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Original Article

Thecal sac ligation in the setting of thoracic spondyloptosis with complete cord transection

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Received: 23 April 2023
Accepted: 24 July 2023
Published: 01 September 2023

DOI
10.25259/SNI_360_2023

Quick Response Code:

ABSTRACT

Background: Traumatic spondyloptosis (TS) with complete spinal cord transection and unrepairable durotomy is particularly rare and can lead to a difficult-to-manage cerebrospinal fluid (CSF) leak.

Methods: We performed a systematic review of the literature on TS and discuss the management strategies and outcomes of TS with cord transection and significant dural tear. We also report a novel case of a 26-year-old female who presented with thoracic TS with complete spinal cord transection and unrepairable durotomy with high-flow CSF leak.

Results: Of 93 articles that resulted in the search query, 13 described cases of TS with complete cord transection. The approach to dural repair was only described in 8 (n = 20) of the 13 articles. The dura was not repaired in two (20%) of the cases. Ligation of the proximal end of the dural defect was done in 15 (75%) of the cases, all at the same institution. One (5%) case report describes ligation of the distal end; one (5%) case describes the repair of the dura with duraplasty; and another (5%) case describes repair using muscle graft to partially reconstruct the defect.

Conclusion: Suture ligation of the thecal sac in the setting of traumatic complete spinal cord transection with significant dural disruption has been described in the international literature and is a safe and successful technique to prevent complications associated with persisting high-flow CSF leakage. To the best of our knowledge, this is the first report of thecal sac ligation of the proximal end of the defect from the United States.

Keywords: Cerebrospinal fluid leak, Cord transection, Dural tear, Thecal sac ligation, Traumatic spondyloptosis

INTRODUCTION

Spondyloptosis is a severe form of spondylolisthesis with complete disruption of all structural elements of the spinal column resulting in severe instability. Traumatic spondyloptosis (TS) is rare but may be caused by high-velocity injury or blunt trauma such as motor vehicle collisions (MVC), falls, or building collapse.[2,5,22,25,31] It is commonly associated with polytrauma, including additional rib or vertebral fractures, hemothorax, pneumothorax, hematomas, or head injuries.[20] It is not uncommon for patients with TS to present with complete neurologic injury on the

American Spinal Injury Association (ASIA) Impairment scale.[9,13,17,24] Neurogenic shock is a life-threatening complication resulting in hemodynamic instability and autonomic dysautonomia from loss of sympathetic tone. In patients with cervical cord injury, shock may occur in up to 29% of patients.[27]

Long-term complications of polytraumatic spinal cord injury are numerous. If the dura is violated, complications regarding cerebrospinal fluid (CSF) leakage may increase the length of stay and impede the rehabilitation process. Complications during the rehabilitation process may be attributed to long hospital stays or long stays in rehabilitation facilities.[4] These include but are not limited to pressure ulcers, autonomic dysreflexia, atelectasis, pneumonia, urinary tract infections, spasticity, muscle atrophy, bone loss, and depressive disorders.[4,23] New innovations to manage these complications are intertwined with advancements in therapeutic strategies. For example, locomotor training has been beneficial for functional improvement and may decrease rates of stasis-related complications.[23] Other innovations include neuromuscular electrical stimulation to decrease muscle atrophy, pharmacologic interventions for spasticity, and increased social support and access to mental health professionals throughout the rehabilitation process.[23]

Few cases have been reported of TS with complete spinal cord transection and associated unrepairable traumatic durotomy.[9,14,29] Associated traumatic durotomy not properly repaired may lead to CSF fistula, which increases the risk of complications including pseudomeningocele, meningitis, impaired wound healing, and subdural hemorrhages.[21,22] Management of a dural tear in this setting is without consensus and depends on the severity of the injury.[20] Suture closure in a watertight fashion, reconstruction with muscle or fat graft, and CSF diversion are management strategies for most dural tears causing CSF leakage.[20,21] However, in the setting of high-flow spinal fluid leakage with complete destruction of the dura that cannot be repaired to reconstitute the normal dural tube, options remain limited. Here, we present a case of TS with complete cord transection and associated traumatic durotomy requiring thecal sac suture ligation to control a high-flow CSF leak. To the best of our knowledge, it is the first reported case of its kind in the United States.

