Nicholas Theodore, MD[‡] Ethan Cottrill, MS[‡]* Samuel Kalb, MD[§] Corinna Zygourakis, MD[‡] Bowen Jiang, MD[‡] Zach Pennington, BS [®][‡] Daniel Lubelski, MD[‡] Erick M. Westbroek, MD[‡] A. Karim Ahmed, BS[‡] Jeff Ehresman, BS[‡] Daniel M. Sciubba, MD[‡] Timothy F. Witham, MD[‡] Jay D. Turner, MD[§] Mari Groves, MD[‡] U. Kumar Kakarla, MD[§]

^{*}Department of Neurosurgery, The Johns Hopkins University School of Medicine, Baltimore, Maryland; ⁵Department of Neurosurgery, Barrow Neurological Institute, St. Joseph's Hospital and Medical Center, Phoenix, Arizona

*Nicholas Theodore and Ethan Cottrill contributed equally to this work.

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Correspondence:

Nicholas Theodore, MD, Department of Neurosurgery, The Johns Hopkins University School of Medicine, 600 N Wolfe St, Meyer 7-113, Baltimore, MD 21287, USA. Email: theodore@jhmi.edu Twitter: @DrNTheodore

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Posterior Vertebral Column Subtraction Osteotomy for Recurrent Tethered Cord Syndrome: A Multicenter, Retrospective Analysis

BACKGROUND: Few have explored the safety and efficacy of posterior vertebral column subtraction osteotomy (PVCSO) to treat tethered cord syndrome (TCS).

OBJECTIVE: To evaluate surgical outcomes after PVCSO in adults with TCS caused by lipomyelomeningocele, who had undergone a previous detethering procedure(s) that ultimately failed.

METHODS: This is a multicenter, retrospective analysis of a prospectively collected cohort. Patients were prospectively enrolled and treated with PVCSO at 2 institutions between January 1, 2011 and December 31, 2018. Inclusion criteria were age \geq 18 yr, TCS caused by lipomyelomeningocele, previous detethering surgery, and recurrent symptom progression of less than 2-yr duration. All patients undergoing surgery with a 1-yr minimum follow-up were evaluated.

RESULTS: A total of 20 patients (mean age: 36 yr; sex: 15F/5M) met inclusion criteria and were evaluated. At follow-up (mean: 23.3 ± 7.4 mo), symptomatic improvement/resolution was seen in 93% of patients with leg pain, 84% in back pain, 80% in sensory abnormalities, 80% in motor deficits, 55% in bowel incontinence, and 50% in urinary incontinence. Oswestry Disability Index improved from a preoperative mean of 57.7 to 36.6 at last follow-up (P < .01). Mean spinal column height reduction was 23.4 \pm 2.7 mm. Four complications occurred: intraoperative durotomy (no reoperation), wound infection, instrumentation failure requiring revision, and new sensory abnormality.

CONCLUSION: This is the largest study to date assessing the safety and efficacy of PVCSO in adults with TCS caused by lipomyelomeningocele and prior failed detethering. We found PVCSO to be an excellent extradural approach that may afford definitive treatment in this particularly challenging population.

KEY WORDS: Detethering, Lipomyelomeningocele, Posterior vertebral column subtraction osteotomy, Tethered cord syndrome

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ethered cord syndrome (TCS) is a constellation of clinical signs and symptoms resulting from stretching of the spinal cord between 2 fixation points.¹⁻³ Although TCS is often caused by spinal malformations, such as

ABBREVIATIONS: CSF, cerebrospinal fluid; ICU, intensive care unit; IRB, Institutional Review Board; MCID, minimum clinically important difference; ODI, Oswestry Disability Index; PVCSO, posterior vertebral column subtraction osteotomy; TCS, tethered cord syndrome

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myelomeningocele, thickened filum terminale, or lipomyelomeningocele,⁴ it can present de novo in adults—for example, after a traumatic event or following a pediatric pathology that was treated surgically.⁵⁻⁷ The current gold standard for treatment of TCS is a traditional intradural detethering procedure, in which arachnoid adhesions are lysed until the cord is visualized to fall freely from its attachments. This procedure often improves symptoms, especially pain.⁸⁻¹⁰ However, complete detethering is not always feasible,¹¹ and symptomatic retethering develops in 5% to 50% of patients over the long term.^{4,8,12-14} In addition, cerebrospinal fluid (CSF) leaks and wound infection occur in



up to 15% of patients who have undergone an intradural detethering procedure.¹⁵⁻¹⁷

In 1995, Kokubun¹⁸ first described a posterior vertebral column subtraction osteotomy (PVCSO) as a primary treatment for TCS in adult patients with a low-placed conus medullaris. In 2006, Grande et al¹⁹ demonstrated in a cadaveric model that the PVCSO procedure can significantly reduce tension on the spinal cord, nerve roots, and filum terminale by shortening the spinal column. This procedure, which involves removing approximately 90% of a thoracolumbar vertebra and subsequently reducing the spinal column, is conceptually illustrated in Figures 1-3.

