spine surgery in cerebral palsy is high and impairs results: multicenter analysis of risk factors and treatment. *Clin Orthop Relat Res* 2010; **468**: 711–6.

23 Sponseller PD, Shah SA, Abel MF, Sucato D, Newton PO, Shufflebarger H, Lenke LG, Letko L, Betz R, Marks M, Bastrom T, for the Harms Study Group. Scoliosis surgery in cerebral palsy: differences between unit rod and custom rods. *Spine* 2009; **34**: 840–4.

24 Tsirikos AI, Mains E. Surgical correction of spinal deformity in patients with cerebral palsy using pedicle screw instrumentation. *J Spinal Disord Tech* 2012; **25**: 401–8.

25 Chidambaran V, Gentry C, Ajuba-Iwuji C, Sponsellar PD, Ain M, Lin E, Zhang X, Klaus SA, Njoku DB. A retrospective identification of gastroesophageal reflux disease as a new risk factor for surgical site infection in cerebral palsy patients after spine surgery. *Anesth Analg* 2013; **117**: 162–8.

26 Ilharreborde B, Hoffmann E, Tavakoli S, Queinnec S, Fitoussi F, Presedo A, Pennecot GF, Mazda K. Intrasacral rod fixation for pediatric long spinal fusion: results of a prospective study with a minimum 5-year follow-up. *J Pediatr Orthop* 2009; **29**: 594–601.

27 Jones KB, Sponseller PD, Shindle MK, McCarthy ML. Longitudinal parental perceptions of spinal fusion for neuromuscular spine deformity in patients with totally involved cerebral palsy. *J Pediatr Orthop* 2003; **23**: 143–9.

28 Teli MG, Cinnella P, Vincitorio F, Lovi A, Grava G, Brayda-Bruno M. Spinal fusion with Cotrel-Dubousset instrumentation for neuropathic scoliosis in patients with cerebral palsy. *Spine* 2006; **31**: E441–7.

29 Bulman WA, Dormans JP, Ecker ML, Drummond DS. Posterior spinal fusion for scoliosis in patients with cerebral palsy: a comparison of Luque rod and Unit Rod instrumentation. *J Pediatr Orthop* 1996; **16**: 314–23.

30 Smucker JD, Miller F. Crankshaft Effect after Posterior Spinal Fusion and Unit Rod Instrumentation in Children with Cerebral Palsy. *J Pediatr Orthop* 2001; **21**: 108–12.

31 Tsirikos AI, Chang WN, Dabney KW, Miller F, Glutting J. Life expectancy in pediatric patients with cerebral palsy and neuromuscular scoliosis who underwent spinal fusion. *Dev Med Child Neurol* 2003; **45**: 677–82.

32 Bohtz C, Meyer-Heim A, Min K. Changes in health-related quality of life after spinal fusion and scoliosis correction in patients with cerebral palsy. *J Pediatr Orthop* 2011; **31**: 668–73.

33 Dias RC, Miller F, Dabney K, Lipton G, Temple T. Surgical correction of spinal deformity using a unit rod in children with cerebral palsy. *J Pediatr Orthop* 1996; **16**: 734–40.

34 Sponseller PD, Whiffen JR, Drummond DS. Interspinous process segmental spinal instrumentation for scoliosis in cerebral palsy. *J Pediatr Orthop* 1986; **6**: 559–63.

35 Tsirikos AI, Chang WN, Dabney KW, Miller F. Comparison of parents' and caregivers' satisfaction after spinal fusion in children with cerebral palsy. *J Pediatr Orthop* 2004; **24**: 54–8.

36 Tsirikos AI, Chang WN, Shah SA, Dabney KW, Miller F. Preserving ambulatory potential in pediatric patients with cerebral palsy who undergo spinal fusion using unit rod instrumentation. *Spine* 2003; **28**: 480–3.

37 Tsirikos AI, Lipton G, Chang WN, Dabney KW, Miller F. Surgical correction of scoliosis in pediatric patients with cerebral palsy using the unit rod instrumentation. *Spine* 2008; **33**: 1133–40.

