Spinal column shortening for tethered cord syndrome: a systematic review and individual patient data meta-analysis

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OBJECTIVE Tethered cord release (TCR) is the gold standard treatment for tethered cord syndrome (TCS); however, there are significant shortcomings including high rates of retethering, especially in complex and recurrent cases. Spinal column shortening (SCS) is an alternative treatment for TCS intended to avoid these shortcomings. Early studies were limited to case reports and smaller case series; however, in recent years, larger case series and small cohort studies have been conducted. Given the increase in available data, a repeat systematic review and meta-analysis is warranted to assess the safety and efficacy of SCS for TCS.

METHODS The authors conducted a systematic review using MEDLINE (OVID), Embase (Elsevier), and Web of Science records dating from 1944 to July 2021 to identify all articles investigating SCS for TCS. They performed standard and individual patient data (IPD) meta-analyses, with 2 independent reviewers using PRISMA-IPD guidelines. Primary outcomes were improvement of preoperative clinical symptoms of pain, motor weakness, and bladder and bowel dysfunction, and also surgical complication rate. Secondary outcomes included urodynamic improvement and health-related quality-of-life outcomes determined using patient-reported outcome tools. Individual study quality assessment was performed using a standardized assessment tool for case reports/series, and publication bias was assessed using funnel plot analyses.

RESULTS The review yielded 15 studies with 191 cases of TCS treated with SCS. IPD were available in 11 studies with 89 cases. The average age at time of surgery was 28.0 years (range 5–76 years). The average follow-up time was 33.2 months (range 7–132 months). Improvement was observed at last follow-up in 60 of 70 (85.7%) patients with preoperative pain, in 38 of 60 (60.3%) patients with preoperative weakness, and in 36 of 76 (47.4%) patients with preoperative bladder or bowel dysfunction. Complications of CSF leak, new neurological deficit, wound infection, or reoperation occurred in 4 of 89 (4.5%) patients.

CONCLUSIONS SCS may be considered a safe and efficacious treatment option for TCS in children and adults (level C evidence; class IIb recommendation), especially for recurrent and complex cases. Current evidence is likely to be affected by selection and publication bias. Prospective comparative studies of SCS and TCR for TCS are recommended to determine long-term duration of outcomes, long-term safety in skeletally immature children, and exact indications of SCS versus traditional TCR.

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KEYWORDS lipomyelomeningocele; myelomeningocele; spina bifida; spinal column shortening; tethered cord syndrome; spine
Tethered cord syndrome (TCS) is defined as the clinical and radiographic manifestation of abnormal attachment of the spinal cord in the thoracic, lumbar, or lumbosacral region, leading to progressive loss of neurological function. Excessive longitudinal stretch on the spinal cord caused by abnormal cord attachments leads to decreased blood supply, altered metabolism, and mechanical damage to neurons of the spinal cord. This damage leads to a constellation of neurological symptoms including back pain, perineal pain, leg pain, leg weakness, sensory dysfunction, sexual dysfunction, and bladder and bowel dysfunction. Additionally, musculoskeletal changes such as scoliosis and foot deformities often occur.

There are several underlying pathologies that may lead to abnormal tethering. Spinal cord tumors, elongated cord, filum terminale thickening, myelomeningocele, lipomyelocele, other spinal dysraphisms, arachnoiditis, and scar tissue from prior neurosurgical procedures are the most frequent etiologies that lead to pathological tethering of the cord to surrounding structures.

Tethered cord release (TCR) is the gold standard treatment for TCS. TCR involves manual resection of the abnormal attachments from the spinal cord to alleviate the excessive tension. TCR has demonstrated excellent outcomes with clinical improvements in pain, motor weakness, and sensory impairment. TCR is also effective in improving bladder and bowel dysfunction, but to a lesser extent. Although it is effective at relieving clinical symptoms, TCR has significant shortcomings, such as risk of spinal cord and nerve root injury, CSF leak, and high rates of retethering due to intradural scar formation. These risks and complications are especially prevalent in recurrent and complex cases such as those due to myelomeningocele and lipomyelocele.