Advantages and disadvantages to this approach have been described (Table 1), but a recent meta-analysis identified only 57 patients across 6 studies who had undergone PVCSO for TCS.²⁰

There is a paucity of evidence on the safety and efficacy of PVCSO for the treatment of recurrent TCS. No study has evaluated the role of PVCSO exclusively in adult patients with TCS caused by lipomyelomeningocele, in whom detethering surgery failed. In this multicenter, retrospective study, we sought to evaluate the outcomes of PVCSO in this patient population, and hypothesized that it is a safe and effective procedure with durable outcomes.



METHODS

Patient Population

This is a 2-center retrospective observational study of PVCSO for the treatment of adult TCS, designed to evaluate the outcomes of a well-described surgical procedure in a prospectively collected cohort. Standard detethering surgery was offered as an alternative to PVCSO and declined by all patients; Institutional Review Board [IRB]/ethics approval was obtained for this study prior to conducting this research. From January 2011 to December 2018, we prospectively enrolled and followed select adult patients who had undergone a detethering procedure for lipomyelomeningocele that failed and who also had a short duration of new progressive symptoms. The 2 centers in this study are Barrow Neurological Institute and The Johns Hopkins Hospital. Inclusion criteria were age \geq 18 yr, diagnosis of TCS caused by lipomyelomeningocele, history of failed detethering surgery, symptom progression <2 yr, and a 1-yr minimum postoperative follow-up. Exclusion criteria were age <18 yr, symptom progression >2 yr, no prior detethering procedure, less than 1 yr of follow-up, or the patient's inability to tolerate extensive spinal surgery. Inclusion was limited to symptom progression <2 yr because we believed that patients in a subacute, or earlier, stage of recurrent TCS would potentially benefit more from PVCSO than those in a more chronic stage of recurrence (>2 yr). Standard operative consent was obtained from all patients.

Data Collection

Changes in postoperative pain, neurological and functional status (clinical notes and Oswestry Disability Index [ODI] scores), radiographical outcomes (spinal column height reduction and changes in the local kyphotic Cobb angle), operative details (operative time, blood loss, intensive care unit [ICU] stay, and length of stay), and complication rates were recorded and analyzed. Radiological outcomes were evaluated by board-certified radiologists on our team. Spinal column height was measured from computed tomography or magnetic resonance imaging as the vertical distance from the superior endplate of the proximal vertebra (relative to the osteotomized vertebra). Local kyphotic Cobb angle was measured from the inferior endplate of the osteotomized vertebra to the superior endplate of the soteotomized vertebra. Complications were defined as durotomy, excessive intraoperative blood loss (>2 L), wound infection, new neurologic abnormality, instrumentation failure,



and pseudarthrosis. Changes in the ODI were assessed for statistical significance using a 2-tailed Mann-Whitney U test, with α set at 0.05.

Procedural Details

All PVCSO procedures were performed at T12 or L1, with fixation one or more levels above and below the osteotomized vertebra, depending on bone quality and intraoperative evaluation. Typical procedural details for a T12 resection are shown in Figures 1-3 and are fully described in **Supplemental Digital Content 1**.

RESULTS

In total, 20 patients met the inclusion/exclusion criteria and were included in this study. Patients' age, sex, body mass index, number of previous detethering procedures, presenting symptoms, levels of osteotomy and fixation, and length of followup are shown in Table 2. The mean age of these patients at time of surgery was 36 yr (range: 20-69 yr). A total of 15 patients were women; 5 were men. All patients had undergone at least 1 previous detethering procedure that provided temporary symptom relief. The mean number of previous detethering



procedures was 3.7 ± 4.3 (range: 1-17). In all patients, the vertebral osteotomy was performed at either T12 (80%) or L1 (20%); in 9 (45%), fixation was performed at 1 level above and below the osteotomy, and in the remainder, fixation was performed at more than 1 level above and below the osteotomy. Those undergoing fixation at 1 level above and below the osteotomy were also the earliest patients in our cohort. Anterior autograft was also placed in 2 patients early in our series. The mean operative time was 5.1 ± 0.7 h (range: 4.2-7.0 h), mean estimated blood loss was 1500 ± 300 mL (range: 1100-2000 mL), mean ICU stay was 2.4 ± 0.7 d (range: 2-4 d), and mean length of stay was 7.3 ± 2.4 d (range: 5-15 d). The mean length of follow-up was 23.3 ± 7.4 mo (range: 13.8-38.0 mo).

