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Tethered Cord Syndrome and Split Cord Malformation: A Retrospective Review with Advanced Surgical Technique

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Research Article

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Abstract

Tethered cord syndrome is a rare condition resulting from compression due to congenital anomalies. This study aims to report surgical findings and outcomes in managing tethered cord syndrome. We retrospectively studied 48 patients diagnosed between October 2009 and December 2017. Demographic, clinical, radiological, and surgical details were reviewed, including neurological, urological, and orthopedic findings. 72.9% pediatric, and 27.1% adults. Neurologic, skin, and orthopedic manifestations were observed in 35%, 31%, and 48% of patients. Lower limb weakness was most common (94%). 44% had a split cord, with type I more common in pediatric patients and type II more common in adults. All underwent surgical intervention, showing significant long-term improvement in neurological, orthopedic, and urological symptoms. Only 8% had moderate improvement; two cases required redoing untethering and adhesiolysis. Timely surgical intervention, including untethering, decompression, and anomaly resection, yields significant neurological outcomes in tethered cord syndrome management.

1 Introduction

Tethered cord syndrome (TCS) encompasses a constellation of congenital anomalies that lead to the development of diverse clinical presentations in different age groups. It is important to define the tethered cord as being under mechanical tension. Usually, this manifests with progressive motor and sensory dysfunction with or without incontinence, especially in children. Excessive tension on the lumbosacral cord, by virtue of specific attachments in the spinal canal, limits its movement [1-3].

Hence, TCS can be either isolated or secondary to other associated congenital anomalies, such as split cord malformation (SCM), intra-dural lipoma, lipomyelomeningocele, neurenteric cyst, and dermal sinus tract [1]. Split cord malformation is a rare spinal cord anomaly due to a defect in the primary neurulation between days 24 and 28 of embryonic life [4, 5]. The etiology is not well defined; however, the unified theory proposes an embryogenetic mechanism of developmental error during the neural tube [6, 7]. This theory identifies two major subtypes of SCM. In Type I, the spinal cord is split into two hemicords by an osseous or cartilaginous septum, where each has its own dura. In type II, the two hemicords are separated by a fibrous septum and are contained in one single dura. The wide spectrum of clinical presentation in patients with TCS warrants a multidisciplinary approach. On routine examination, signs of a tuft of hair, dermal sinus, dimple, and cutaneous hemangioma should be noted. These signs are highly associated with underlying spinal anomalies [1, 2].

The treatment of TCS, especially in its isolated form, is controversial. Surgical management is gaining preference, even for adult patients [8, 9]. Furthermore, early antenatal diagnosis by detailed morphology scan in 1st and 2nd trimesters of pregnancy might be helpful in the planning for the proper surgical management and caring in the post-natal period [10]. This retrospective review presents the clinical and radiological findings, surgical interventions, and outcomes of patients diagnosed with both subtypes of TCS. We highlight the importance of decompressive laminectomy or laminoplasty, in addition to surgical treatment for spinal cord malformations, to improve outcomes and prevent neurological deterioration.

2 Materials and Methods2.1 Study Design and Settings:

This study was approved by the Institutional Research Board at Jordan University of Science and Technology (JUST) and King Abdullah University of Science and Technology and finally approved by the University Review Committee for Research on Humans at JUST. The study group consists of 48 cases, they were treated for symptomatic split cord syndrome and tethered cord at King Abdullah University Hospital from December 2009 to December 2017. Untethering of the spinal cord, resection of diastematomyelia spur, resection of dermal sinus, reconstruction of the dura, and decompressive laminectomy or laminoplasty were carried out. We conducted a retrospective analysis of patients' demographics and clinical outcomes following surgery. Postoperative clinical assessments were performed at 2 weeks, 3, 6, 12, and 24 months, and up to 5 years to monitor progress. The inclusion criteria for our study comprised all cases with symptomatic tethered cords, diastematomyelia, and lipomyelomeningiocele, while the exclusion criteria included asymptomatic cases such as myelomeningocele, meningocele, and spinal cord tumors. Magnetic resonance imaging (MRI) was the modality of choice for evaluating TCS and identifying other associated anomalies such as SCM, dermal sinus tract, lipomatous tumors, and dermoid cysts as shown in Fig. 1, Fig. 2, and Fig. 3. It provided detailed information for surgical planning, including the conus level. All patients underwent whole spine MRI to exclude other congenital anomalies. In five cases with a split cord, a myelogram and postmyelogram CT scan were requested when the MRI failed to identify the spur's origin. The radiological criteria for inclusion were based on evidence of tethered cord, when conus medullaris ended below L3 in the pediatric group, and below L2 in the adult group, with thickened filum terminale. This was either isolated tethering or secondary to the associated congenital anomalies. None had evidence of Chiari malformation.