To avoid these shortcomings of TCR, particularly in complex and recurrent cases, spinal column shortening (SCS) has been proposed as an alternative treatment option. SCS involves intentionally reducing spinal column height to indirectly alleviate tension on the spinal cord. Various techniques have been used to shorten the spinal column, including pedicle subtraction osteotomy (PSO), vertebral column subtraction (VCS), and homogeneous spinal-shortening axial decompression (HSAD). All these techniques reduce tension on the spinal cord without requiring entry into the dura mater or direct manipulation of neural elements.

With increased interest and available data in the literature in recent years, there is need for a thorough and robust investigation of the safety and clinical efficacy of SCS in both children and adults. Here we present an updated systematic review of the literature as of July 2021. The primary search term was the MeSH of “Tethered Cord Syndrome,” and secondary terms included the following: AND spinal column shortening OR vertebral column resection OR pedicle subtraction osteotomy OR homogeneous spinal-shortening axial decompression. Additionally, the reference lists from prior review articles were searched to identify any additional articles.

Eligibility Assessment and Data Collection

Assessment for eligibility was performed by two independent reviewers (L.G.M. and M.C.A.) in a standardized manner. Any disagreements between reviewer assessments were resolved by consensus or by consulting a senior reviewer (A.J.). Both reviewers (L.G.M. and M.C.A.) performed data collection using a standardized data extraction template created specifically for this review. Each reviewer then checked the extracted data from the other reviewer. Corresponding authors were contacted via email to obtain any unreported data points. If no response was received, the unreported data were termed NR (not reported). If a response was received and the corresponding...
author confirmed that the data point was not available, it was termed NA (not available).

**Data Recorded**

Data items recorded included the following: age; sex; country; indication for surgery; number of prior untethering procedures; surgical procedure performed; spinal levels operated; size of osteotomy/magnitude of shortening; estimated blood loss; operation duration; surgical complications (CSF leak, new neurological deficit, wound infection, retethering, hardware failure, proximal junctional kyphosis, reoperation, and death); and subjective preoperative symptoms of pain, weakness, and bladder and bowel dysfunction as well as improvement in symptoms at final follow-up. The Japanese Orthopaedic Association (JOA) motor score, visual analog scale (VAS) pain score, urodynamics, and any HRQOL outcomes determined using PROs were collected when available.

**Methodology Quality Assessment**

The system described by Murad et al. in their 2018 article was used to perform the methodological quality assessment. This system was specifically designed to assess case series and case studies. For our review, we used the system to grade the articles as high, medium, or low quality. Questions 4–6 regarding causality were not included because they relate to adverse drug events, which was not applicable to our review. For length of follow-up, postoperative assessments at more than 1 year for patients ≥ 18 years of age were considered high quality and postoperative assessments at more than 3 years for patients < 18 years of age were considered high quality.

**Quantitative Analysis**

For traditional meta-analysis, we calculated rate estimates with the variance-stabilizing, double-arcsine transformation method. We used an inverse-variance weighting method in both fixed and random-effects models. The Clopper-Pearson interval (e.g., exact binomial interval method) was used to calculate the 95% confidence intervals. Heterogeneity between studies was estimated using Cochran’s Q with reported p values, between-study variance τ², and the I² statistic. We report the results from the random-effects models. Studies with zero cases or missing values were excluded from the summary results. R package “meta” was used for the meta-analysis modeling process (R Foundation for Statistical Computing).

