

Clinical criteria for filum terminale resection in occult tethered cord syndrome

Petra M. Klinge, MD, PhD,¹ Owen P. Leary, BS,¹ Philip A. Allen, PhD,² Konstantina Svokos, DO, MS,¹ Patricia Sullivan, MD,¹ Thomas Brinker, MD, PhD,³ and Ziya L. Gokaslan, MD¹

¹Department of Neurosurgery, Warren Alpert Medical School of Brown University, Rhode Island Hospital, Providence, Rhode Island; ²Department of Psychology, University of Akron, Ohio; and ³Department of Neurosurgery, Medical School Hannover, Germany

OBJECTIVE Tethered cord syndrome (TCS) comprises three symptom categories: back/leg pain, bowel/bladder, and neurological complaints. MRI typically reveals a low-lying conus medullaris, filum terminale (FT) pathology, or lumbosacral abnormalities. FT resection is established in TCS but not in radiologically occult TCS (OTCS). This study aims to identify patients with OTCS who are likely to benefit from FT resection.

METHODS The authors recruited 149 patients with OTCS (31 pediatric, 118 adult) treated with FT resection—including only cases with progressive TCS, negative spine MRI, and no concurrent neurological/urological conditions. A comprehensive questionnaire collected patient self-reported symptoms and clinical findings at the preoperative and at 3- and 12-month follow-up examinations. Based on questionnaire data, the authors extracted a 15-item symptoms and findings scale to represent the three TCS symptom categories, assigning 1 point for each item present.

RESULTS OTCS presents without radicular/segmental sensorimotor findings, but with leg/back pain and conus dysfunction, in addition to leg fatigue and spasticity; the latter indicating an upper motoneuron pathology. The 15-item scale showed clinical improvement in 89% of patients at the 3-month follow-up and 68% at the 12-month follow-up. Multivariate analysis of the scale revealed that it accurately predicts outcome of FT resection in 82% of cases. Patients with a preoperative score exceeding 6 points are most likely to benefit from surgery.

CONCLUSIONS By applying the study's inclusion criteria and incorporating the novel 15-item scale, surgeons can effectively select candidates for FT resection in patients with OTCS. The observed outcomes in these selected patients are comparable to those achieved in degenerative spine surgery.

<https://thejns.org/doi/abs/10.3171/2024.1.SPINE231191>

KEYWORDS occult tethered cord syndrome; outcome assessment; symptom scale; congenital; lumbar; sacral; pain

TETHERED cord syndrome (TCS) is a condition that can be caused by a diseased filum terminale (FT), leading to stretch injury to the lower spinal cord, particularly the conus medullaris.¹ Typically, lumbar MRI shows a low-lying conus medullaris or a diseased, thickened, and/or fatty FT. MRI often reveals additional pathologies like congenital or acquired lumbosacral abnormalities.² Clinically, TCS presents with a triad of symptom categories, which consist of back and leg pain, neurological leg symptoms, and bowel and bladder symptoms. Neurocutaneous signs and orthopedic findings like scoliosis are also common. Detethering, accomplished through sectioning or excision of the FT, has been shown to improve the symptomatology (reviewed by O'Connor et al.³).

Interestingly, the symptomatology of TCS has also

been observed in cases with negative MRI findings.⁴ This pathology has been termed occult tethered cord syndrome (OTCS) (reviewed by Rezaee and Keykhosravi).⁵ More recently, we found that OTCS is associated with congenital or acquired ultrastructural collagen damage of the FT. Applying biomechanical testing of FT specimens, we have shown that this pathology can cause stretch injury to the conus and lower spinal cord even in cases with a radiologically normal FT.⁶ In that study we also analyzed the clinical outcomes following FT resection in OTCS. We found a significant improvement in preoperative symptoms with outcomes assessed up to 12 months.⁶ However, that study was based on clinician-reported observations that were eventually found to be prone to investigator bias.

Research in spine surgery has emphasized the signifi-

ABBREVIATIONS EDS = Ehlers-Danlos syndrome; FT = filum terminale; OTCS = occult tethered cord syndrome; PROM = patient-reported outcome measure; ROC = receiver operating characteristic; TCS = tethered cord syndrome.

SUBMITTED November 2, 2023. **ACCEPTED** January 8, 2024.

INCLUDE WHEN CITING Published online March 15, 2024; DOI: 10.3171/2024.1.SPINE231191.

cance of patient self-reported symptoms for outcome studies (reviewed by Beighley et al.⁷). Accordingly, the present study analyzes the outcome of FT resection in OTCS by evaluating patient self-reported symptoms in addition to neurological signs found on clinical examination. We used a comprehensive questionnaire to gather such data before surgery and at 3- and 12-month follow-up appointments. We used questionnaire data to construct a 15-item scale that includes the 5 most prevalent symptoms or examination findings of each of the three TCS symptom categories. We analyzed this scale for tracking the progress of patients with OTCS after FT resection. Additionally, we explored the scale's potential in predicting outcomes based on preoperative symptoms and examination findings.

Methods

Applying the strict inclusion and exclusion criteria described below, this prospective single-center study enrolled 149 consecutive adult and pediatric patients with OTCS who completed a 3- and 12-month follow-up after FT excision between 2017 and 2021.

Inclusion and Exclusion Criteria

Patients were considered for surgery when presenting with symptoms from all three components of the classic TCS symptom triad.³ Although the presence of neurocutaneous signs of spina bifida occulta and/or scoliosis were supportive, they were not mandatory diagnostic elements.

