IJMS
Vol 48, No 2, March 2023

Original Article

Comparison of Ultrasonographic Findings between Patients with Tethered Cord Syndrome and Healthy Children

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Received: 06 December 2021
Revised: 26 January 2022
Accepted: 23 March 2022

Abstract

Background: Tethered cord syndrome (TCS) is a type of occult spinal dysraphism, which necessitates early detection as an essential component of patient management in reducing complications. This study aimed to compare the findings of spinal cord ultrasonography between TCS patients and healthy individuals.

Methods: The current study is a case-control study of patients who were admitted to the Akbar and Ghaem Hospitals (Mashhad, Iran) in 2019. The study population comprised 30 children with TCS aged under two years old, and the control group included 34 healthy peers of the same age. The maximum distance of the spinal cord from the posterior canal wall was measured in millimeters using ultrasonography. Demographic and sonographic findings of each participant were recorded in checklists, which were then entered into SPSS software. P values less than 0.05 were considered statistically significant.

Results: The study included 30 children with TCS and 34 healthy individuals with a mean age of 7.67±6.39 months. TCS patients had a significantly shorter maximum distance of the spinal cord from the posterior wall of the spinal canal than the control group (1.75±0.62 mm vs. 2.79±0.76, P<0.001). After performing corrective surgery, the TCS patients indicated significant improvement in this interval (1.57±0.54 mm to 2.95±0.49 mm, respectively, P=0.001).

Conclusion: In comparison to children without TCS, the spinal cord was substantially closer to the posterior canal wall in TCS patients. However, these outcomes were improved significantly in patients after surgery.

Please cite this article as: Keykhosravi E, Faravani E, Dehghani Dashtabi S, Emadzadeh M, Alamdaran SA, Aminzadeh B. Comparison of Ultrasonographic Findings between Patients with Tethered Cord Syndrome and Healthy Children. Iran J Med Sci. 2023;48(2):130-136. doi: 10.30476/IJMS.2022.93848.2517.

Keywords ● Neural tube defects ● Spinal dysraphism ● Ultrasonography

What's Known

- Tethered cord syndrome (TCS) is a neurological disorder that restricts spinal cord movements within the spinal canal.
- For TCS patients, early corrective surgery prevents the progression of related symptoms.

What's New

- The current modalities do not provide an accurate factor for diagnosing postoperative relapse. The success of corrective surgeries can be measured utilizing ultrasound from the spinal cord to the posterior wall of the spinal canal in a prone position in TCS patients and healthy controls.

Introduction

Tethered cord syndrome (TCS), also known as a neuro-orthopedic syndrome, is a constellation of neurological disorders that restrict the movements of the spinal cord inside the spinal canal due to mechanical traction,[1] causing various orthopedic, urologic, or dermatologic manifestations.[2-4] TCS clinical symptoms are extensive and differ with the age of diagnosis and the primary etiology. The primary method for determining TCS is physical

examination.[5] The most typical TCS presentations include cutaneous symptoms, neurological impairments, pain, and orthopedic deformities. These presentations change depending on age and the underlying pathologic condition.[6]

Lumbosacral magnetic resonance imaging (MRI) of the spine is the gold standard imaging modality in assessing TCS patients.[7-9] Nonetheless, ultrasound can also be used to diagnose and evaluate TCS, as well as to monitor TCS-related alterations in the urinary system, such as bladder thickening (indicating detrusor hypertrophy) or atrophy of kidneys.[10] The prognosis depends on the severity and location of the anomaly. There were reports of asymptomatic patients with radiographic regression. However, almost 75% of the patients experienced a long-term intensification of symptoms and neural injuries. A delayed diagnosis and course of TCS treatment may cause various neurologic symptoms and sphincter complications. Even after corrective surgery, symptoms may not resolve completely, and there is always a chance of recurrence, which necessitates postoperative neurological and radiological follow-up observations.[11-13] Given this, as well as the fact that other spinal abnormalities are frequently present in TCS patients, a need for determining the effectiveness of current diagnostic measures becomes more evident.