IPD were used to perform quantitative analysis for frequency, mean, and confidence intervals when applicable for each outcome variable. IPD meta-analysis statistical testing was performed using Microsoft Excel (Microsoft Corp.). For dichotomous variables, proportions were calculated for IPD assessment by using the combined frequencies. Additionally, subgroup analyses were performed for the major outcomes by age group, sex, number of prior untethering procedures, and length of follow-up. Chi-square statistical testing was used to assess for significant differences between subgroups in the outcomes of interest (alpha = 0.05). To perform heterogeneity testing and bias risk assessment, the results were pooled across studies in a random-effects proportion meta-analysis for the major outcomes, using the DerSimonian and Laird model when possible. Cochran’s Q statistic to determine I² values was used to perform heterogeneity testing. For outcomes measured with continuous variables, mean, standard deviation, standard error, and confidence intervals were calculated when applicable.

**Bias Risk Assessment**

Assessment of publication bias was done using funnel plot analyses, with the x-axis containing proportion and the y-axis containing standard error, as well as being discussed in narrative form. A statistician (S.C.C.) was consulted to assist with the meta-analysis, subgroup analyses, heterogeneity testing, and bias risk assessment.

**Recommendations**

The GRADE (Grading of Recommendations, Assessment, Development and Evaluation) system was used to create recommendations.

**Results**

**Literature Search**

Our systematic search of OVID (n = 97), Web of Science (n = 57), Embase (n = 45), and cross-referencing from prior review reference lists (n = 6) produced 205 records (PRISMA-IPD flow diagram, Fig. 1)—98 records remained after removal of duplicates. Titles and abstracts were screened by two independent reviewers to produce 34 remaining articles for full-text review. Full-text assessment yielded 15 articles that fully met criteria for inclusion, and IPD were then collected. Ten articles originally provided IPD, whereas 5 articles provided only aggregate data. From 5 requests made via email to corresponding authors, 1 response was received providing additional data.

In total, the 15 included articles included 191 patients (Table 1). Of the 15 included studies, 7 were from Japan (n = 25 total patients), 5 were from the United States (n = 67), and 3 were from China (n = 99). Ten studies used vertebral column resection (VCR) (n = 83 total patients), 3 used HSAD (n = 99), and 2 used PSO (n = 9). Subjective changes in symptoms of pain and weakness were reported in 13 of the 15 included studies, and 14 studies reported subjective changes in bladder/bowel dysfunction. Five studies included perioperative urodynamic analysis. JOA motor score assessments were performed in 5 studies. The VAS pain scale was used in 4 studies. For PROs, 2 studies used the Oswestry Disability Index, 1 study used the Scoliosis Research Society Health Related Quality of Life Questionnaire (i.e., SRS-22), and 1 study used the Pediatric Quality of Life Inventory. Eleven articles noted the use of intraoperative neurophysiological monitoring, whereas 4 articles did not specify whether monitoring was used. No articles included detailed neurophysiological monitoring data, and there were no specific episodes of monitoring changes reported.

The 11 articles including IPD described 89 cases of SCS for TCS. There were 4 case reports (1–2 cases), 2 retrospective case series (at least 3 consecutive cases), and 9 prospective case series (at least 3 consecutive cases).
16,23,25,27,30–32 and 2 two-armed retrospective cohort studies,26,29 Of note, none of the 3 studies using HSAD were able to be included in IPD analysis. Hou et al.33 displayed some IPD; however, due to missing key data points such as individual follow-up lengths and data inconsistencies with regard to IPD, it was not included in the IPD analysis. Complete listing of cases and obtained IPD outcomes can be found in Supplemental Table 1.

**Standard Meta-Analysis Primary Outcomes and Secondary Outcomes**

Aggregate data of all included studies are displayed in Table 2. The estimated improvement rates of preoperative pain, weakness, and bowel and bladder dysfunction as well as the estimated major complication rate (CSF leak, new neurological deficit, wound infection, hardware failure, proximal junctional kyphosis, reoperation, and death) are displayed in Fig. 2. Due to limited data and significant heterogeneity in outcome reporting between studies, a formal meta-analysis was not performed for any of the secondary outcomes, including JOA motor score, urodynamics, and any HRQOL outcomes determined using PROs.