Specific criteria applied to confirm surgical eligibility were as follows:

- Clinical symptoms demonstrated progression and resistance to conservative treatment for a minimum of 6 months.
- In accordance with the study protocol, the urologist was asked to exclude nonneurogenic bladder dysfunction, and had the discretion to select appropriate urological diagnostic procedures. Urodynamic studies were not obligatory for confirming neurogenic bladder dysfunction due to their low sensitivity and specificity in OTCS.⁸
- MRI of the entire neuraxis was conducted by study-independent radiologists to exclude any alternative pathology explaining the symptoms. Particularly, patients with radiological presence of low-lying conus medullaris (at or below the L2/3 level), radiologically altered FT, spinal dysraphism, or intraspinal masses were excluded. In addition, based on the MRI findings and a clinical examination by the senior study surgeon (P.M.K.), patients with a degenerative spine abnormality significant enough to contribute to the symptomatology were excluded.

Surgical Technique

Our surgical technique for FT resection has been separately published, including a video showing the surgical details.⁹ In brief, surgery was performed through a lumbar interspinous approach one vertebral level below the radiographic termination of the conus medullaris. Following durotomy under microscopic magnification, the FT was

isolated with intraoperative electrophysiological monitoring. Then the FT was cauterized and transected. Given that the FT is vascularized by just one artery originating at the conus level,¹⁰ following cauterization the distal portion of the FT is effectively devascularized. Consecutive FT degeneration bears the risk of scar formation leading to future retethering. To address this risk, gentle stretch forces were applied to the distal severed FT until a snap was felt indicating its rupture. Then it could be completely pulled out. The technical feasibility of this extraction is based on biomechanical studies revealing the FT's plastic properties in response to gentle but unphysiological stretch forces until it breaks at its thinnest portion,^{6,11,12} which is the sacral dura insertion site. Notably, we observed no instances of intra- or postoperative hemorrhage. All FT specimens underwent routine pathological examination.

The dura mater was closed primarily and sealed with fibrin sealant including an autologous fat graft (Tisseel, Baxter) and a Duraform patch (Natus Medical, Inc.). Patients underwent a complete postoperative bed rest period of 24 hours.

Questionnaire

A comprehensive questionnaire collected patient self-reported symptoms in addition to neurological examination findings before surgery and at 3- and 12-month follow-up visits. Pediatric patients who were able to engage with survey questions participated with parental assistance if needed. Patients older than 18 years at the time of index surgery strictly self-completed the patient-reported symptom questionnaires.

The questionnaire covered previously reported TCS symptoms and findings,^{3,5,13} and included those reported in our previous TCS/OTCS study.⁶ We also recorded the general medical history including orthopedic and connective tissue comorbidity. Table 1 shows OTCS-specific questions and answers offered by the patient as well as the requested clinician examination findings. Of note, questionnaire data were just collected but not analyzed prior to the termination of the data collection period, which was 12 months after the inclusion of the last study patient.

We used the questionnaire data to construct a 15-item scale that represents the five most prevalent symptoms and signs in the neurological, bowel and bladder, and pain categories (Table 2). The 15-item scale scores each present symptom with a single point, resulting in a total score range of 0 to 15. To streamline the questionnaire data, we allocated only 1 point for each symptom and finding regardless of whether they occurred on the left or right side or were bilaterally. A point was given for foot clonus with a level greater than 2/5. Bowel symptoms, which encompassed incontinence and/or obstipation, scored as a single point. We excluded segmental or radicular sensorimotor findings from the scale due to their rare occurrence. Additionally, we omitted observations that would remain unchanged after surgery, such as neurocutaneous signs, connective tissue disorders, and scoliosis. Applying a variety of statistical methods, we analyzed the applicability of the scale for assessment and prediction of outcome following FT excision for OTCS.

TABLE 1. Questionnaire for preoperative (baseline) and follow-up symptoms examinations

Category & Questions	Offered Answers
Bowel & bladder	
Bladder Sxs: leaking, urgency, frequency, hesitation , retention	Y/N , duration (none, <1 yr, 1–3 yrs, 3–5 yrs, 5–10 yrs, >10 yrs)
Urinary infection	None, ≤3 times/yr, >3 times/yr
Pain during urination	Y/N
Obstipation	Y/N
Bowel incontinence	Y/N
Pain	
Leg	Pain: Y/N , duration (<1 yr, 1–3 yrs, 3–5 yrs, 5–10 yrs, >10 yrs) Distribution (variable, lt, rt, both sides) Quality (can not describe, burning, aching, spastic, stabbing) Occurrence (day, night, both, constant, daily, sometimes, occasionally, rarely) Cramps: Y/N , occurrence (as above) Sensory signs: Y/N , duration (in yrs, as above)
Back	Y/N , duration, distribution, quality, occurrence, cramps Y/N , sensory signs
Sacral	Y/N
Neurology	
Fatigue leg	Y/N, lt/rt , increase w/ activity Y/N
Paresthesia leg	Y/N, lt/rt , increase w/ activity Y/N
Motor examination (C5–T1 & L2–S1)*	Motor score (5/5) for each myotome lt & rt
Hyperreflexia*	Y/N, UE, LE, rt, lt
Foot clonus*	Y/N, >2 on the 5/5 scale (0 = none, 2 = normal), rt, lt
Increased muscle tone*	Y/N, UE, LE, rt, lt
Other findings	
Scoliosis*	Y/N
Kyphosis*	Y/N
Neurocutaneous signs*	Dimple, discoloration, asymmetrical gluteal fold, hairy patch

LE = lower extremity; Sxs = symptoms; UE = upper extremity; Y/N = yes/no.