38 Watanabe K, Lenke LG, Daubs MD, Watanabe K, Bridwell KH, Stobbs G, Hensley M. Is spine deformity surgery in patients with spastic cerebral palsy truly beneficial?: a patient/parent evaluation. *Spine* 2009; **34**: 2222–32.

39 Cassidy C, Craig CL, Perry A, Karlin LI, Goldberg MJ. A reassessment of spinal stabilization in severe cerebral palsy. *J Pediatr Orthop* 1994; **14**: 731–39.

40 Gersoff WK, Renshaw TS. The treatment of scoliosis in cerebral palsy by posterior spinal fusion with Luque-rod segmental instrumentation. *J Bone Joint Surg Am* 1988; **70**: 41–4.

41 Modi HN, Hong JY, Mehta SS, Srinivasalu S, Suh SW, Yi JW, Yang JH, Song HR. Surgical correction and fusion using posterior-only pedicle screw construct for neuropathic scoliosis in patients with cerebral palsy: a three-year follow-up study. *Spine* 2009; **34**: 1167–75.

42 Rinsky LA. Surgery of spinal deformity in cerebral palsy. Twelve years in the evolution of scoliosis management. *Clin Orthop Relat Res* 1990; **253**: 100–109.

43 Hoffer MM, Feiwell E, Perry R, Perry J, Bonnett C. Functional ambulation in patients with melomeningocele. *J Bone Joint Surg Am* 1973; **55**: 137–48.

44 Comstock CP, Leach J, Wenger DR. Scoliosis in total-body-involvement cerebral palsy. Analysis of surgical treatment and patient and caregiver satisfaction. *Spine* 1998; **23**: 1412–24.

45 Auerbach JD, Spiegel DA, Zgonis MH, Reddy SC, Drummond DS, Dormans JP, Flynn JM. The correction of pelvic obliquity in patients with cerebral palsy and neuromuscular scoliosis: is there a benefit of anterior release prior to posterior spinal arthrodesis? *Spine* 2009; **34**: E766–74.

46 Benson ER, Thomson JD, Smith BG, Banta JV. Results and morbidity in a consecutive series of patients undergoing spinal fusion for neuromuscular scoliosis. *Spine* 1998; **23**: 2308–18.

47 Caird MS, Palanca AA, Garton H, Hensinger RN, Ayyangar RN, Drongowski A, Farley FA. Outcomes of posterior spinal fusion and instrumentation in patients with continuous intrathecal baclofen infusion pumps. *Spine* 2008; **33**: E94–9.

48 Fuhrhop SK, Keeler KA, Oto M, Miller F, Dabney KW, Bridwell KH, Lenke LG, Luhmann SJ. Surgical treatment of scoliosis in non-ambulatory spastic quadriplegic cerebral palsy patients: A matched cohort comparison of unit rod technique and all-pedicle screw constructs. *Spine Deform* 2013; **1**: 389–94.

49 Gau YL, Lonstein JE, Winter RB, Koop S, Denis F. Luque-Galveston procedure for correction and stabilization of neuromuscular scoliosis and pelvic obliquity: a review of 68 patients. *J Spinal Disord* 1991; **4**: 399–410.

50 Keeler KA, Lenke LG, Good CR, Bridwell KH, Sides B, Luhmann SJ. Spinal fusion for spastic neuromuscular scoliosis: is anterior releasing necessary when intraoperative halo-femoral traction is used? *Spine* 2010; **35**: E427–33.

51 McElroy MJ, Sponseller PD, Dattilo JR, Thompson GH, Akbarnia BA, Shah SA, Snyder BD. Growing rods for the treatment of scoliosis in children with cerebral palsy: a critical assessment. *Spine* 2012; **37**: E1504–E10.

52 Montgomery DM, Aronson DD, Lee CL, LaMont RL. Posterior spinal fusion: allograft versus autograft bone. *J Spinal Disord* 1990; **3**: 370–5.