**IPD Meta-Analysis Primary and Secondary Outcomes**

Primary outcomes using IPD are displayed in Table 3. Sex was reported in all 89 cases (41 males, 48 females). Age was reported in all 89 cases, with a mean of 28.0 years (range 5–76 years). TCS etiology was reported in 81 of 89 cases and included myelomeningocele (n = 21), lipomeningocele (n = 3), lipomyelomeningocele (n = 42), spinal lipoma (n = 9), transitional spinal lipoma (n = 4), TCS unspecified (n = 1), and TCS without occupying lesion (n = 1). Number of prior untethering procedures was reported in all 89 cases, with a mean of 1.8 prior TCRs (range 0–17). Length of follow-up was included in all cases, with a mean of 33.2 months (range 7–132 months). Level of osteotomy was reported in all 89 cases and included T6 (n = 1), T9 (n = 1), T10 (n = 6), T11 (n = 6), T12 (n = 39), L1 (n = 34), L3 (n = 1), and L4 (n = 1). Size of osteotomy was reported in 73 cases, with a mean of 18.4 mm (range 9–28 mm). Estimated blood loss was reported in 45 cases, with a mean of 631.3 ml (range 100–1700 ml), and operation duration was reported in 45 cases, with a mean of 289.2 minutes (range 138–330 minutes).

Although not included as a primary outcome, it was noted that sexual dysfunction was reported as a symptom in 3 of the 15 included articles.15,29,32 Within these 3 articles listing sexual dysfunction, 3 total patients reported sexual dysfunction as a preoperative symptom. For 2 patients sexual function improved postoperatively, and for 1 patient it was not specified whether symptoms improved. There were major complications reported in 4 of 89 (4.5%) cases from articles that reported IPD; however, in only one of these complications was it specified which patient in the series had the complication. These complications included 2 cases of hardware failure requiring revision surgery, 1 case of wound infection, and 1 case of new sensory abnormality (L1 numbness corresponding to level of osteotomy).

Of note, there were no cases of retethering reported in any of the articles found in our review. There was no statistically significant difference in outcomes across age groups, sex, number of prior untethering procedures, or length of follow-up for any of the primary outcomes (Table 4).
and efficacy of SCS for the treatment of TCS. Improvement in preoperative weakness has been reported to range from 48% to 63%, and improvement in bowel and bladder dysfunction has been reported to range from 14% to 56% of patients. Of note, recovery rates of pain, weakness, and bladder dysfunction after a traditional TCR have been found to be reduced in complex pathologies of TCS such as myelomeningocele and lipomyelomeningocele, as well as in recurrent cases. Based on these literature findings, SCS appears to provide similar results with regard to improvement in preoperative symptoms.

TCR has an estimated complication rate of 4%–17% for CSF leak or new neurological deficit when used to treat TCS following at least 1 prior TCR. Additionally, retethering rates have been reported to range from 3% to 32% after untethering of myelomeningocele, and from 5% to 50% after untethering of lipomyelomeningocele. Based on our review, despite longer operative times and increased average blood loss compared to TCR, SCS does not appear to present additional risk of major complications for these complex and recurrent cases. There were no reported cases of retethering in our review, indicating that SCS may be a more durable treatment for cases that are at high risk for retethering. However, there were 2 cases of severe instrumentation failure that required reoperation, which highlights one of the additional complication risks associated with SCS compared to TCR. Aldave et al. previously described 1 case of catastrophic instrumenta-

### Quality Assessment, Risk Ratios, Heterogeneity, and Risk of Bias Assessment

Of the included articles, 4 were graded as having high methodological quality, 10 were graded as medium quality, and 1 was graded as low quality (Supplemental Table 2). Funnel plot analyses for risk of bias are shown in Supplemental Fig. 1.