2.2 The Surgical Technique:

For all patients, the same surgeon performed the surgical technique in a consistent manner. The procedure involved using a microscope to make a skin incision at the lumbosacral level, with extension to the dermal opening if required. Following the wound and muscle strip opening, laminoplasty was performed in the pediatric age group. Subsequently, a durotomy was carried out and reflected. During the procedure, the diastematomyelia spur, conus medullaris, and filum terminale were identified first. The standard surgical technique included identification of diastematomyelia spur, dissection of the spur off adjacent neural tissues, followed by full resection of the spur from its origin. The identified filum terminale was dissected off nerve roots, coagulated from two sides before resection, and sent for histopathology for confirmation. During the surgical procedure, any neural compromise and tension on the spinal cord at the split level was avoided and full release of the spinal cord was ensured. The dura was reconstructed to cover the spinal cord and the laminae were replaced and fixed in situ. This was followed by wound closure in layers. In patients with kyphoscoliosis, spinal column rotation, and angulation led to additional mechanical compression against the spinal cord at the site of the deformity.

As a result, in 14 cases, the operation site was extended to perform decompressive laminectomy in adults or laminoplasty in children. This was done to relieve compression, to unrestricted movement of the untethered spinal cord. Postoperatively, all patients were required to stay on bed rest for 24 hours and were discharged from the hospital within a week once their wound was healing appropriately. Follow-up appointments were scheduled for all patients at 2 weeks, 3, 6, 12, and 24 months, and yearly for up to five years. Additionally, all patients were referred to a rehabilitation program upon discharge. In patients with intra-dural lipomatous mass and tethered cord, surgery was conducted in a similar fashion, where laminoplasty was performed, followed by midline durotomy, tacking up of dural flaps, followed by debulking of the lipomatous lesion and untethering of the spinal cord by cutting the thickened filum as shown in Fig. 4.

2.3 Statistical Analysis:

Data analyses were performed using the IBM Statistical Package for the Social Sciences (SPSS) software for Windows, version 26.0. Descriptive statistics were used to present the demographic and clinical information of the patients. Categorical data were presented by frequencies and percentages (%), while continuous data were presented by means ± standard deviations for continuous data.

3 Results

3.1 Patient Characteristics

The patient characteristics of the study population, which included 48 patients with tethered cord syndrome. All the patients' demographics and clinical characteristics were summarized in Table 1. Out of all the patients, 35 (72.9%) were pediatric patients with a mean age of 5.04 years ± 4.8, and 13 (27.1%) were adult patients with a mean age of 23.6 years ± 8.8. Most patients were female (71%) across the entire cohort, but males were more prevalent in the adult subgroup (62%) than in the pediatric subgroup (17%). 16 patients (35%) presented with neurologic manifestations, 15 patients (31%) with skin manifestations, and 23 patients (48%) with orthopedic deformities. The most common neurologic manifestation was lower limb weakness, which was present in 15 patients (94%). Bladder dysfunction was observed in 6 patients (38%), and bowel incontinence was observed in 3 patients (19%). Among skin manifestations, a soft mass was observed in 8 patients (53%), followed by tuft of hair in 9 patients (60%). Orthopedic deformities included scoliosis/kyphoscoliosis in 17 patients (74%) and foot deformities in 9 patients (19%). The anomalies associated with tethered cord syndrome are shown in Table 2, with a syrinx being present in 8 patients (17%), filum terminal in 9 patients (19%), and spinal lipoma in 9 patients (19%). The study found a split cord in 21 patients (44%), with type I split present in 10 patients (48%) and type II split present in 11 patients (52%). Type I split was more common among pediatric patients while type II split was the only type of split found among adult patients. 2 (10%) patients were found to harbor two levels of splitting. Most of the splits were in the lumbar region in 14 patients (67%). All patients underwent surgical intervention. Apart from recurrent UTIs, all children reported excellent improvement. Postoperatively, the patients had an uneventful course, only 7 patients

developed superficial wound infection and one of them resulted in meningitis. All patients recovered smoothly. In the long-term follow-up of a maximum of 5 years and a mean of 3 years. Most patients demonstrated a significant improvement in neurological, orthopedic, and urological symptoms. No new neurological deficits or worsening of the primary neurological status was observed except in one case which resulted in increased weakness as the power went down from a 4/5 to a 2-3/5 in lower limbs with no sphincteric deterioration, however, he displayed a significant improvement with rehabilitation and recovered into his baseline power in a year's time. On the 5-year follow-up, all cases were satisfied and displayed a significant improvement. Only 4 cases (8%) out of 48 had moderate improvement with two cases having re-tethering of the cord which required to redo the untethering and adhesiolysis. They later proved to have excellent improvement.