MATERIALS AND METHODS

Search strategy

We systematically reviewed the literature in accordance with the preferred reporting items for systematic reviews and meta-analysis guidelines. One author (S.V.) queried PubMed on April 15, 2023, using the search term "traumatic spondyloptosis." There were no limits on the year of publication. The search query is shown in Figure 1.

Selection process and data collection

After the articles returned in the search were organized, two authors (S.V. and M.D.K.) independently screened the abstracts and titles for relevance. Articles not available in English or articles without full manuscripts were removed. Our inclusion and exclusion criteria were applied to the remaining articles. Articles were included in our study if they met all of the following inclusion criteria: (1) Reported case(s) described TS and (2) complete transection of the cord of dural sac/nerve roots was explicitly stated. Articles were excluded from our study if they met any one of the following exclusion criteria: (1) Article was a literature review, systematic review, or meta-analysis and (2) operative approach to spondyloptosis was not described.

Data items extracted from the included articles were country of origin, year of publication, level of spondyloptosis, approach to the spinal cord or dural repair, presence of postoperative CSF leak, and overall outcomes. A case of TS was also reviewed for presentation.

Study risk of bias assessment

Two reviewers (S.V.) and (E.S.) assessed the risk of bias using the Joanna Briggs Institute Critical Appraisal Checklist for Case Series and Case Reports, respectively. Low risk was defined as an answer of Yes for >75% of the questions [Supplementary Table 1].

RESULTS

Case presentation

A 26-year-old female was transferred from an outside hospital after sustaining a rollover motor vehicle accident (MVA). She was an unrestrained driver and was violently ejected from the driver's seat into the back of the car, requiring subsequent extrication by Emergency Medical Services. The patient had no significant past medical history except for a COVID (SARS-CoV-2) infection several months prior and a persistent asymptomatic positive COVID test.

On presentation to our medical center, the patient was awake and talking with normal neurological function in the upper extremities and no motor or sensory function below the T6 level consistent with an ASIA grade A injury. Computed tomography imaging showed bilateral pleural effusions, multiple fractures of the ribs, and traumatic thoracic spondyloptosis of T7 on T8 with anterior and lateral subluxation of the T7 vertebral body compared to the T8 vertebral body [Figure 2a]. Magnetic resonance imaging revealed complete disruption of all the supporting ligaments at that level and complete transection of the spinal cord. The two ends of the spinal cord were not in continuity. The rostral segment of the transected spinal cord was noted to terminate