### Discussion

The objective of this study was to investigate the safety and efficacy of SCS for the treatment of TCS. TCR remains the gold standard surgical treatment for TCS; however, in recent years there has been increased interest in SCS as an alternative treatment that circumvents many of the drawbacks of TCR procedures, especially in recurrent and complex cases. Due to the absence of comparative cohort studies and randomized controlled trials, it is difficult to make direct comparisons of the effectiveness of SCS versus TCR, and thus it is challenging to make strong recommendations. To date, Nakashima et al. and Zhang et al. are the only comparative cohort studies of SCS and TCR, with limited sample sizes of 3 and 8 patients treated with SCS, respectively. Although a direct comparison is limited due to few comparative studies, current literature estimates an improvement in preoperative back/leg pain in roughly 75%–83% of patients with TCS treated with TCR. Improvement in preoperative weakness has
tion failure and proximal joint kyphosis in detail.\textsuperscript{15} Revision surgery including extension of spinal instrumentation resulted in adequate repair and resolution of all preoperative symptoms at 31 months of follow-up. Longer follow-up is needed to understand the natural history of retethering after SCS. Authors have reported retethering to occur at intervals ranging from 1 to 9 years following traditional TCR, most commonly manifesting 5 years after TCR.\textsuperscript{12,38} Given the long intervals for retethering, this complication cannot be ruled out based on the limited follow-up times in many of the articles in this review. With questions remaining with regard to long-term risks and complications of SCS, it should be used with caution in more straightforward cases in which TCR is likely to provide adequate results with limited risk of complication.

Secondary outcomes of formal urodynamics and PROs were used in multiple studies in our review. However, differences in methodology and measurement tools made a meta-analysis unfeasible. Hou et al.\textsuperscript{33} and Wang et al.\textsuperscript{16,27} demonstrated significant improvement in urodynamics using HSAD, whereas Hsieh et al.\textsuperscript{32} and our previous series\textsuperscript{31} demonstrated improvement in formal urodynamics at a rate that was comparable to symptomatic improvement in urological dysfunction. Additionally, included studies demonstrated statistically significant quality-of-life improvements using tools such as the Scoliosis Research Society Health Related Quality of Life Questionnaire, the Oswestry Disability Index, and the Pediatric Quality of Life Inventory (Table 2).\textsuperscript{15,30,31} These results demonstrate more objective evidence of the efficacy of SCS.

There was statistically significant heterogeneity in the primary outcomes of subjective improvement of preoperative weakness and bowel/bladder dysfunction. Additionally, there is nonstatistically significant heterogeneity in outcomes of pain improvement and complication rate. This is not unexpected, given that there are observable differences between studies that make generalizability difficult. Management of TCS is highly dependent on the etiology and classification of spinal dysraphism.\textsuperscript{39} A majority of cases in this review are due to complex etiologies.