The position of the conus medullaris is the most important MRI and ultrasound observation in the diagnosis of the tethered cord. The aim of TCS surgery is to release the spinal cord by making an incision on the filum terminale. Throughout the surgery, it is critical to appropriately recognize the filum terminale and sacral rootlets. Through the TCS surgery, the scar from the previous closure will be opened down to the overlaying dura mater. A minor part of the laminae may be removed to gain better contact or decompress the spinal cord and cauda equina. Then, the dura is opened, and the spinal cord and rootlets are mildly separated from the scarred attachments to the surrounding dura.[14] On the other hand, since corrective surgery does not alter this position by restoring it to normal values, the conus medullaris cannot be used to assess postoperative retethering. Currently, imaging techniques are not much efficient in diagnosing retethering. Thus, physicians rely heavily on indirect symptoms, such as failure to resolve or recurrence of neurological symptoms. Consequently, current modalities do not provide an accurate factor for diagnosing postoperative relapse.

The identification of imaging techniques efficient in diagnosing retethering can help in the earlier and more reliable detection of recurrence. One of these techniques is ultrasound, which has the advantage of evaluating a child's spine in different conditions without the need for anesthesia. One of the factors that can be measured in ultrasound is the distance between the spinal cord and the posterior spinal canal, which is reduced in patients with TCS due to the spinal cord being pulled down.[15] Therefore, finding new quantitative imaging parameters can help evaluate the success of corrective surgeries while also providing better means of diagnosis for retethering.

Accordingly, this study was conducted to compare the distance of the spinal cord from the posterior wall of the spinal canal in prone positions between TCS patients and healthy controls using B-mode ultrasound. It also aimed to evaluate the ultrasound findings in a group of patients following corrective surgery.

Materials and Methods

This case-control study was conducted at the Radiology Department and Pediatric Ultrasound Clinics of Ghaem and Akbar Children's Hospitals (Mashhad, Iran), in 2019. The study was approved by the Ethics Committee of Mashhad University of Medical Sciences (IR.MUMS.MEDICAL.REC.1397.210). Before a patient could be enrolled in the study, they were informed about the goals of the research as well as the whole process of the study. Written informed consent was obtained from the patients' parents or their guardians. All the participants were informed that they could withdraw from the study at any time during the research process. The required information was gathered anonymously using sequential numbers, and the patients' identities were kept completely confidential.

Participants

The statistical population of this case-control study included all patients who were referred to the Neurosurgery Clinics of Ghaem and Akbar Children's Hospitals in 2019. The samples were selected using a census sampling method. The children under the age of two, who were referred to and diagnosed with TCS by an expert neurosurgeon were eligible patients.

Magnetic resonance imaging is the gold standard for determining the various causes of TCS. Therefore, TCS was diagnosed based on comprehensive clinical evaluations and confirmed by MRI (based on low-lying conus medullaris and a filum terminal measurement of more than 2 mm).[7, 16] Patients with open neural tube defects (NTD) or spinal canal masses, as

well as those who refused to participate in the study, were excluded. During this period, a total of 33 patients were diagnosed with TCS, among which three cases were excluded due to having an open NTD (n=1) and spinal canal mass (lipoma) (n=2).

The control group also included a group of healthy children (n=34) of the same age range who were referred to the Radiology Department and Pediatric Ultrasound Clinics of Ghaem and Akbar Children's Hospitals for various unrelated reasons and were found to be normal in both their ultrasound and clinical examinations.

Data Collection Procedure

The checklists were used to gather demographic information, such as age and sex, as well as personal and clinical features. Following that, a radiologist with experience in pediatric ultrasonography, who was blind to the MRI results of the participants, performed the ultrasound scans on each participant. Thirteen patients with TCS were accessible for post-operative evaluation, and the effects of corrective surgery on the research variables in these patients were also evaluated.