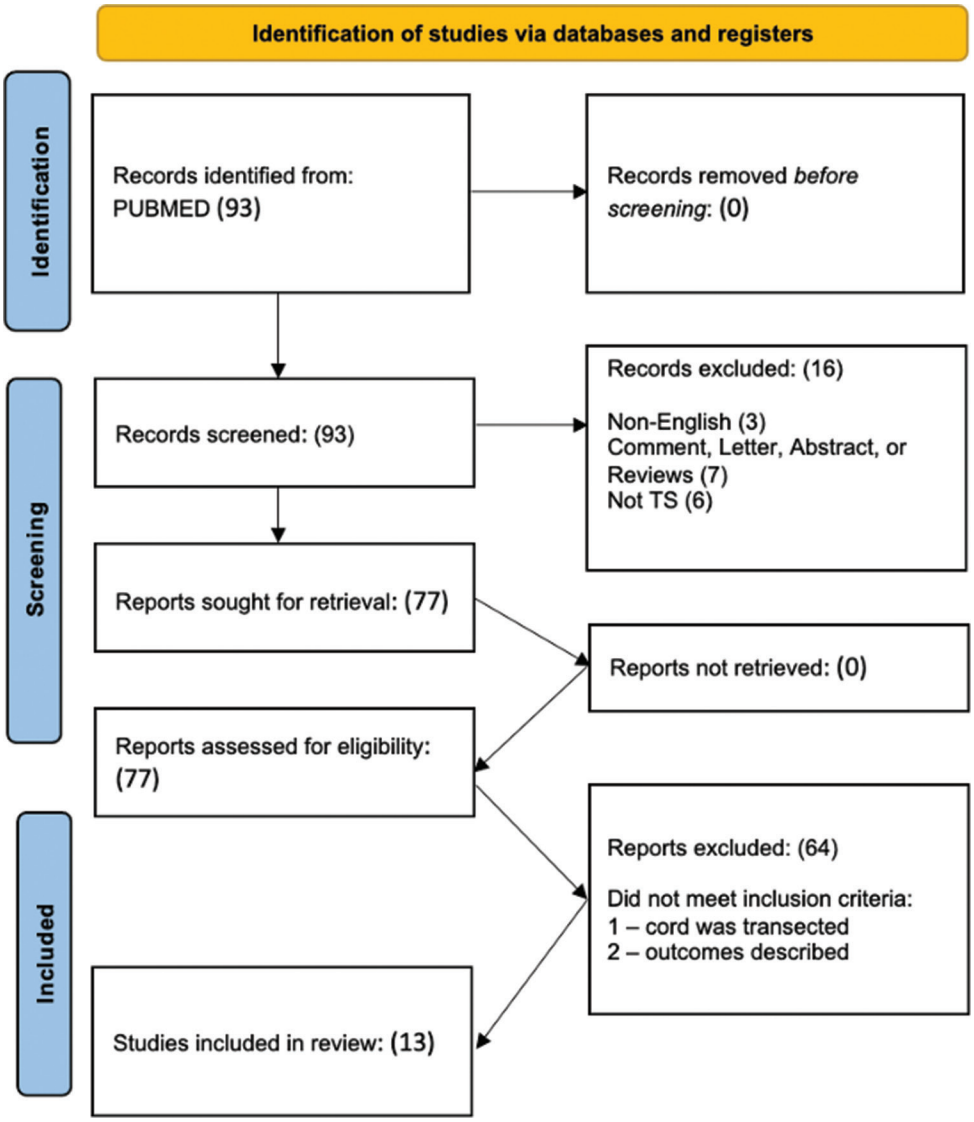

Figure 1: Preferred reporting items for systematic reviews and meta-analysis diagram depicting search query and final included articles. TS = Traumatic Spondyloptosis.

at the T6 level and a distal segment started at the T8 level [Figure 2b], with a separation of about 1.4 cm between the two segments. Given the patient's highly unstable spinal injury, she was taken to surgery for open thoracic spinal reduction and stabilization.

Operation

The patient was anesthetized and positioned prone on an open Jackson table (Jackson Spinal Table; Mizuho OSI, Union City, California, USA) with her arms at her sides. Neurophysiologic monitoring was used for this surgery. Prepositioning baseline showed no electrophysiologic signals distal to her injury. Partial closed reduction was performed under fluoroscopy using manual traction and manipulation of the patient's hips and shoulders. While performing the subperiosteal dissection, several areas of fascial disruption with brisk CSF leakage were encountered. Pedicle screws were placed at T2–T11 with intraoperative stereotactic navigation using the O arm (Medtronic; Minneapolis, Minnesota, USA). After the pedicle screws were placed, attention was turned to exploring the spinal canal. The posterior elements were completely disrupted at T7 and T8. A standard laminectomy was performed from T5 to T8 to expose the spinal canal and examine the dural tube. The dura was severely and circumferentially injured with a large defect of no intervening dura from approximately T6 to T7. From the disrupted ends of the dura, the injured thoracic nerve roots and the transected ends of the spinal cord are shown [Figure 3a]. The thoracic spinal cord was not in continuity. There was high-flow spinal