3.2 Tables

	All Patients (n = 48)	Pediatric (n = 35)	Adult (n = 13)	
Age (years), mean (SD)	10.07 (10.29)	5.04 (4.8)	23.6 (8.8)	
Gender n (%)				
Male	14 (29)	6 (17)	8 (62)	
Female	34 (71)	29 (83)	5 (38)	
Neurologic manifestations n (%)	16 (35)	9 (40)	7 (23)	
Lower limb weakness	15 (94)	8 (89)	7(100)	
Bladder dysfunction	6 (38)	4 (44)	2 (29)	
Bowel Incontinence	3 (19)	3 (33)	0 (0)	
Skin manifestations n (%)	15 (31)	14 (40)	1 (8)	
Tuft of hair	9 (60)	8 (57)	0 (0)	
Skin tag	1 (7)	1 (7)	0 (0)	
Dimple	1 (7)	0 (0)	1 (100)	
Hyperpigmentation	2 (13)	2 (14)	0	
Soft mass	8(53)	8 (57)	0	
Orthopedic deformities n (%)	23 (48)	18 (51)	5 (38)	
Scoliosis/Kyphoscoliosis	17 (74)	13 (72)	4 (80)	
Foot deformities	9 (19)	7 (39)	2 (40)	
¹ SD: Standard deviation, n: Number of patients				

Table 1 Patients' demographics and clinical characteristics.

	All Patients (n = 48)	Pediatric (n = 35)	Adult (n = 13)	
Syrinx	8 (17)	5 (14)	3 (23)	
Filum terminal	9 (19)	7 (20)	2 (15)	
Dermal sinus tract	3 (6)	3 (9)	0 (0)	
Spinal lipoma	9 (19)	7 (20)	2 (15)	
Meningocele	3 (6)	3 (9)	0 (0)	
Lipomeningocele	1 (2)	1 (3)	0 (0)	
Presence of split n (%)	21 (44)	16 (46)	5 (38)	
Type of split n (%)				
Type I split	10 (48)	10 (62)	0 (0)	
Type II split	11 (52)	6 (37)	5 (100)	
Location of split n (%)				
Thoracic	4 (19)	2 (12)	2 (40)	
Thoracolumbar	2 (9)	2 (12)	0 (0)	
Lumbar	14 (67)	11 (69)	3 (60)	
Lumbosacral	1 (5)	1 (6)	0 (0)	
¹ SD: Standard deviation, n: Number of patients				

Table 2 Associated anomalies with tethered cord syndrome

4 Discussion

Tethered cord syndrome (TCS) is a complex congenital anomaly associated with spinal dysraphism. The pathophysiological basis and natural history of TCS have been the subject of various theories. Huang et al. described TCS as a stretch-induced functional disorder of the spinal cord caused by excessive tension between the cord and dura matter [11]. The destruction of the filum terminal, leading to limited spinal cord movement within the lumbar cistern, is thought to play a significant role [3, 12]. In the secondary TCS, the associated congenital malformation may clarify the reason for cord tethering. The pathogenesis of the underlying embryological origin of neural tube defects and the associated spinal cord deformities are poorly understood. There is no single theory that could explain the exact mechanism by which these congenital defects develop. However, different theories have been proposed. In the unified theory of Mclone, the neurulation theory, proposed that the mechanism of NTD development is a defect in the apposition of neuroceles during primary neurulation, resulting in failure to maintain distention of the

primitive ventricular system [7]. Pang et al. proposed the unified theory of split cord malformation (SCM), which suggests that an accessory neurenteric canal formed by adhesions between the ectoderm and endoderm leads to the formation of an osseocartilaginous septum dividing the spinal canal [6]. On some extremely rare occasions, 3 spinal dysraphism are formed, in combination with Chiari type II malformation. Dhandapani et al. proposed the hypothesis of the cumulative cascading contiguous theory of the previous two [13]. Mahapatra introduced A new sub-classification of type I SCM based on the location of the bony spur responsible for the split, which can affect surgical dissection and outcome [14].