53 Sink EL, Newton PO, Mubarak SJ, Wenger DR. Maintenance of sagittal plane alignment after surgical correction of spinal deformity in patients with cerebral palsy. *Spine* 2003; **28**: 1396–403.

54 Swank SM, Cohen DS, Brown JC. Spine fusion in cerebral palsy with L-rod segmental spinal instrumentation. A comparison of single and two-stage combined approach with Zielke instrumentation. *Spine* 1989; **14**: 750–9.

55 Tsirikos AI, Chang WN, Dabney KW, Miller F. Comparison of one-stage versus two-stage anteroposterior spinal fusion in pediatric patients with cerebral palsy and neuromuscular scoliosis. *Spine* 2003; **28**: 1300–5.

56 Vialle R, Delecourt C, Morin C. Surgical treatment of scoliosis with pelvic obliquity in cerebral palsy: the influence of intraoperative traction. *Spine* 2006; **31**: 1461–6.

57 Cobb JR. Outline for the study of scoliosis. Ann Arbor: JW Edwards, 1948.

58 Cahill PJ, Warnick DE, Lee MJ, Gaughan J, Vogel LE, Hammerberg KW, Sturm PF. Infection after spinal fusion for pediatric spinal deformity: thirty years of experience at a single institution. *Spine* 2010; **35**: 1211–7.

59 Duckworth AD, Mitchell MJ, Tsirikos AI. Incidence and risk factors for post-operative complications after scoliosis surgery in patients with Duchenne muscular dystrophy : a comparison with other neuromuscular conditions. *Bone Joint J* 2014; **96B**: 943–9.

60 Jevsevar DS, Karlin LI. The relationship between preoperative nutritional status and complications after an operation for scoliosis in patients who have cerebral palsy. *J Bone Joint Surg Am* 1993; **75**: 880–84.

61 Lonstein JE, Koop SE, Novachek TF, Perra JH. Results and complications after spinal fusion for neuromuscular scoliosis in cerebral palsy and static encephalopathy using luque galveston instrumentation: experience in 93 patients. *Spine* 2012; **37**: 583–91.

62 Sarwahi V, Sarwark JF, Schafer MF, Backer C, Lee M, King ECB, et al. Standards in anterior spine surgery in pediatric patients with neuromuscular scoliosis. *J Pediatr Orthop* 2001; **21**: 756–60.

63 Szoke G, Lipton G, Miller F, Dabney K. Wound infection after spinal fusion in children with cerebral palsy. *J Pediatr Orthop* 1998; **18**: 727–33.

64 Lipton GE, Miller F, Dabney KW, Altiok H, Bachrach SJ. Factors predicting postoperative complications following spinal fusions in children with cerebral palsy. *J Spinal Disord* 1999; **12**: 197–205.

65 Borkhuu B, Nagaraju D, Miller F, Moamed Ali MH, Pressel D, Adelizzi-Delany J, Miccolis M, Dabney K, Holmes L, Jr. Prevalence and risk factors in postoperative pancreatitis after spine fusion in patients with cerebral palsy. *J Pediatr Orthop* 2009; **29**: 256–62.

66 Mohamed Ali MH, Koutharawu DN, Miller F, Dabney K, Gabos P, Shah S, Holmes L, Jr. Operative and clinical markers of deep wound infection after spine fusion in children with cerebral palsy. *J Pediatr Orthop* 2010; **30**: 851–7.

67 Sewell MD, Malagelada F, Wallace C, Gibson A, Noordeen H, Tucker S, Molloy S, Lehovsky J. Does spinal fusion and scoliosis coorection improve activity and particiaption for children with GMFCS level IV and V cerebral palsy? *Medicine* 2015; **94**: 1–8.

68 Sewell MD, Malagelada F, Wallace C, Gibson A, Noordeen H, Tucker S, Molloy S, Lehovsky J. A preliminary study to assesses whether spinal fusion for scoliosis

improves carer-assessed quality of life for children with GMFCS level IV or V cerebral palsy. *J Pediatr Orthop* 2016; **36**: 299–304.