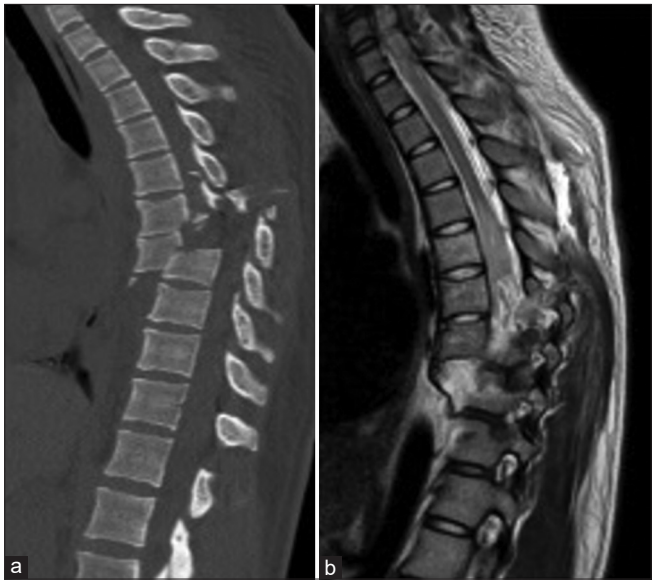

Figure 2: Sagittal reconstruction of thoracic computed tomography showing spondyloptosis of T7 on T8 with anterior and lateral subluxation of the T7 vertebral body compared to the T8 vertebral body (a). Sagittal T2-weighted thoracic magnetic resonance imaging showing the disruption and complete transection of the spinal cord at the T7–8 (b).

fluid leakage from the rostral end of the dural defect. Without proper control of this high-flow CSF leak, there could be impaired wound healing, meningitis, pseudomeningocele, and subdural hemorrhages. The laminectomy was extended both rostrally and caudally to identify dura amenable to repair. Because the defect was too large to adequately reconstruct and the spinal cord was completely transected and without neurologic function distal to T6, it was felt that the safest and most definitive way to stop the CSF leak and obtain watertight closure was to suture ligate the open end of the thecal sac of the rostral segment [Figure 3b]. Two 3-0 silk suture ties were placed next to each other and used to ligate the open end of the rostral dura. The closure was challenged by performing a Valsalva maneuver that confirmed no egress of CSF. The repair was reinforced with Adherus tissue sealant (Stryker; Kalamazoo, Michigan, USA).

Further reduction of the spine was attempted with the instrumentation. A quad-rod construct was utilized at T7–T8 due to the significant deformity and instability at this level. The posterior lateral fusion was then carried out in the standard fashion with local morselized autograft and allograft protein. The wound was closed in a standard multilayered fashion.

Postoperative course

The postoperative neurologic examination remained unchanged, with no neurologic function below the level of the spinal cord injury but preserved neurologic function above the level of the injury. She made a good recovery from her other traumatic injuries and had no issues from COVID infection. She suffered no CSF leakage or wound healing issues and was ultimately discharged from the hospital to spinal cord rehabilitation.

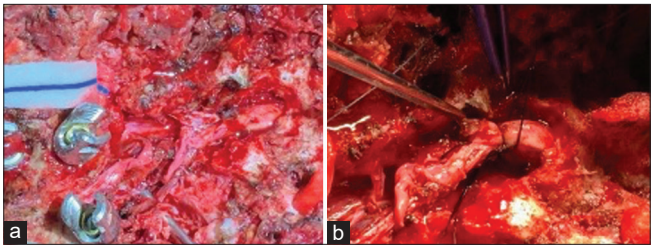

Figure 3: Intraoperative picture showing the extent of the traumatic damage to the dura and the transected spinal cord and thoracic nerve roots (a). Suture ligation of the rostral end of the thecal sac to obtain a water tight dural closure (b).

SYSTEMATIC REVIEW

Search results

Our search query returned 93 articles, of which 13 met the inclusion/exclusion criteria [Table 1]. Most articles focused on operative fixation and did not specify whether the dural defects were repaired. Eight of the 13 (61.5%) articles described the approach to dural repair.[7,9,12-14,22,30,31] These eight articles included 20 patients with complete cord transection.