Boldface type indicates items that were used for the construction of the 15-item symptom scale shown in Table 2.

* Clinical findings; all other items were patient reported.

Statistics

Statistical analysis of the clinical data was performed using R software (version 4.0.3, www.r-project.org). Various statistical techniques were applied, including exploratory and confirmatory factor analysis, Cronbach's alpha test, Friedman rank testing with post hoc Wilcoxon test, generalized multivariate linear modeling, logistic regression, and receiver operating characteristic (ROC) curve analysis. Detailed explanations and references for these procedures can be found in the *Results* section.

Ethical Approval

The patient's or legal guardian's informed consent was obtained and documented. The project was approved by the local institutional review board.

Results

Patient Population

The study included 149 patients with a median age of 24.1 years (IQR 25.1). Twenty-three patients (15%) were male, 123 (83%) were female, and 3 (2%) identified as non-

binary. A total of 31 (21%) were less than 18 years old, with 7 of those being younger than 6 years.

Of the participants, 87 (58%) had a previous diagnosis of comorbid Ehlers-Danlos syndrome (EDS), and 59 (40%) presented with scoliosis. Additionally, we observed a sacral dimple in 49 (33%) patients, local skin discoloration in 7 (5%), a hairy spot in 10 (7%), and an asymmetrical gluteal fold in 13 (9%).

Preoperative Symptoms and Findings

Among the patients, 29% reported experiencing the symptom triad of TCS within the past 6–12 months, 33% within 1–5 years, 7% within 5–10 years, and 17% for more than 10 years. Additionally, 14% acknowledged experiencing the triad for at least 6 months but were unable to specify a more definite duration due to the insidious onset of symptoms.

Only a small number of patients displayed clear radicular or segmental sensorimotor deficits. Figure 1 illustrates the motor strength findings. Table 2 presents the most prevalent patient self-reported symptoms and neurological examination findings. Notably, neurological findings are

TABLE 2. Prevalence of symptoms on the 15 item-scale

Category or Sx	No. of Patients (%)
Neurological scale	
Increased muscle tone legs*	42 (28)
Hyperreflexia legs*	98 (66)
Foot clonus*	83 (56)
Fatigue legs	127 (85)
Paresthesia legs	84 (56)
Pain scale	
Leg	132 (89)
Sacral	62 (42)
Low-back	119 (80)
Cramps in leg & back	84 (56)
Fluctuating pain Sxs	42 (28)
Bowel & bladder scale	
Urinary leakage	85 (57)
Urinary urgency	87 (58)
Urinary frequency	78 (52)
Urinary hesitation	72 (48)
Bowel Sxs	92 (62)

* Objective clinical findings.

characterized by the presence of foot clonus, heightened lower-extremity hyperreflexia, and increased leg tone, all symptoms indicative of upper motoneuron dysfunction.

Figure 2 presents histograms depicting the 15-item scale and its symptom categories (subscales). Most patients (n = 130, 87.2%) exhibited TCS symptoms across all 3 categories, followed by 17 patients (11.4%) with symptoms in 2 categories, and only 2 patients (1.3%) with symptoms in

a single category. This indicates that the scale, based on only 15 symptoms and findings, has a limited diagnostic sensitivity. However, it accurately validated the preoperative clinical diagnosis of a symptom triad with an 87.2% confirmation rate.

Follow-Up and Prediction of Outcome

Postoperative complications necessitating surgical revision included CSF leak (n = 3), cauda equina tethering diagnosed on MRI (n = 8 within 12 months postsurgery), and local wound revision (n = 5). No occurrences of acute cauda syndrome or new radicular or segmental sensorimotor findings were observed postoperatively.

We used the 15-item scale to assess follow-up outcomes after FT resection. Applying the Friedman test with paired post hoc Wilcoxon test, all differences comparing the follow-up results with the preoperative scores indicated statistically significant improvement with p < 0.001, except the 12-month outcome of both bowel and bladder symptoms (p < 0.0026) and pain (p < 0.084) (Fig. 3).

We assessed clinical improvement by calculating the difference between preoperative and 12-month follow-up scores on the 15-item scale. Using multivariate generalized linear modeling, we found that the preoperative scores of the three subscales served as predictors for the improvement parameter. Notably, when examining the preoperative presence of individual items of the scale, only pain fluctuation exhibited predictive power. Interestingly, comorbidities (EDS, scoliosis, and neurocutaneous signs) did not significantly predict the improvement parameter (Table 3).