### Table 2. Included study aggregate data

<table>
<thead>
<tr>
<th>Authors &amp; Year</th>
<th>No. of Prior Untethering Ops (range)</th>
<th>FU, Mos (range)</th>
<th>Level of Resection (no.)</th>
<th>Size of Osteotomy, mm (range)</th>
<th>EBL, ml (range)</th>
<th>Op Duration, mins (range)</th>
<th>Complications</th>
</tr>
</thead>
<tbody>
<tr>
<td>Kanno et al., 2008\textsuperscript{22}</td>
<td>0</td>
<td>24</td>
<td>T12 (1)</td>
<td>22</td>
<td>NR</td>
<td>NR</td>
<td>None</td>
</tr>
<tr>
<td>Miyakoshi et al., 2009\textsuperscript{23}</td>
<td>0</td>
<td>48 (36–60)</td>
<td>L1 (3)</td>
<td>20</td>
<td>NR</td>
<td>NR</td>
<td>None</td>
</tr>
<tr>
<td>Hsieh et al., 2009\textsuperscript{32}</td>
<td>4.5 (4–5)</td>
<td>16.5 (15–18)</td>
<td>T12 (1), L1 (1)</td>
<td>18.5 (17–20)</td>
<td>NR</td>
<td>NR</td>
<td>None</td>
</tr>
<tr>
<td>Matsumoto et al., 2009\textsuperscript{24}</td>
<td>1</td>
<td>20</td>
<td>T9 (1)</td>
<td>21</td>
<td>540</td>
<td>366</td>
<td>None</td>
</tr>
<tr>
<td>Kawamura et al., 2010\textsuperscript{25}</td>
<td>0</td>
<td>21 (18–24)</td>
<td>L3 (1), L4 (1)</td>
<td>14</td>
<td>NR</td>
<td>NR</td>
<td>None</td>
</tr>
<tr>
<td>Kokubun et al., 2011\textsuperscript{15}</td>
<td>0.4 (0–1)</td>
<td>74.6 (30–132)</td>
<td>T12 (2), L1 (6)</td>
<td>21.0 (19–23)</td>
<td>NR</td>
<td>NR</td>
<td>Aggressive neurological deterioration (1)</td>
</tr>
<tr>
<td>Nakashima et al., 2016\textsuperscript{26}</td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td></td>
<td>Intraop bleeding &gt;3000 ml (1)</td>
</tr>
<tr>
<td>Aldave et al., 2017\textsuperscript{15}</td>
<td>1 (0–3)</td>
<td>30.8 (27–37)</td>
<td>T6 (1), T11 (1), L1 (5)</td>
<td>15</td>
<td>578.6 (250–900)</td>
<td>401.1 (270–519)</td>
<td>Instrument failure requiring reop (1)</td>
</tr>
<tr>
<td>Hou et al., 2018\textsuperscript{31}</td>
<td>0.3 (0–1)</td>
<td>21.5 (NR)</td>
<td>NR</td>
<td>17.2 (NR)</td>
<td>720.0 (NR)</td>
<td>265.6 (NR)</td>
<td>None</td>
</tr>
<tr>
<td>Wang et al., 2019\textsuperscript{17}</td>
<td>NR</td>
<td>31.2 (12–48)</td>
<td>L1–S1 (8), L2–S1 (2), L1–5 (2), L2–5 (6), other (2)</td>
<td>NR</td>
<td>744.4 (460–1155)</td>
<td>262.9 (192–330)</td>
<td>None</td>
</tr>
<tr>
<td>Ide et al., 2021\textsuperscript{28}</td>
<td>0.6 (0–1)</td>
<td>56.6 (12–108)</td>
<td>L1 (7)</td>
<td>16 (11–20)</td>
<td>827.9 (411–1252)</td>
<td>328.4 (291–414)</td>
<td>None</td>
</tr>
<tr>
<td>Zhang et al., 2020\textsuperscript{29}</td>
<td>1.3 (1–3)</td>
<td>25.0 (7–43)</td>
<td>T10 (2), T11 (3), T12 (3)</td>
<td>NR</td>
<td>731 (NR)</td>
<td>358 (NR)</td>
<td>None</td>
</tr>
<tr>
<td>Theodore et al., 2021\textsuperscript{30}</td>
<td>3.7 (1–17)</td>
<td>23.3 (14–38)</td>
<td>T12 (16), L1 (4)</td>
<td>22.9 (9–28)</td>
<td>NR</td>
<td>NR</td>
<td>Duorotomy (1), wound infection (1), new neurological deficit (1), instrumentation failure (1)</td>
</tr>
<tr>
<td>McVeigh et al., 2021\textsuperscript{31}</td>
<td>1.9 (0–6)</td>
<td>27.3 (13–45)</td>
<td>T10 (4), T11 (2), T12 (16), L1 (8)</td>
<td>15</td>
<td>578.3 (100–1700)</td>
<td>248.7 (138–423)</td>
<td>Duorotomy (1), acute blood loss anemia (2)</td>
</tr>
</tbody>
</table>