Three of the 13 (23.1%) articles were from the USA.[10,29,30] Five (38.5%) were from India, and one (7.7%) was from Korea, Japan, Greece, Iran, and Turkey each.[1,3,9,12-14,16,22,28,32] Cases from Iran, Korea, India, Turkey, and USA described their approach to the dural defect.[7,9,12-14,16,22,30,31]

Repair of durotomy

The dura was deemed nonrepairable and therefore not repaired in two (20%) of the 20 cases.[9,16] The cord was transected at T7–8 level in one case and C5–6 level in the other.[7,16] There was no postoperative CSF complication in the case of thoracic spondyloptosis.[9] In the case of cervical spondyloptosis, postoperative CSF leak required a second operation during which the dura was sutured both cranially and caudally to the defect.[16] There were no further CSF leaks. Neither case had improvement in paraplegia or quadriplegia, respectively.[9,16]

Three of the included articles were published from the Department of Neurosurgery of the All India Institute of Medical Science.[12,13,22] These articles collectively

Table 1: Author, title, year, country of origin, injury characteristics, approach to dura repair and outcomes in patients included in the articles.

First Author	Title	Year (Country)	Level of Spondyloptosis	Level of Cord Transected	Dura Repair	Post-operative CSF Leak	Reported Outcomes
Fattahi A[9]	Traumatic thoracic spine spondyloptosis treated with spondylectomy and fusion	2018 (Iran)	T7-8	T7-8	Not repaired	Not described	Paraplegia
Kwun JW[16]	Management of Traumatic Cervical Spondyloptosis with an Unsealable Dura Tear: A Case Report	2021 (Korea)	C5-6	C5-6	Not repaired	Yes – required second operation in which the dura was sutured cranially and caudally to the defect	Quadriplegia
Garg M[12]	Management of Pediatric and Adolescent Traumatic Thoracolumbar Spondyloptosis	2022 (India)	1. L1-2 2. L4-5 3. T12-L1 4. L1-2 5. T11-12	1. L1-2 2. L4-5 3. T12-L1 4. L1-2 5. T11-12	Ligation with purse-string suture and reinforcement with fibrin glue of the proximal end	Not described	ASIA A
Mishra A[22]	Traumatic spondyloptosis: a series of 20 patients	2015 (India)	(n=7) Not specified	(n=7) Not specified	Ligation with purse-string suture and reinforcement with fibrin glue of the proximal end	1 of 7 patients had postoperative CSF leak	ASIA A
Garg M[13]	Traumatic Lateral Spondyloptosis: Case Series	2018 (India)	1. T6-7 2. L1-2 3. T12-L1	1. T6-7 2. L1-2 3. T12-L1	Ligation with purse-string suture and reinforcement with fibrin glue of the proximal end	Not described	ASIA A One patient died from bedsore complications
Chandrashekhara SH[3]	Unusual traumatic spondyloptosis causing complete transection of spinal cord	2011 (India)	1. L4-5 2. L3-4 3. T12-L1 4. T11-12	1. L3-4 (Low lying cord) 2. L3-4 (Complete cauda equina and nerve root transection) 3. T12-L1 4. T11-12	Not described	Not described	1. Mild improvement 2. paraplegia 3. paraplegia 4. paraplegia
Sasagawa T[28]	Traumatic spondyloptosis at the thoracolumbar junction in a patient with diffuse idiopathic skeletal hyperostosis: A case report	2020 (Japan)	T10-11	T10-11	Not described	Not described	ASIA A

(*Contd...*)

Table 1: (*Continued*).