The patients were placed in a prone position and a B-mode gray-scale ultrasound examination of the spinal cord was performed using a Samsung WS80A device (Samsung Electronics, Korea) equipped with an 8-12 MHz linear-array transducer.

The main outcome of this study was the maximum distance of the spinal cord from the posterior wall of the spinal canal. The maximum distance of the spinal cord from the posterior wall of the spinal canal at T12-L2 levels (except the inflated conus medullaris region) was measured using B-mode ultrasonography. B-mode ultrasonography was also used to determine the endpoint location of conus medullaris, which was then recorded in both groups.

Statistical Analysis

All statistical analyses were performed with the statistical package for social sciences, version 23.0 (SPSS Inc., Chicago, IL, USA). The Kolmogorov-Smirnov test and histograms were used to determine the normality of data distributions. Mean±SD was used to express normally distributed quantitative variables, whereas median and interquartile range (IQR) were employed to describe non-normally distributed quantitative variables. The Chi square test, Mann-Whitney test, and independent samples *t* test were used to compare the data between the two groups. Paired samples *t* test was used to compare preoperative and postoperative data. P<0.05 was considered statistically significant for all statistical tests.

Results

This study included 33 children under the age of two, who were diagnosed with TCS. However, three children were excluded; one patient due to open NTD and two other subjects due to spinal canal mass. Therefore, the data from 30 cases were finally analyzed. Thirteen cases were followed up, and the results of the ultrasound examination of the spinal cord were compared to those obtained before the surgery. The control group consisted of a group of 34 peers of the same age, and sex, with no history of TCS. This study included 30 (46.9%) males and 34 (53.1%) females. The participants had a mean age of 7.67±6.39 months, ranging from three days to 23 months.

Table 1 compares the baseline characteristics of healthy children with TCS patients. There was no significant difference in sex or age between the two groups, P>0.301 and P>0.608, respectively.

Based on the B-mode ultrasound, the location of conus medullaris is presented in table 2. In the healthy group, the location of conus medullaris varied between L1 and L2 vertebral bodies; however, it was most commonly found at the L1-L2 interspace and L1 level. In the TCS group, nevertheless, the L4 level was the most common site where conus medullaris was located, and the range expanded between L3 and L5 vertebrae.

The primary outcome of this study was comparing the TCS patients with healthy controls. The maximum distances of the spinal cord from the posterior wall of the canal were estimated at 2.79±0.76 mm and 1.75±0.62 mm in the normal and TCS groups, respectively, which was significantly lower in TCS patients than in the control group (P<0.001) (figure 1).

Table 1: Basic characteristics of the normal and tethered cord syndrome groups

Variable		Normal n=34	TCS n=30	P value
Age (months)	Median (IQR)	6.00 (2.00-13.25)	6.50 (3.75-10.25)	0.608*
Sex N (%)	Male	18 (52.9%)	12 (40.0%)	0.301**
	Female	16 (47.1%)	18 (60.0%)	

IQR: Interquartile range; TCS: Tethered cord syndrome; *Mann-Whitney test; **Chi square test

Table 2: Conus medullaris position in the normal and tethered cord syndrome groups

Endpoint location of conus medullaris	Normal (n=34) N (%)	TCS (n=30) N (%)
L1 vertebral body	14(41.2)	-
L1-L2 interspace	14 (41.2)	-
L2 vertebral body	6 (17.6)	-
L3 vertebral body	-	2 (6.7)
L3-L4 interspace	-	5 (16.7)
L4 vertebral body	-	16 (53.3)
L5 vertebral body	-	7 (23.3)