To further evaluate the predictive potential of the scale in individual patients, we divided the cases into two groups. Responders (n = 102, 68%) were those who exhibited an improvement of at least 1 point between their preoperative

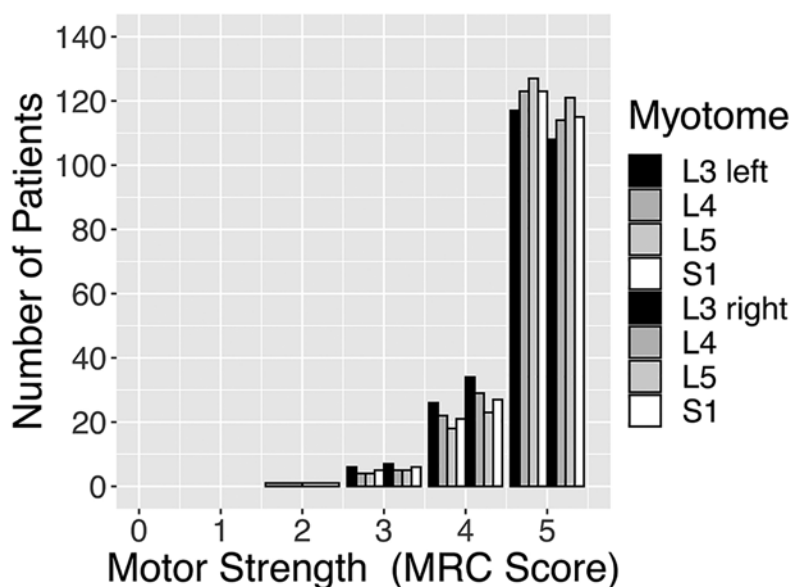


FIG. 1. Motor strength was assessed using the Medical Research Council (MRC) scale, which revealed a grade of 4/5 or 5/5 in most cases. Only 9 patients in total had a grade of 3/5 (n = 8) or 2/5 (n = 1). Among these 9 cases, 4 had a remote medical history that explained the paresis. The other 5 patients experienced severe pain, and examination results were inconsistent.

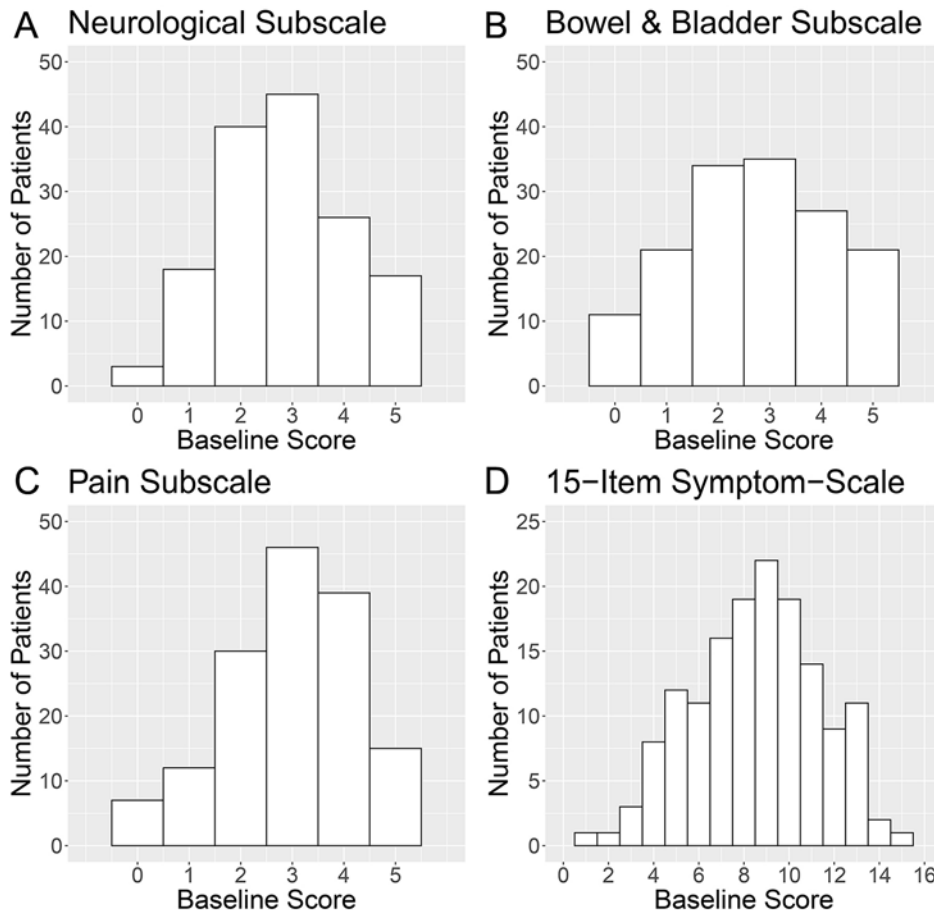


FIG. 2. Histograms of the baseline scores for each symptom category and the total 15-item scale.

assessment and the 12-month follow-up. All other cases were categorized as nonresponders ($n = 47$, 32%). When we applied logistic regression analysis, we observed that among patients with a preoperative score greater than 6, the likelihood of improvement at the 12-month follow-up exceeded 60%. When the preoperative score exceeded 8, this likelihood increased to more than 80%, and for those with a score higher than 10, it reached beyond 90%. Furthermore, the ROC curve¹⁴ showed that the scale accurately distinguishes surgical responders from nonresponders in 82% of cases. This suggests that the scale is a useful tool for predicting surgical outcomes (Fig. 4).

Statistical Validation of the 15-Item Scale

Because we used questionnaire data, we applied psychometric statistical procedures to assess the validity and reliability of the scale.¹⁵ Cronbach's alpha coefficient measures the internal consistency, or reliability, of a set of survey items.¹⁶ Testing the set of the three categories of our 15-item scale, Cronbach's alpha was 0.507 at the preoperative baseline, 0.72 at the 3-month follow-up, and 0.552 at the 12-month follow-up. A test result spanning from 0.5 to 0.8 indicates moderate, but still acceptable reliability.