First Author	Title	Year (Country)	Level of Spondyloptosis	Level of Cord Transected	Dura Repair	Post-operative CSF Leak	Reported Outcomes
Gabel BC[10]	Traumatic L5 Posterolateral Spondyloptosis: A Case Report and Review of the Literature	2015 (USA)	L5-S1	L5-S1 (Complete obliteration of the thecal sac)	Not described	Not described	Paraplegia
Hasturk AE[14]	Unusual traumatic midthoracic spondyloptosis and its surgical management: case report	2013 (Turkey)	T5-6	T5-6	Duraplasty	Not described	Paraplegia
Zygogiannis K[32]	Traumatic Fracture of the Thoracic Spine With Severe Posterolateral Dislocation: A Case Report	2022 (Greece)	T3-4	T3-4	Not described	Not described	Paraplegia, died in 1 year
Sekhon LH[29]	Surgical management of traumatic thoracic spondyloptosis: review of 2 cases	2007 (USA, Australia)	T12-L1	T12-L1	Muscle graft used to partially reconstruct the defect.	Not described	Paraplegia
Toms J[30]	Traumatic lumbar spondylolisthesis resulting in complete thoracic spinal cord avulsion: an unusual presentation	2018 (USA)	L3-4	T11-12	Dura was closed around caudal portion of defect	Cystic replacement of normal thecal sac within spinal canal	Paraplegia
Bailyan V[1]	Unusual cord transection in a patient with traumatic spondylolisthesis	2016 (India)	L2-3	T11-12	Not described	Not described	Not described

CSF: Cerebrospinal fluid, ASIA: American spinal injury association impairment scale

described 15 of the 20 (75%) of the total reported cases with complete cord transection. The dural defect was repaired with proximal ligation using a purse-string suture and reinforcement with fibrin glue.[12,13,22] Of all cases with this approach to dural repair, one patient has a postoperative CSF leak.[22] Management of this leak was not described, none of the patients had any improvement in neurologic function and remained ASIA A.[12,13,22]

One case describes the repair of a dural defect at T5–6 level with duraplasty and no postoperative CSF leak.[14] Muscle graft was used to partially repair the dural defect at T12–L1 level in Sekhon *et al.*'s article.[29] This article does not describe a postoperative CSF leak.[29] Toms *et al.*'s case report describes cord transection at T11–12 level.[30] In this case, the dura was closed at the caudal portion of the defect.[30] At 2 years follow-up, there was the cystic replacement of the normal thecal sac within the spinal canal cranial to the durotomy.[30]

DISCUSSION

TS is a rare but devastating injury that may occur throughout the spine.[8,9,13,18,24,25] The common causes in the reported literature are MVC and fall from a tree or building.[2,9,18,24] Associated complete cord transection with associated ASIA A neurologic examination has been reported in thirteen papers, but limited attention has been directed at the management of any associated disruption of the dura. However, without proper repair, the possibility of a high-flow CSF leak can cause significant morbidity and possibly mortality.[9,13,18,24,28,29]

A retrospective review by Luszczyk *et al.* analyzed 187 patients at a level 1 trauma center with traumatic dural tears.[20] They cite a 9.9% rate of dural tear in thoracic spinal fractures. This was higher than the rate of dural tear in the cervical spine (9.1%), but markedly lower than that in the lumbar spine (17.6%).[20] Of thoracic dural tears, 77.8% resulted from fracture or dislocation injuries, as seen in this case.[18] Another study reports an association between disruption of the ligamentum flavum and CSF leak with dural tear in the cervical spine.[17]

While the specific management of dural traumatic dural tear is case-specific, the general principles involve an attempt to create a watertight primary suture closure or repair using graft material that may be autograft from fascial lata or other sources, or synthetic dural substitute.[7,20] Fibrin glue or synthetic tissue sealants and placement of a lumbar drain or lumboperitoneal shunt may also be employed if a water-tight closure is difficult to achieve.[7,19,20] However, in cases of complete cord transection and spinal cord injury, durotomy may not always be repaired.[9,16] One case of traumatic TS with complete cord transection at T7–8 reports success with the insertion of a drain without dural repair.[9] However, Kwun *et al.* report a postoperative CSF leak that required reoperation to ligate the thecal sac both cranially and caudally to the defect.[16] In one review, there were no differences in rates of complications between those who underwent dural repair and those who did not.[20] Of note, the cord was transected in only 48.5% of the included patients, and the severity of the CSF leak in patients was not fully described.[20] The size of the durotomy and characteristics of the torn dural edges may be important characteristics to consider when deciding whether to repair the dural defect and prevent a CSF leak. In fact, durotomy with high-flow CSF leak has been associated with persistent CSF leak necessitating irrigation and debridement with operative wound re-closure, pseudomeningocele, meningitis, and postural headaches from persistent CSF leak in the literature.[11,20] Long-term, a pseudomeningocele may progress to subdural hygromas and communicating hydrocephalus requiring ventriculoperitoneal shunting.[6]