69 Abousamra O, Nishnianidze T, Rogers KJ, Bayhan IA, Yorgova P, Shah SA. Correction of pelvic obliquity after spinopelvic fixation in children with cerebral palsy: a comparision study with minimum two-year follow-up. *Spine Deform* 2016; **4**: 217–24.

70 Nishnianidze T, Bayhan IA, Abousamra O, Sees J, Rogers KJ, Dabney KW, Miller F. Factors predicting postoperative complications following spinal fusions in children with cerebral palsy scoliosis. *Eur Spine J* 2016; **25**: 627–34.

71 Dindo D, Demartines N, Clavien PA. Classification of surgical complications: a new proposal with evaluation in a cohort of 6336 patients and results of a survey. *Ann Surg* 2004; **240**: 205–13.

72 Funk S, Lovejoy S, Mencio G, Martus J. Rigid instrumentation for neuromuscular scoliosis improves deformity correction without increasing complications. *Spine* 2016; **41**: 46–52.

73 Majd ME, Muldowny DS, Holt RT. Natural history of scoliosis in the insitutionalized adult cerebral palsy population. *Spine* 1997; **22**: 1461–6.

74 Saito N, Ebara S, Ohotsuka K, Kumeta H, Takaoka K. Natural history of scoliosis in spastic cerebral palsy. *Lancet* 1998; **351**: 1687–92.

Table I: GRADE summary of findings: outcomes of spinal surgery for children with scoliosis and cerebral palsy

Outcome	No. of participants (studies)	Effect size	Quality of evidence components								Overall GRADE	Com
			Reasons to downgrade					Reasons to upgrade				
			Study limitations	Consistency	Directness	Precision	Publication bias	Magnitude of effect	Dose response gradient	Bias would reduce effect		
Overall function	990 (12)	Unable to determine	Serious (–1)	No important inconsistency	Some indirectness (–1)	Imprecision (–1)	Unlikely	Unable to determine	Not apparent	Not apparent	Very low	In tw with ques trend chan func
QoL	194 (3)	Unable to determine	Serious (–1)	No important inconsistency	Some indirectness (–1)	Imprecision (–1)	Unlikely	Unable to determine	Not apparent	Not apparent	Very low	Impr when with and ques
Gross motor function	294 (8)	Unable to determine	Serious (–1)	No important inconsistency	Some indirectness (–1)	Imprecision (–1)	Unlikely	Unable to determine	Not apparent	Not apparent	Very low	Mixe impr two mini chan rema

Parent or caregiver related	775 (10)	Unable to determine	Serious (–1)	Some in inconsistency	Some indirectness (–1)	Imprecision (–1)	Unlikely	Unable to determine	Not apparent	Not apparent	Very low	In st repo satis outc posit repo care mixe
% CC	Short term 805 (20)	Mean 59% Range 36%–77%	Serious (–1)	No important inconsistency	No important indirectness	No important imprecision	Unlikely	Strong evidence of effect (+1)	Not apparent	Not apparent	Low	All s impr and l curv
	Longer term 1070 (14)	Mean 61.4%, Range 55%–78%										
POC	Short term 675 (12)	Mean 65.5% Range 48.2%–82.8 %	Serious (–1)	No important inconsistency	Some indirectness (–1)	No important imprecision	Unlikely	Strong evidence of effect (+1)	Not apparent	Not apparent	Very low	All s impr and l pelvi corre
	Longer term 218 (7)	Mean 55.3% Range										

		43.1%–81%										
Overall postoperative complication rate	506 (12)	Average 1:3 odds of having at least one complication. Mean proportion 38.1% (95% CI 27.3% to 53.3%).	Serious (–1)	No important inconsistency	Some indirectness (–1)	No important imprecision	Unlikely	Strong evidence of effect (+1)	Not apparent	Not apparent	Very low	Mixe over comp in re studi

QoL, quality of life; CC, curve correction; POC, pelvic obliquity correction; CI, confidence interval.