EBL = estimated blood loss; FU = follow-up.
such as lipomyelomeningocele and secondary causes of TCS such as scar tissue formation from prior surgery (81 of 89 cases). However, there are several cases of primary TCS included such as spinal lipomas with no prior operations (8 of 89 cases). Furthermore, surgical technique varied significantly between studies. Given the high level of specialization of these techniques, results may vary based on individual technique and surgeon experience with the procedure. Whereas most studies used techniques involving an osteotomy (VCR or PSO), HSAD does not involve an osteotomy and instead resects multiple intravertebral discs to shorten the spinal column.33 Of note, no studies in which HSAD was used were able to be included in the IPD meta-analysis. Even among studies using VCR, the technique varied significantly between authors. For example, in our previous series we used a 3-column resection, with resection of 15 mm of the vertebral body, but this method left superior and inferior endplates intact and did not involve the intervertebral discs.31 However, other techniques used by Kokubun et al.,13 Hsieh et al.,32 and Theodore et al.30 involved resection of the vertebral body of interest as well as the adjacent intravertebral disc. Other differences that may have a significant impact on outcomes include size of spinal column reduction, number of prior TCRs, age of patient, and duration of symptoms before SCS. Unfortunately, this review is underpowered to distinguish the significance of these variables, and future research with larger sample sizes will be needed to optimize these factors. We performed subgroup analyses for age, sex, number of prior TCRs, and length of follow-up; however, no statistically significant differences were observed in any of the primary outcomes. Potential confounding factors previously mentioned along with small sample sizes are likely to contribute to the lack of statistical significance in these subgroups.

**SCS in Skeletally Immature Children**

TCS affects both children and adults; however, it is primarily a disease that manifests in childhood. For this reason, use of SCS in children is of particular interest. There are multiple concerns that arise with spinal fusion in a skeletally immature child. Continued skeletal growth of nonfused portions of the spinal column may increase the risk of reoccurrence of pathological cord tension. Previous studies have demonstrated spinal fusion in children to result in growth retardation of 1 mm per year of remaining growth per spinal level fused.40 Additionally, growth of the anterior and middle segments of the fused spinal seg-

### TABLE 3. Major outcomes: IPD summary results and heterogeneity testing

<table>
<thead>
<tr>
<th>Outcome</th>
<th>Frequency</th>
<th>Random-Effects Meta-Analysis (95% CI)</th>
<th>P</th>
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</thead>
<tbody>
<tr>
<td>Preop pain improvement</td>
<td>60/70 (85.7%)</td>
<td>81% (70–89)</td>
<td>0%</td>
</tr>
<tr>
<td>Preop weakness improvement</td>
<td>38/63 (60.3%)</td>
<td>61% (40–78)</td>
<td>38%</td>
</tr>
<tr>
<td>Preop B/B dysfunction improvement</td>
<td>36/76 (47.4%)</td>
<td>50% (38–61)</td>
<td>0%</td>
</tr>
</tbody>
</table>

B/B = bowel and/or bladder.
ments while fusion hardware secures the posterior column can result in a crankshaft deformity. For these reasons, it could be best to delay SCS until skeletal maturity when possible. Seven studies in this review included both pediatric and adult cases. On subgroup analysis there was not a significant difference in efficacy in skeletally immature children compared to adults. To date, there has not been a reported case of retethering or other serious complication associated with SCS due to continued skeletal growth, although follow-up length has been limited. In practice, treatment should be approached on an individual basis, weighing risks of SCS with severity of current symptoms, risks associated with additional TCR, and concerns for permanent damage if treatment is delayed.