Confirmatory factor analysis¹⁷ was used to test validity and revealed a root mean square error of approximation of 0.053, which indicates a moderate model fit and suggests

that the total scores of each of the 3 categories effectively represent the entire 15-item scale. Although lacking statistical significance, exploratory factor analysis revealed a 3-factor model that exhibited the best fit, resulting in the following categories: 1) pain (leg and back); 2) conus dysfunction; and 3) spasticity (leg and bladder combined).

Discussion

Patient-reported outcome measures (PROMs) have been recognized as a valuable instrument for outcome studies in spine surgery.⁷ Our study is the first demonstrating the potential of PROMs for outcome assessment following FT resection in OTCS.

Upper Motoneuron Dysfunction in OTCS

We found that radiologically occult TCS presents upper, but not lower motoneuron dysfunction given that the clinical examination revealed leg spasticity (leg hyperreflexia, increased muscle tone, and foot clonus) without segmental or radicular sensorimotor deficits.¹⁸ Besides those objective clinical findings, patient self-reported leg muscle fatigue has been also recognized as a symptom of upper motoneuron diseases.¹⁹ Leg fatigue is described by the patients as severe muscle weakness related to exertion of limb muscles and the inability to sustain motor function

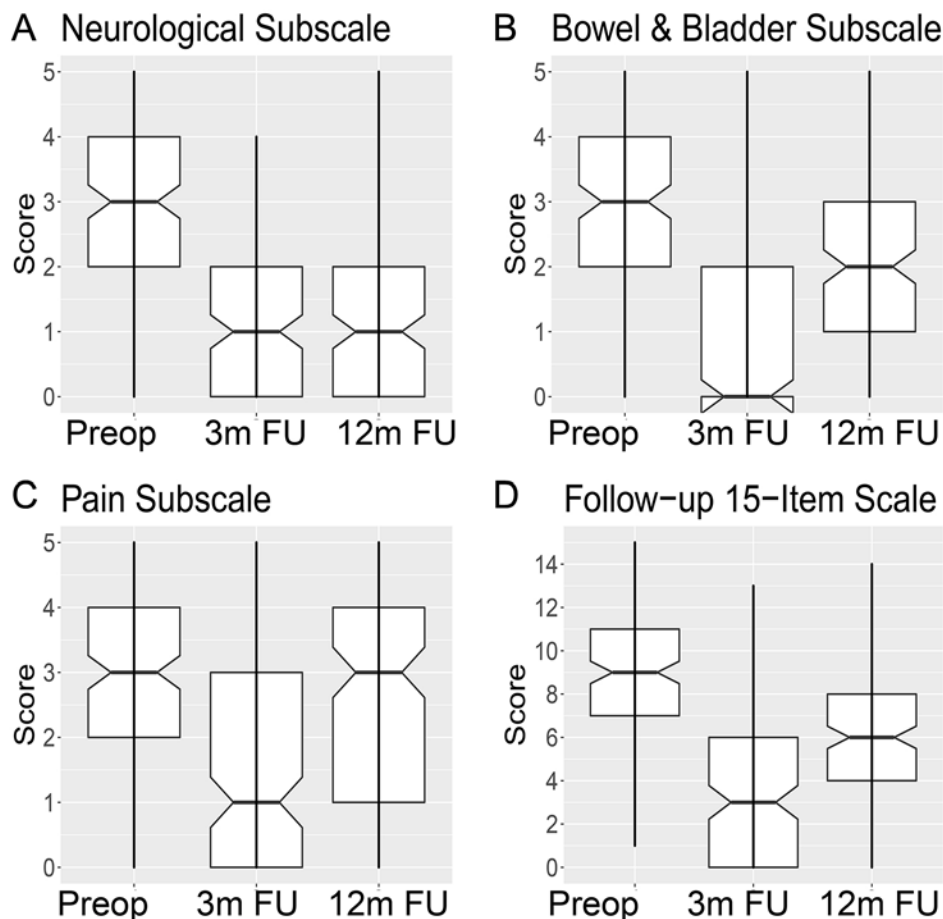


FIG. 3. Preoperative and postoperative scores of the 15-item scale and its subscales. The box plot shows the median, IQR, and the 95th percentiles of the scores at the preoperative baseline and at 3-month and 12-month follow-ups. The improvement following FT excision was most remarkable at the 3-month follow-up, but the 12-month follow-up revealed long-term improvement too. The 12-month pain score was the only one that was not significantly improved. Statistics results are shown in the text. FU = follow-up; m = month.

and motor strength. Leg fatigue was prevalent in 82% of our cases. Clinically, muscle fatigue is a disabling symptom, and many patients need walking aids or even a wheelchair after short walking distances.

What Could Cause Isolated Upper Motoneuron Dysfunction in OTCS?

Our previous experimental and clinical work has shown that a diseased but normal-length FT may expose the co-

TABLE 3. Generalized linear modeling of improvement

	Estimate	SE	t Value	Pr(> t)	Significance
(Intercept)	-2.6651	0.7861	-3.39	0.000904	***
Neurological subscale	0.8495	0.2134	3.98	0.000109	***
Bowel & bladder subscale	0.3677	0.1661	2.213	0.028493	*
Pain subscale	0.6216	0.2381	2.61	0.010014	*
EDS	-0.6049	0.5421	-1.116	0.266362	NS
Scoliosis	-0.2564	0.5495	-0.467	0.641493	NS
Neurocutaneous signs	0.4442	0.504	0.881	0.379601	NS

NS = not significant.