Symptomatic changes (bowel incontinence, motor weakness, back/leg pain, sensory abnormalities, and urinary incontinence) and functional changes (ODI) from the preoperative to last follow-up time points are shown in Tables 3 and 4, respectively. Figure 4 shows the aggregate symptomatic changes for all patients at the time of last follow-up (mean: 23.3 mo). Of the 19 patients who presented with back pain, 16 (84%) experienced either symptomatic improvement (11/16) or resolution (5/16) at the final follow-up. Of the 15 patients who presented with leg pain, 93% (14/15) experienced either symptomatic improvement (11/15) or resolution (3/15). A total of 8 (50%) of the 16 patients who presented with urinary incontinence experienced symptomatic improvement (5/8) or resolution (3/8), while 6 (55%) of the 11 patients who presented with bowel incontinence experienced symptomatic improvement (4/6) or resolution (2/6). Improvement in motor function was seen in 80% (8/10) of patients who presented with motor deficits. Of the 10 patients who presented with sensory abnormalities, 80% saw symptomatic improvement (6/10) or resolution (2/10). One patient developed L1-level numbness, corresponding to the osteotomized vertebra. However, additional imaging (magnetic resonance imaging and radiographs) of this patient showed excellent instrumentation position, solid fusion, and increased cross-sectional area of the spinal cord, suggesting tension relief. Furthermore, this patient reported overall satisfaction with surgery and has not undergone further operative treatment. No patient developed new bowel incontinence, motor deficits, back/leg pain, or urinary incontinence at last follow-up. Surgery-associated back pain was

TABLE 2. Characteristics of 20 Patients with TCS Who Were Included in the Study; the Etiology of TCS for All Patients Was Lipomyelomening ocele								
Pt	Age (yr)	Sex	BMI (kg/m ²)	Previous detetherings (no.)	Presenting symptoms ^a	Level of osteotomy	Levels of fixation	Length of follow-up (mo)
1	25	М	23.7	3	B, F, L, M, U	T12	T11-L1	33.8
2	29	F	37.9	2	B, F, L	T12	T11-L1	20.2
3	23	F	32.3	2	B, L, M, U	T12	T11-L1	30.9
4	48	F	25.8	1	B, F, D, U	L1	T11-L1	26.2
5	69	F	22.6	2	B, F, L, M, U	T12	T11-L1	32.9
6	27	F	26.0	1	B, F, M, S, U	T12	T11-L1	31.2
7	50	М	23.8	2	B, F, L, M, U	T12	T11-L1	19.7
8	27	М	35.0	1	B, S, U	T12	T11-L1	19.5
9	42	F	30.5	1	B, L, S, U	T12	T11-L1	14.6
10	48	М	29.5	3	L, U	T12	T10-L2	13.8
11	63	F	25.1	1	B, F, L, U	T12	T10-L2	21.9
12	23	F	38.0	2	B, L	T12	T10-L2	31.2
13	20	F	42.0	1	B, F, M, S	L1	T4-ilium	17.7
14	20	F	25.0	1	B, U	L1	T10-L3	38.0
15	28	F	24.0	12	B, L, M, S, U	T12	T10-L2	19.9
16	37	F	52.0	10	B, F, L, M, S, U	T12	T10-L2	25.1
17	49	М	29.0	1	B, L, M, S	L1	T9-L4	22.4
18	36	F	38.0	17	B, L, M, S, U	T12	T10-L2	16.4
19	34	F	28.7	6	B, F, L, S, U	T12	T10-L2	14.3
20	27	F	32.9	5	B, F, L, S, U	T12	T10-L2	15.9
Mean	36	15F:5M	31.1	3.7	B: 19 (95%) D: 1 (5%) F: 11 (55%) L: 15 (75%) M: 10 (50%) S: 10 (50%) U: 16 (80%)	T12: 16 (80%) L1: 4 (20%)	1 level above/below: 9 (45%) >1 level above/below: 11 (55%)	23.3

^a Symptoms: B, back pain; D, deformity (kyphotic); F, fecal incontinence; L, leg pain; M, motor deficit; S, sensory abnormalities; U, urinary incontinence. BMI, body mass index; M, male; F, female.

considered to be absent at last follow-up (minimum: ≥ 12 mo postoperatively).