The well-known classification for SCM was first introduced by Pang as type I, namely diastematomyelia, where a dural-sheathed rigid osseous-cartilaginous median septum (bony spur) split the cord into two hemicords, each with its own dural sheath and central canal. While type II, diplomyelia, a nonrigid fibrous median septum split the cord into two hemicords, which are encased by one dural sheath [6]. This can be confirmed intraoperatively, as it is difficult to be diagnosed by preoperative imaging. This can be confirmed intraoperatively, as it is difficult to be diagnosed by preoperative imaging. Many risk factors could predispose to neural tube defects. Preconceptions of maternal nutrition, health, and psychological status may play a role. Jia et al. suggest that pre-gestational diabetes, low birth weight, and neonatal female gender as risk factors for NTD, while low maternal education and low paternal age are moderately associated with NTD [15]. Aydin et al., indicate that vitamin B12 supplementation and folates may help in lowering NTD frequency, with a possible relationship between increased NTD risk and factor V Leiden [16]. Maternal coffee consumption during pregnancy was not significantly associated with the occurrence of total NTD or the spina bifida subtype of NTD [17]. Others have concluded that Maternal H. pylori infection can increase the risk of occurrence of NTDs in newborns [18]. Ratan et al highlighted the possible role of the paternal side, where the father's serum homocysteine levels are considered an independent risk factor for NTD and other congenital anomalies [19]. Many risk factors could predispose to neural tube defects. Preconceptions of maternal nutrition, health, and psychological status may play a role. Jia et al. suggest that pre-gestational diabetes, low birth weight, and neonatal female gender as risk factors for NTD, while low maternal education and low paternal age are moderately associated with NTD [15]. Aydin et al., indicate that vitamin B12 supplementation and folates may help in lowering NTD frequency, with a possible relationship between increased NTD risk and factor V Leiden [16]. Maternal coffee consumption during pregnancy was not significantly associated with the occurrence of total NTD or the spina bifida subtype of NTD [17]. Others have concluded that Maternal H. pylori infection can increase the risk of occurrence of NTDs in newborns [18]. Ratan et al highlighted the possible role of the paternal side, where the father's serum homocysteine levels are considered an independent risk factor for NTD and other congenital anomalies [19].

In this cohort, the main presentation in the adult group of patients was back pain, progressive lower limb weakness and gait instability. Conversely, pediatric patients had more prevalent spinal column deformities and skin stigmata, being consistent with other reports [2]. This necessitated a multidisciplinary approach in the form of neurological, urological, pediatric, and orthopedic assessment as necessary. All adults with SCM in this study were type II. This could explain the late presentation. The caudal spinal cord would have been under tension due to possible filum fibrosis and thickening, growth

spur, increased physical activity, and development of spinal stenosis as part of the aging process [2]. Imaging modalities are important for the appropriate diagnosis of the tethered cord and secondary anomalies. The antenatal diagnosis is important for the early detection of the condition and for appropriate parents counseling, and discussion of the best management strategy. This, when performed efficiently, will decrease the burden on both caregivers and all medical teams, as they should be more prepared in advance. Hence, the surgical intervention could be planned and done with less complications and effort [10, 20]. Surgical management is the treatment of choice for symptomatic patients. Some studies recommend surgery for adult patients regardless of clinical status [21, 22]. However, other studies recommend observation for the asymptomatic patient with regular follow-up [5]. Surgical intervention remains the treatment of choice for symptomatic patients regardless the spinal cord at the site of kyphotic angulation, relieving mechanical compression and ensuring the free release of the spinal cord.

5 Conclusion

Tethered cord syndrome is a complex congenital pathology with diverse clinical presentations and progression patterns. A multidisciplinary approach involving various specialties is essential for effective management. Early detection and appropriate surgical intervention, particularly in the pediatric population, can prevent further neurological damage and lead to significant neurological and orthopedic improvement, resulting in satisfactory outcomes. Antenatal diagnosis, advanced imaging techniques, and regular follow-up play crucial roles in optimizing patient care. By employing these strategies, healthcare professionals can reduce the burden on caregivers and improve overall patient outcomes. Further research is needed to better understand the underlying pathogenesis and risk factors associated with TCS.

Declarations

Acknowledgements

Not Applicable

Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

Data Availability

The data used in this study is available upon request from the corresponding author.