In the case presented in this report, the dural disruption was so extensive that there was a high level of concern for complications associated with persisting, high-flow CSF leak, and ligation of the dura was felt to be the most effective method to avoid complications. Since CSF is produced in the ventricles of the brain proximal to the complete spinal cord transection, we felt thecal sac ligation at the proximal end was sufficient to prevent CSF leakage. Closure of the caudal portion of the defect has been reported and was associated with cystic replacement of the normal thecal sac within the spinal canal. There is precedent for ligation of the dural sac in very specific situations in which satisfactory alternatives are lacking, as in instances of resection of malignancy such as sacral chordoma in which margins are an important goal and very distal.[15,26] This technique has been described with success in reported cases from India but is, to our knowledge, the first such report from the United States. We emphasize that this technique is limited to instances of gross transection of the spinal cord with absolutely no neurologic function below the level of injury and would not be appropriate in instances wherein there is any anatomic continuity of the spinal cord, even if there is no appreciated neurological function on examination.

CONCLUSION

Suture ligation of the thecal sac in the setting of traumatic complete spinal cord transection with significant dural disruption has been described in the international literature and is a safe and successful technique to prevent complications associated with persisting high-flow CSF leakage. To the best of our knowledge, this is the first report of this technique undertaken in the United States.

Declaration of patient consent

Patients' consent not required as patients' identities were not disclosed or compromised.

Financial support and sponsorship

Nil.

Conflicts of interest

There are no conflicts of interest.

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How to cite this article: Vazquez S, Houten JK, Stadlan ZT, Greisman JD, Vaserman G, Spirollari E, *et al*. Thecal sac ligation in the setting of thoracic spondyloptosis with complete cord transection. Surg Neurol Int 2023;14:304.

SUPPLEMENTARY MATERIAL

Supplementary Table 1: Joanna Briggs critical appraisal checklist for case series.

	Q1	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Total (%)	Risk of bias
Fattahi and Daneshi (2018)	Y	Y	Y	N	Y	Y	Y	Y	Y	NA	89	Low
Kwun *et al.* (2021)	Y	Y	Y	N	Y	Y	Y	Y	Y	NA	89	Low
Garg *et al.* (2022)	Y	Y	Y	Y	Y	Y	Y	N	Y	NA	89	Low
Mishra *et al.* (2015)	Y	Y	Y	Y	Y	Y	Y	Y	Y	NA	100	Low
Garg *et al.* (2018)	Y	Y	Y	Y	Y	Y	Y	Y	Y	NA	100	Low
Chandrashekhara (2011)	Y	Y	Y	UN	Y	Y	Y	Y	Y	NA	89	Low
Sasagawa (2020)	Y	Y	Y	N	Y	Y	Y	Y	Y	NA	89	Low
Gabel *et al.* (2015)	Y	Y	Y	N	Y	Y	Y	Y	Y	NA	89	Low
Hasturk *et al.* (2013)	Y	Y	Y	N	Y	Y	Y	Y	Y	NA	89	Low
Zygogiannis (2022)	Y	Y	Y	N	Y	Y	Y	Y	Y	NA	89	Low
Sekhon *et al.* (2007)	Y	Y	Y	UN	Y	Y	Y	Y	Y	NA	89	Low
Toms *et al.* (2018)	Y	Y	Y	N	Y	Y	Y	Y	Y	NA	89	Low
Bailyan (2016)	Y	Y	Y	N	Y	Y	Y	N	Y	NA	78	Low

Results from the JBI Critical Appraisal Checklist for case series. Y: Yes, N: No, UN: Unclear, NA: Not available. Total: Percentage of yes answers, where NA did not contribute to the overall sum. Q1-Q10 refer to the questions included in the JBI Critical Appraisal Checklist for case series.