Nineteen patients (95%) completed ODI forms both preoperatively and at the final follow-up. For these patients, ODI improved significantly postoperatively, from a mean of 57.7 preoperatively to 36.6 at last follow-up (P < .01; Table 4 and Figure 5). This improvement is considered clinically significant, surpassing the minimum clinically important difference (MCID) of 12.8 points on the ODI for patients undergoing lumbar spine surgery.²¹ Furthermore, all patients indicated overall satisfaction with their surgical treatment, and all indicated they would elect to undergo the PVCSO surgery again.

Spinal column height reduction and changes in the local kyphotic Cobb angle for all patients are shown in Table 5. The mean spinal column height reduction was 23.4 ± 2.7 mm (range: 18-28 mm). The mean local kyphotic Cobb angle changed slightly from a mean of 8.6° preoperatively to 5.9° postoperatively, suggesting maintenance of local spine alignment. Furthermore, all patients demonstrated excellent hardware position and radiologic fusion at the time of the final

follow-up. Representative pre- and postoperative magnetic resonance images are provided in Figure 6, which illustrates spinal column reduction and cord tension relief after PVCSO. The majority of our patients did not receive postoperative magnetic resonance imaging, which limits our ability to perform statistical analysis of changes pertaining to cord tension relief. However, representative intraoperative ultrasound videos illustrating cord tension relief immediately before and after PVCSO are provided (**Supplemental Digital Contents 2** and **3**).

A total of 4 patients experienced complications, including an intraoperative durotomy (not requiring reoperation), a wound infection requiring debridement, instrumentation failure requiring revision, and new sensory abnormality (L1-level numbness, corresponding to the osteotomized vertebra).

DISCUSSION

Key Results

The purpose of this study was to evaluate surgical outcomes after PVCSO in adult patients with TCS caused

TABLE 3. Symptomatic Changes in Patients at the Time of Last Follow-up ^a						
			Pa	ain		
Patient	Bowel	Motor	Back	Leg	Sensory	Urinary
1	Same	Improved	Resolved	Resolved	-	Same
2	Improved	-	Improved	Improved	-	-
3	-	Same	Resolved	Improved	-	Improved
4	Improved	-	Improved	-	-	Improved
5	Improved	Improved	Improved	Improved	-	Improved
6	Same	Improved	Same	-	Same	Same
7	Same	Improved	Same	Improved	-	Same
8	-	-	Improved	-	Improved	Resolved
9	-	-	Improved	Improved	Improved	Improved
10	-	-	-	Improved	-	Same
11	Resolved	-	Improved	Improved	-	Resolved
12	-	-	Resolved	Improved	-	-
13	Same	Improved	Improved	-	Improved	-
14	-	-	Improved	-	Worsened ^b	Same
15	-	Improved	Improved	Improved	Improved	Same
16	Improved	Improved	Improved	Improved	Improved	Improved
17	-	Improved	Improved	Improved	Improved	-
18	-	Same	Same	Same	Same	Same
19	Resolved	-	Resolved	Resolved	Resolved	Resolved
20	Same	-	Resolved	Resolved	Resolved	Same
Total						
Worsened	0	0	0	0	1 ^b	0
Same	5	2	3	1	2	8
Improved	4	8	11	11	6	5
Resolved	2	0	5	3	2	3

^aHyphen (-) indicates that the patient did not present with this symptom, and the symptom did not develop.

^bPatient did not present with symptom, but did develop the symptom postoperatively.