Conflict of interest

The authors of this article have carefully considered any potential conflicts of interest and have found none to report. They have no relevant financial or non-financial interests that could impact the article's content, and they have no affiliations or involvement with any organizations with a financial or proprietary interest in the material discussed. The authors declare that they have no competing interests related to the manuscript's subject matter and certify that they have no ties to any entity that could present a conflict.

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Figures

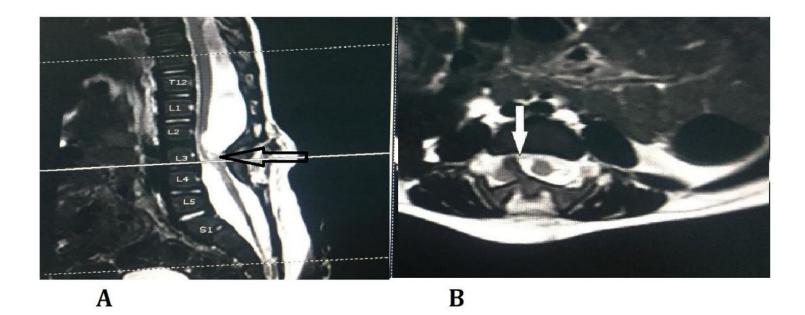
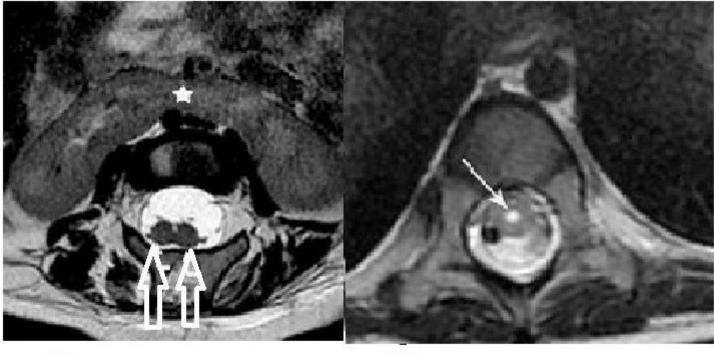


Figure 1

(A): Sagittal thoracolumbar MRI. (B): Axial MRI at L3 level showing the split spinal cord by the median septum into two hemicords (arrows), each with its own dural sheath (diastematomyelia).

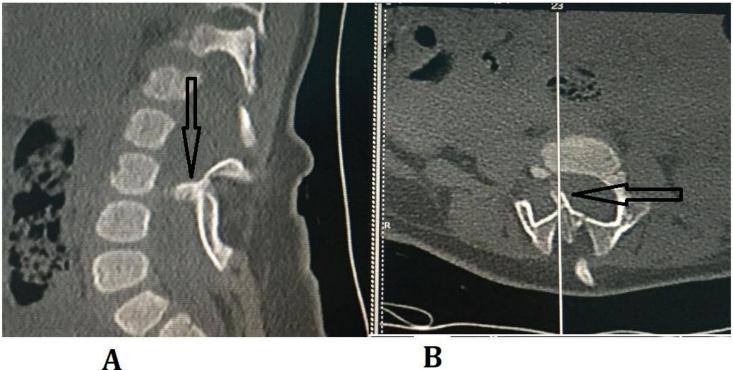




B

Figure 2

Axial T2 MRI of the same patient in Figure 1 (A): Shows splitting of the cord into two hemicords (diplomyelia) encased with same dural sheath (2 arrows). (B): Shows syringomyelia (one arrow).



A

Figure 3

Sagittal thoracolumbar spinal CT scan. (B): Axial CT scan at L2 level showing the bony spur projecting through the spinal canal at L2 level (arrows).

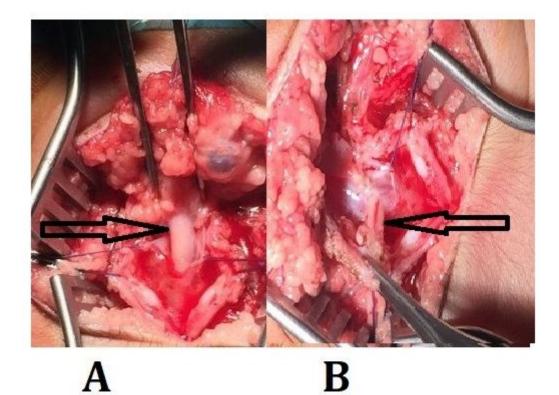


Figure 4

Intraoperative gross image. (A): Shows low insertion of spinal cord with tethered cord (B): Shows thickened filum terminale