Dependent variable: improvement calculated as the difference between preoperative and 12-month postoperative scores on the 15-item scale. Independent variables: the 3 subscales, EDS, scoliosis, and neurocutaneous signs.

* $p < 0.05$; *** $p < 0.001$.

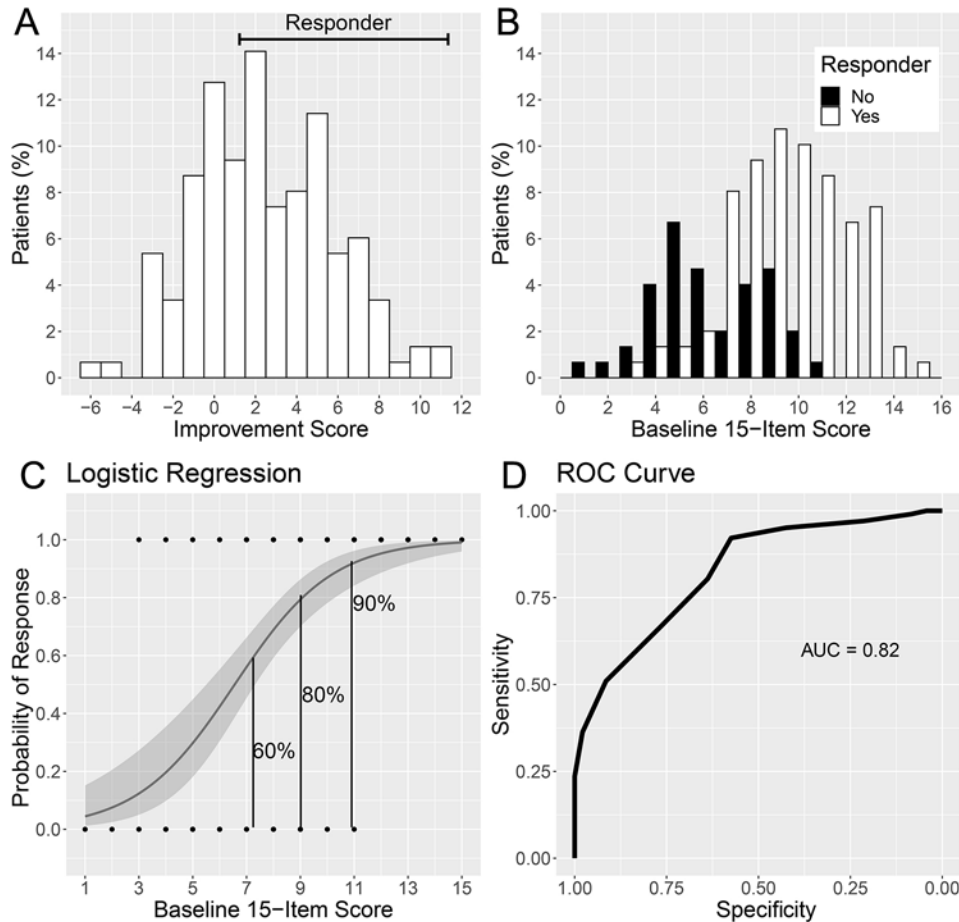


FIG. 4. The 12-month outcome prediction. **A:** Histogram of the improvement score calculated as the difference between baseline and 12-month 15-item score. Patients with at least a 1-point improvement were categorized as responders. **B:** The histogram shows responders and nonresponders at the 12-month follow-up in relation to the preoperative baseline score. Improvement was found in 68% of the patients. **C:** Logistic regression indicates that the probability for improvement is 60% for cases with a preoperative score of more than 6; 80% for those with a score of more than 8; and 90% for those with a score of more than 10 points. The shaded area indicates the 95% confidence interval. **D:** The ROC curve analysis¹⁴ illustrates the high specificity and sensitivity of the 15-item symptom scale indicating that the scale accurately predicts the outcome in 82% of the cases. AUC = area under the curve.

nus to pathological stretch forces.⁶ Such a diseased FT has impaired biomechanical elastic properties, which may be caused by acquired tendinopathy and/or congenital structural collagen fibril weakness in connective tissue disorder such as EDSs.^{6,20} Other groups have experimentally shown in cats¹ and human cadaver studies¹² that axial stretch forces applied to the FT extend through the entire lower spinal cord. Because the FT is the only axial ligament connecting the spinal cord with the skeleton, transmitted stretch forces should specifically affect axial spinal cord structures, including the conus and corticospinal tracts. Interestingly, it was shown in motoneuron cell cultures that even very mild forces can cause axonal stretch injury when applied repeatedly.²¹ It should also be recognized that pathological FT stretch forces can contribute to lumbosacral pain, because stretch-sensitive nociceptive structures are integrated into the collagen fibrous structures of the FT.²² Furthermore, it is recognized that muscle spasticity is correlated with significant pain sensation.²³

Ultimately, our study supports previous suggestions^{1,6,24}

that stretch injury contributes to the clinical presentation of both TCS and OTCS. This notion is strongly supported by our observation that FT resection improved the OTCS symptomatology.