by lipomyelomeningocele, whose previous detethering surgery had failed. A total of 20 patients (mean age: 36 yr; mean number of previous detethering procedures: 3.7) were evaluated. At a mean follow-up of $\overline{23.3} \pm 7.4$ mo, symptomatic improvement/resolution was seen in 93% of those presenting with leg pain, 84% of those with back pain, 80% of those with sensory abnormalities, 80% of those with motor deficits, 55% of those with bowel incontinence, and 50% of those with urinary incontinence. ODI improved from a mean of 57.7 preoperatively to 36.6 at final follow-up (n = 19; P < .01). We considered this improvement to be clinically significant, surpassing the MCID of 12.8 points for patients undergoing lumbar spine surgery; however, given the large variability in MCID values, the use of other reference values could alter this assessment.²² Complications occurred in 4 patients: 1 each for intraoperative durotomy (not requiring reoperation), wound infection requiring debridement, instrumentation failure requiring revision instrumentation, and new sensory abnormality (L1-level numbness, corresponding to the osteotomized vertebra). In all cases, PVCSO was performed at T12 or L1, which we believe is the ideal location for TCS. The last dentate ligaments are at T12, and so shortening above T12 may not relieve tension on the cord; in contrast, PVCSO below L1 would put major lumbar nerve roots at risk for injury and would therefore increase the risk for morbidity.

Limitations

There are several limitations to this study. We report the results of a relatively small sample size (n = 20). This nonetheless adds substantially to the available literature in terms of patient number, especially for adult patients with TCS caused by lipomyelomeningocele in whom previous detethering procedure(s) has failed. Our study also has a lengthy follow-up period of close to 2 yr. The patients in this cohort will continue to be followed so that long-term data, including the durability of symptomatic changes, may be added to the literature. In addition, we limited inclusion to patients with symptom progression < 2 yr; it is possible that those with long-standing symptom progression (>2 yr) may also benefit from PVCSO but were not included in this study. Furthermore, we assessed symptomatic outcomes via history and physical exam and did not utilize standardized assessment scales for all outcomes (eg, urodynamic studies for urinary incontinence or anorectal manometry for bowel incontinence). We also did not investigate trends in symptomatic

TABLE 4. Changes in ODI					
	ODI				
Patient	Preoperative	Last follow-up	Change		
1	n/a	n/a	n/a		
2	66	42	-24		
3	87	66	-21		
4	46	36	—10		
5	46	33	—13		
6	52	40	-12		
7	50	38	-12		
8	60	55	—5		
9	90	12	-78		
10	54	44	-10		
11	58	10	-48		
12	50	50	0		
13	80	80	0		
14	20	0	-20		
15	50	20	-30		
16	40	23	-17		
17	38	30	-8		
18	90	80	-10		
19	58	12	-46		
20	62	25	-37		
Average	57.7	36.6	-21.1		
P-value	<.	.01 ^a	n/a		

^aStatistically significant.

improvements over the course of the follow-up period, which would be helpful for defining the natural history of recovery after PVCSO, given the inherent limitations of our study design. Future studies using a priori-defined assessment time points and standardized scales for all outcomes are necessary to define the role of PVCSO for TCS more clearly. Additionally, our study was not designed to directly compare detethering vs PVCSO in this population, which is why we compared the population to historical controls published in the literature. Additional studies directly comparing these treatment options are needed to inform decision-making in this area. Future studies are also needed to determine the optimal column reduction. While the mean reduction in this series was 23.4 mm, cadaveric studies have suggested that the optimal reduction to relieve tension on the cord while minimizing the risk of dural buckling may be between 12 and $16 \text{ mm}.^{23}$

Interpretation and Generalizability

Lin et al²⁰ performed a systematic review and meta-analysis of patients with TCS treated with PVCSO, which included 57 patients across 6 retrospective studies. They found that rates of symptom improvement ranged from 60% to 96% for pain or numbness, 13% to 67% for sensory abnormalities, and 79% to 100% for urinary and bowel dysfunction. However, only 1 of those studies²⁴ included patients with a history of failed detethering surgery (n = 3), which limits its direct comparison





with other studies. Additionally, our results compare favorably to symptomatic improvement after detethering procedures, with reduced risk of CSF leak and wound infection. Table 6 offers a comparison of the outcomes of the 2 techniques.^{10,13,14,17,20,25-37}

Achieving favorable results in patients with recurrent TCS is particularly challenging. Patients with symptomatic TCS who have undergone prior detethering, such as those in the current study, tend to be less likely to experience symptom resolution compared to those undergoing index surgery.^{17,38-42} The decreased likelihood of improvement with repeat detethering is unclear. Some suggest that adult TCS is associated with scarring of the spinal cord in the intradural space, which leads to decreased CSF circulation, rather than mechanical traction of the spinal cord.^{20,24,43} Thus, repeat intradural exposure and intradural surgery would lead to further adhesions and scarring, limiting its effectiveness. PVCSO may tackle this problem using a superior approach, whereby shortening of the spinal column directly leads to shortening of the stretched and tethered spinal cord. Further investigation is needed to substantiate this.