**Limitations**

The primarily positive outcomes reported by studies in this review are probably impacted by selection and publication bias, and this potentially underestimates the rate of poor outcomes in the true population. All studies in our review were retrospective in nature, and there are very few cohort studies, thus adding to the potential for publication bias. Furthermore, the primary outcomes of most included studies involved subjective symptomatic improvement measured via history and physical examination and did not use standardized assessment tools for primary outcomes. This form of outcome measurement in combination with the retrospective nature of these studies increases likelihood of bias. Many studies did use objective measures (urodynamics, VAS pain scales, PROs); however, these testing methods were not able to be included in formal meta-analysis. Additionally, our previous series provided more than one-third of the pooled patients for the IPD meta-analysis. As a result, the outcomes and limitations of our prior series probably have a large impact on the results of this analysis.

Length of follow-up was a significant limitation for this review. A few studies demonstrated longer-term follow-up; however, most of the studies included had an average follow-up length of fewer than 3 years. Durability of symptomatic improvement is essential to the effectiveness of SCS, and the primary complication of retethering is a process that occurs over many years. Therefore, long-term complications such as retethering cannot be completely ruled out based on the results of this review.

Although this review demonstrates an increasing level of evidence demonstrating safety and efficacy of SCS, many questions remain. Further studies are needed to explain inconsistencies in outcomes and to assess long-term durability of outcomes. Timing of surgery and precise indications for patient selection, including etiologies of TCS most suitable for SCS, still need to be determined. Particularly for children who have yet to reach skeletal maturity, timing of surgery is critically important. To answer these remaining questions, prospective comparative studies of SCS and TCR for TCS that include pediatric patients are essential.

**Recommendations**

This meta-analysis provides the highest level of evidence to date with regard to the efficacy and safety of SCS for the treatment of TCS. We conclude that SCS is safe and effective in improving symptoms of pain, weakness, and bowel and bladder dysfunction associated with TCS. Due to our reliance on primarily case reports, case series, and small cohort studies, the level of evidence (level C) col-

<table>
<thead>
<tr>
<th>TABLE 4. Major outcomes: subgroup analyses</th>
</tr>
</thead>
<tbody>
<tr>
<td>Characteristic</td>
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<tr>
<td>----------------</td>
</tr>
<tr>
<td>Age</td>
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<td>Statistical analysis</td>
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<tr>
<td>Sex</td>
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<tr>
<td>Statistical analysis</td>
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lected in this meta-analysis would support a weak recommendation (class IIb) for the use of SCS for the treatment of TCS based on the GRADE guidelines.21 We believe that SCS may have particular utility in treating complex and recurrent cases of secondary TCS. However, this review cannot make a recommendation with regard to direct comparison of SCS to TCR for secondary TCS. With regard to use of SCS in children, the current evidence indicates that SCS has similar efficacy and safety in children and adults—however, with relatively short follow-up there remains an increased risk associated with spinal fusion in skeletally immature patients. For these reasons, we do not advocate for widespread use of SCS, but rather cautious application under a research protocol, particularly in skeletally immature children.

Conclusions

SCS is a safe and efficacious treatment option (class IIb) for TCS in children and adults, particularly for complex and recurrent cases. Prospective comparative studies of SCS and TCR for TCS are recommended to determine duration of outcomes, long-term safety in skeletally immature children, and absolute and relative indications of SCS versus TCR.

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Disclosures
The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions
Conception and design: Jea, McVeigh. Acquisition of data: McVeigh, Anokwute, Chen. Analysis and interpretation of data: McVeigh, Anokwute, Chen. Drafting the article: all authors. Critically revising the article: all authors. Reviewed submitted version of manuscript: all authors. Approved the final version of the manuscript on behalf of all authors: Jea. Statistical analysis: McVeigh, Anokwute, Chen. Administrative/technical/material support: McVeigh, Anokwute, Chen. Study supervision: Jea.

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Abstract Presentations
An abstract detailing these findings has been accepted for oral presentation at the 2022 AANS Annual Scientific Meeting taking place April 29–May 2, 2022, in Philadelphia, PA.

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