TCS: Tethered cord syndrome

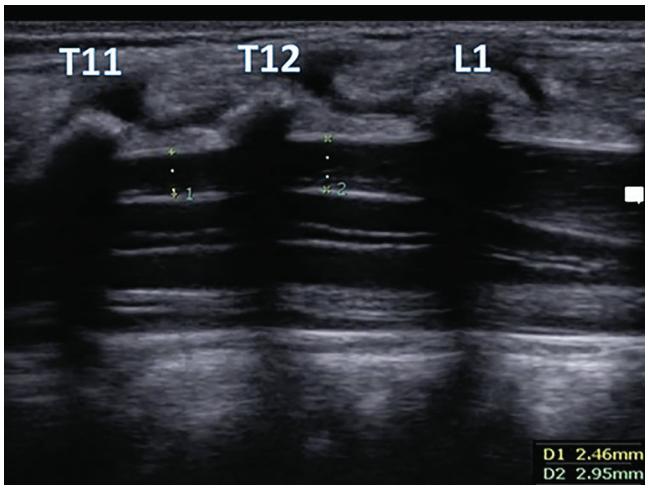

Figure 1: An ultrasound image shows the maximum distance of the spinal cord from the posterior wall of the canal between T12 and L1(2.95 mm) in a healthy infant.

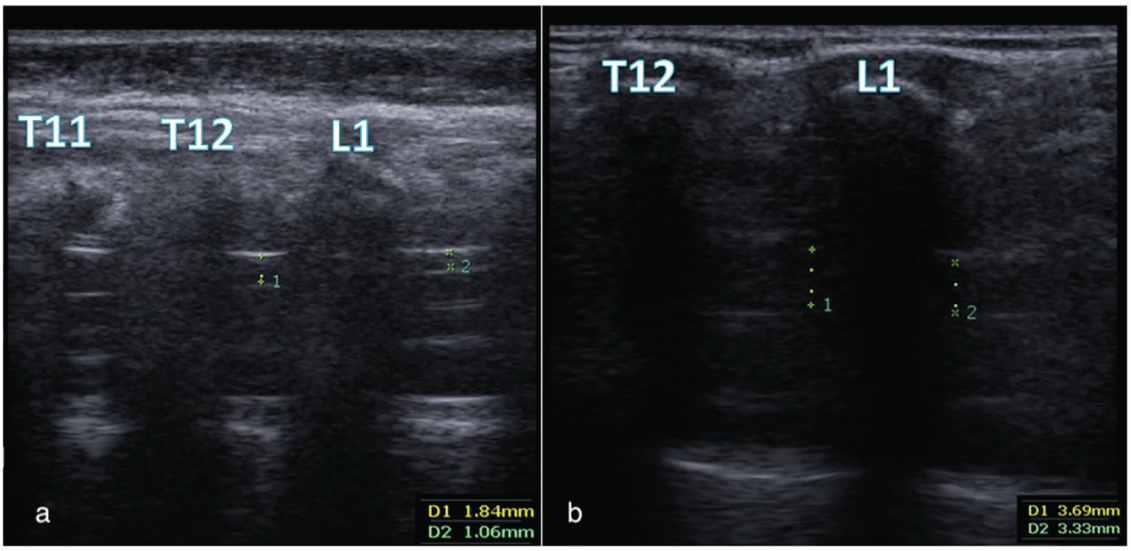

Figure 2: Ultrasound images show the maximum distance of the spinal cord from the posterior wall of the canal between T12 and L1 in a patient with tethered cord syndrome before (a) (1.84 mm) and after (b) (3.69 mm) the corrective surgery.

Postoperative information was accessible for only 13 patients. As shown in figure 2, the mean value for the maximum distance of the spinal cord from the posterior wall of the spinal canal was significantly increased following the surgery, 1.57±0.54 mm and 2.95±0.49 mm preoperatively and postoperatively, respectively (P=0.001).

Discussion

This case-control study compared the ultrasound findings between TCS children and healthy children of the same age and sex. The maximum distance of the spinal cord from the posterior wall of the canal varied significantly between the two groups. Consequently, the

TCS group had significantly lower values. These findings were also compared with the postoperative values, which showed a significant improvement after the corrective surgery. The implications of these findings can affect the way TCS patients are diagnosed and followed up after corrective surgery.