Follow-Up Assessment

PROMs have been extensively used for the assessment of outcome in spine surgery, but not yet in TCS or OTCS. A recent review recognized “a limited, though effective use”; accordingly, an increased use in spine surgery is advocated.⁷ The 15-item scale may be considered as a PROM, and it revealed significant improvements at 3- and 12-month follow-ups after surgery. All three symptom categories improved significantly at the 3-month follow-up, but at 12 months, the pain scale did not show the same level of improvement as bowel and bladder and neurological subscales. Overall, the novel scale revealed improvement rates that are on par with those reported in degenerative spine surgery.^{25,26}

The absence of sustained pain improvement at the

12-month follow-up appears to contradict the findings from our prior TCS/OTCS study, which had similar selection criteria for surgery. It is important to note that in the previous study, patient-reported symptoms were not recorded without clinical assessment.⁶ The discrepancy between the studies can be attributed to several factors. First, there is the potential for bias in the interpretation of patient symptoms by clinicians, leading to inconsistent reporting. Second, various unrecorded variables may have influenced patients' self-reported pain perceptions in the present study. These could include changes in medication, increased physical activity, placebo effects, and factors like chronic pain-related anxiety and depression. Furthermore, it is worth noting that our questionnaire only inquired about the presence of pain but did not assess its intensity. As a result, our scale may underestimate the actual extent of pain improvement.

Although our data suggest positive 1-year improvement trends, we acknowledge the possibility of these trends diminishing over an extended period.

Prediction of Outcome

Preoperative patient-reported data can be used to develop regression-based statistical models to identify patients who are likely to achieve a clinically important improvement of symptoms following spine surgery.²⁵ In our study population, multivariate linear modeling of the 15-item scale confirmed the predictive capabilities of its three symptom categories. Logistic regression analysis revealed that outcome in individual cases can be predicted: the higher the individual preoperative score, the higher the probability that the patient responds to surgery. ROC curve analysis demonstrated that this discrimination is highly specific and sensitive. Predicting outcomes of individual cases was accurate in 82% of cases.

It is important that the multivariate analysis demonstrated that the 3 symptom categories, rather than individual symptoms, serve as predictors of the surgical outcome. This finding could explain why the only randomized controlled OTCS trial failed to demonstrate the benefits of FT resection, because it included patients with urinary symptoms only.⁸

We conclude therefore that surgery is most favorable in patients who present all three symptom categories. That such symptom categories but not individual symptoms predict the outcome following OTCS surgery has been suggested previously by a retrospective analysis of 22 pediatric patients.²⁷ Also, it has been observed in acute conus medullaris syndrome²⁸ that only the combination of cauda dysfunction along with neurological and pain symptoms is highly specific and sensitive for diagnosing.

Limitations

Applying statistical methods like factor analysis and Cronbach's test, we recognize that the novel scale needs refinements to optimize its validity and reliability. Such refinements may include scaled pain assessments and quality of life measures. Eventually, this will require future studies because of the complexity of the process for the validation of medical scales.¹⁵ Also, the novel scale needs external validation—that is, the application of the

scale in a patient cohort not used for the generation of the scale.

It is important to note that our scale may not be suitable for young children due to its reliance on patient-reported data, making it more appropriate for adult patients. Also, its applicability for classic TCS cases needs further investigation.

Conclusions

By applying the study's inclusion criteria and incorporating the novel 15-item symptom scale, we can effectively select patients with OTCS who are likely to benefit from FT resection. The observed outcomes in these selected patients are comparable to those achieved in patients who undergo spine surgery for degenerative disease. However, before widespread clinical application, it is imperative that future studies focus on refining and optimizing the current scale.

Given that objective clinical findings are limited to signs of leg spasticity, the assessment and prediction of outcomes of FT resection in OTCS heavily relies on patient self-reported symptoms. Notably, the observation of upper motoneuron dysfunction sheds light on a specific pathomechanism in OTCS; that is, FT-transmitted axonal stretch injury to the lumbar corticospinal tracts. As spine surgeons, we should acknowledge the existence of OTCS as a manageable disorder. Diagnosis should be contemplated in cases with normal findings on spine MRI but with a symptom triad, including urinary dysfunction, leg/back pain, and lower-extremity neurological symptoms.

Acknowledgments

We thank our team of clinical research assistants who contributed to the collection of the patient symptom questionnaires, in particular Sarah Brown, Michelle Zhu, Shanzeh Sayied, Kevin Ma, and Darlene Gaudet.

References

1. Yamada S, Iacono RP, Andrade T, Mandybur G, Yamada BS. Pathophysiology of tethered cord syndrome. *Neurosurg Clin N Am.* 1995;6(2):311-323.
2. Hoffman HJ, Hendrick EB, Humphreys RP. The tethered spinal cord: its protean manifestations, diagnosis and surgical correction. *Childs Brain.* 1976;2(3):145-155.
3. O'Connor KP, Smitherman AD, Milton CK, et al. Surgical treatment of tethered cord syndrome in adults: a systematic review and meta-analysis. *World Neurosurg.* 2020;137:e221-e241.
4. Warder DE, Oakes WJ. Tethered cord syndrome and the conus in a normal position. *Neurosurgery.* 1993;33(3):374-378.
5. Rezaee H, Keykhosravi E. Effect of untethering on occult tethered cord syndrome: a systematic review. *Br J Neurosurg.* 2022;36(5):574-582.
6. Klinge PM, Srivastava V, McElroy A, et al. Diseased filum terminale as a cause of tethered cord syndrome in Ehlers-Danlos syndrome: histopathology, biomechanics, clinical presentation, and outcome of filum excision. *World Neurosurg.* 2022;162:e492-e502.
7. Beighley A, Zhang A, Huang B, et al. Patient-reported outcome measures in spine surgery: a systematic review. *J Craniovertebr Junction Spine.* 2022;13(4):378-389.
8. Steinbok P, MacNeily AE, Hengel AR, et al. Filum section for urinary incontinence in children with occult tethered cord