The advantages of PVCSO, including the potential for more durable symptom relief compared to detethering in patients with TCS, must be weighed against the potential risks of the procedure (Table 1). Complication rates after PVCSO are not well defined in the TCS literature. However, the International Spine Study Group has reported complication rates as high as 78% in adult patients with spinal deformity after 3-column osteotomy at a minimum 2-yr follow-up, including most commonly rod breakage (32%) and dural tear (21%).⁴⁴ Proximal junctional kyphosis was also noted in 9.8% of patients; the mean number of instrumented levels was 12.9, and the mean estimated blood loss was 3.3 liters.⁴⁴ Complication rates in our cohort were much lower, which may be explained by the decreased number of instrumental levels and bony work required for treatment.

CONCLUSION

We report a multicenter, retrospective analysis on the safety and efficacy of PVCSO in 20 adult patients with TCS caused by lipomyelomeningocele, in whom previous detethering surgery had failed. Our results suggest that PVCSO is a safe and effective treatment that may ultimately serve as an alternative to repeat intradural detethering in this population. Our series significantly contributes to the body of literature on the subject and shows that excellent initial results can be achieved in the particularly challenging population of adult patients with prior failed detethering for lipomyelomeningocele.

TABLE 5. Radiological Outcomes of Patients at Final Follow-up (Mean: 23.3 Months)						
	Spinal column boight	Local kyphotic Cobb angle ^a				
Patient	reduction, mm	Preoperative	Final follow-up	Change		
1	24	8.5°	2.0 °	-6.5°		
2	23	4.3°	2.4 °	−1.9 °		
3	28	0.0°	0.0°	0.0°		
4	19	36.9°	16.5°	−20.4°		
5	n/a	n/a	14.9°	n/a		
6	23	3.8°	0.0°	-3.8°		
7	25	2.9 °	-3.3 °	−6.2°		
8	22	10.9°	9.5°	−1.4 °		
9	24	n/a	n/a	n/a		
10	26	3.6°	4.6°	1.0°		
11	27	n/a	1.3°	n/a		
12	26	3.7°	9.7 °	6.0°		
13	20	1.0°	1.0°	0.0°		
14	26	3.0°	15.0 °	12.0°		
15	18	n/a	6.7°	n/a		
16	21	7.6 °	2.0°	-5.6°		
17	23	30.6°	15.7°	−14.9°		
18	24	14.9°	10.3°	-4.6°		
19	21	2.3°	3.8°	1.5°		
20	25	3.5°	1.0°	-2.5°		
Average	23.4	8.6°	5.9 °	—2.9 °		

^aMeasured from the superior endplate of the proximal vertebra to the inferior endplate of the distal vertebra (relative to the osteotomized vertebra). A positive value indicates kyphosis, whereas a negative value indicates lordosis.

TABLE 6. Comparison of Outcomes for TCS Based on Operative Technique: Detethering Procedure or PVCSO					
	Treatment				
		PVCS	0		
	Detethering procedures	Literature ^{20 a}	This series		
Improvement in:					
Back/leg pain	56%-100% ^{17,23-26}	60%-96%	88%		
Fecal/urinary incontinence	14%-75% ²⁶⁻³²	79%-100%	52%		
Motor deficits	31%-80% ^{23,26,29,32,33}	80%-100%	80%		
Sensory abnormalities	35%-79% ^{17,25,28,33,34}	13%-67%	80%		
TCS recurrence:	5%-50% ^{10,13,14,26,35}	0%	0%		

^a Meta-analysis of 57 patients treated with PVCSO for TCS that resulted from a wide variety of pathologies. TCS, tethered cord syndrome; PVCSO, posterior vertebral column subtraction osteotomy.



cord tension relief after PVCSO for a 34-yr-old patient with TCS caused by lipomyelomeningocele. This patient had undergone 6 previous detethering procedures, all of which failed. This patient's column height was reduced by 21 mm, relieving tension on the cord. In C and D, the lateral and anteroposterior cord diameters are indicated at the middle of the osteotomized vertebrae (T12).

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Supplemental Digital Content 1. Procedural details for a typical PVCSO at the level of T12.

Supplemental Digital Content 2. Video. Intraoperative ultrasound video depicting spinal cord tethering immediately before PVCSO.

Supplemental Digital Content 3. Video. Intraoperative ultrasound video depicting cord tension relief in the same patient immediately after PVCSO.