In the healthy group, the location of conus medullaris varied between L1 and L2 vertebral bodies; nonetheless, it was most commonly found at the L1-L2 interspace (41.2%) or L1 level (41.2%). This was fairly in line with the results of a previous study on the location of the distal part of the spine, which determined the normal range for the position of the conus medullaris by evaluating 26 healthy neonates under the age of three months. The position of the conus medullaris apparently ranged from the superior border of the L1 vertebra to the superior border of the L3 vertebra, with the mean position being midway between the L1-L2 interspace and the inferior border of the L2 body, which was almost similar to what was found in the present study.[15] As previously stated, the conus medullaris position does not return to the normal range after corrective surgery. Therefore, it is only effective in the primary diagnosis of tethering and is of no use in subsequent evaluations of retethering.

To the best of our knowledge, few studies were conducted to introduce new quantitative ultrasound findings that might be effective in diagnosing postoperative tethering recurrence. Lam and others introduced ultrasound as a practical modality in the examination of suspected TCS patients in 108 neurologically healthy children and compared the results with 16 children having TCS. They also measured the posterior and anterior subarachnoid space interval at the L5-S1 level below the conus medullaris (using B-mode). In addition, cerebrospinal fluid pulsation of the cauda equina was assessed (using M-mode). According to Lam and others, TCS might be indicated by a posterior subarachnoid space/anterior subarachnoid space ratio of ≤1. Furthermore, in a symptomatic patient, any oscillation amplitude of less than 0.3 mm can be a sign of TCS. Their findings showed that the spinal cord was significantly closer to the posterior spinal canal wall in TCS patients than in healthy children. The cord should be closer to the anterior spinal canal wall; in other words, the posterior subarachnoid space/anterior subarachnoid space ratio should be more than 1.[17] The findings of the previously mentioned study were consistent with those of the current study.

In the present study, the distance of the spinal cord from the posterior wall of the spinal canal was presented quantitatively, using the ultrasound scan, as a simple, accessible, and quick modality.

Although the current study provided useful information for diagnosing TCS, it had some limitations. The results of this study should not be generalized to other populations, because it was a non-random, single-centered study. Since this was a case-control study, a connection could not be inferred. Thus, the assessment of diagnostic accuracy, positive predictive value, and negative predictive value of measured parameters was impossible. In this regard, further studies must be conducted with larger sample sizes to evaluate the effectiveness of these parameters in comparison to the preoperative and postoperative imaging results. Further evaluation of clinical symptoms, their impact on treatment response, and the relationship between them and imaging findings are all required. With a particular focus on different diagnostic methods, more research is required to determine the best modality for diagnosing TCS in neonates and infants.

Conclusion

The present study aimed to find new criteria in imaging for the diagnosis of TCS and retethering after surgery. In this study, the distance between the spinal cord and the posterior wall of the spinal canal was measured. Based on the findings of this study, this criterion showed a significant difference between healthy children and TCS patients. In addition, the infants' performance on this criterion improved after the surgery. It can be concluded that the above criteria can be helpful not only in diagnosing TCS but also to diagnose retethering after surgery. In this study, the patients were not followed up for a long time after the surgery. Future studies can determine the role of the aforementioned criteria based on long-term follow-up of patients and the relationship between the long-term recurrence of the disease and the criteria reported in ultrasound.

Acknowledgment

This work was supported by the Research Council of Mashhad University of Medical Sciences, Mashhad, Iran (code: 970025). The authors would like to thank the Clinical Research Development Unit of Ghaem Hospital for their assistance with data analysis.

Authors' Contribution

E.K: Data analyzing, drafting the manuscript and critical revision; E.F: Performing experiments, data acquisition, and critical revision; S.DD: Study design, drafting the manuscript and critical revision; M.E: Performing experiments, data acquisition, and critical revision; SA.A: Data acquisition, data analyzing, and critical revision; B.A: Study conception, and critical revision. All authors have read and approved the final manuscript and agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

Conflict of Interest: None declared.

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