- syndrome: a randomized, controlled pilot study. *J Urol*. 2016; 195(4 Pt 2):1183-1188.
9. Abdulrazeq H, Shao B, Sastry RA, Klinge PM. Microsurgical approach for resection of the filum terminale internum in tethered cord syndrome—a case demonstration of technical nuances and vignettes. *Acta Neurochir (Wien)*. 2023;165(11): 3505-3509.
 10. Djindjian M, Ribeiro A, Ortega E, Gaston A, Poirier J. The normal vascularization of the intradural filum terminale in man. *Surg Radiol Anat*. 1988;10(3):201-209.
 11. Yamada S, Knierim D, Yonekura M, Schultz R, Maeda G. Tethered cord syndrome. *J Am Paraplegia Soc*. 1983;6(3): 58-61.
 12. De Vloot P, Monea AG, Sciort R, van Loon J, Van Calenbergh F. The filum terminale: a cadaver study of anatomy, histology, and elastic properties. *World Neurosurg*. 2016;90:565-573.e1.
 13. Klekamp J. Tethered cord syndrome in adults. *J Neurosurg Spine*. 2011;15(3):258-270.
 14. Mandrekar JN. Receiver operating characteristic curve in diagnostic test assessment. *J Thorac Oncol*. 2010;5(9):1315-1316.
 15. Boateng GO, Neilands TB, Frongillo EA, Melgar-Quinonez HR, Young SL. Best practices for developing and validating scales for health, social, and behavioral research: a primer. *Front Public Health*. 2018;6:149.
 16. Cho E, Kim S. Cronbach's coefficient alpha: well known but poorly understood. *Organ Res Methods*. 2015;18(2):207-230.
 17. Rosseel Y. lavaan: an R package for structural equation modeling. *J Stat Softw*. 2012;48(2):1-36.
 18. Emos MC, Agarwal S. Neuroanatomy, upper motor neuron lesion. In: *StatPearls*. StatPearls Publishing; 2023. Accessed January 22, 2024. <https://www.ncbi.nlm.nih.gov/books/NBK537305/>
 19. Gibbons CJ, Thornton EW, Young CA. The patient experience of fatigue in motor neurone disease. *Front Psychol*. 2013;4:788.
 20. Hills S, Pugacheva A, Weltin P, et al. Tethered cord syndrome in KBG syndrome. *Am J Med Genet A*. 2023;191(5): 1222-1226.
 21. Yap YC, King AE, Guijt RM, et al. Mild and repetitive very mild axonal stretch injury triggers cystoskeletal mislocalization and growth cone collapse. *PLoS One*. 2017;12(5): e0176997.
 22. Klinge PM, McElroy A, Leary OP, et al. Not just an anchor: the human filum terminale contains stretch sensitive and nociceptive nerve endings and responds to electrical stimulation with paraspinal muscle activation. *Neurosurgery*. 2022;91(4): 618-624.
 23. Marsden J, Stevenson V, Jarrett L. Treatment of spasticity. *Handb Clin Neurol*. 2023;196:497-521.
 24. Henderson FC Sr, Austin C, Benzel E, et al. Neurological and spinal manifestations of the Ehlers-Danlos syndromes. *Am J Med Genet C Semin Med Genet*. 2017;175(1):195-211.
 25. Halicka M, Wilby M, Duarte R, Brown C. Predicting patient-reported outcomes following lumbar spine surgery: development and external validation of multivariable prediction models. *BMC Musculoskelet Disord*. 2023;24(1):333.
 26. Daniell JR, Osti OL. Failed back surgery syndrome: a review article. *Asian Spine J*. 2018;12(2):372-379.
 27. Fabiano AJ, Khan MF, Rozzelle CJ, Li V. Preoperative predictors for improvement after surgical untethering in occult tight filum terminale syndrome. *Pediatr Neurosurg*. 2009; 45(4):256-261.
 28. Rider LS, Marra EM. Cauda equina and conus medullaris syndromes. In: *StatPearls*. StatPearls Publishing; 2023. Accessed January 22, 2024. <https://www.ncbi.nlm.nih.gov/books/NBK537305/>

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: Klinge, Svokos, Gokaslan. Acquisition of data: Leary, Svokos. Analysis and interpretation of data: Klinge, Leary, Allen, Svokos, Brinker. Drafting the article: Klinge, Brinker. Critically revising the article: Klinge, Leary, Allen, Svokos, Brinker, Gokaslan. Reviewed submitted version of manuscript: Klinge, Leary, Allen, Sullivan, Brinker, Gokaslan. Approved the final version of the manuscript on behalf of all authors: Klinge. Statistical analysis: Allen, Brinker. Administrative/technical/material support: Leary, Sullivan. Study supervision: Klinge, Leary.

Correspondence

Petra M. Klinge | Warren Alpert School of Medicine of Brown University, Rhode Island Hospital, Providence, RI. petra_klinge@brown.edu