



DOI: 10.5137/1019-5149.JTN.40660-22.2



Received: 12.05.2022 Accepted: 27.07.2022

Published Online: 27.02.2023

# **Changes in the Lumbosacral Angle After Spinal Cord** Untethering in 23 Children with Tethered Cord Syndrome

Bing XIA, Honagian WANG, Yingmei DONG, Wenjing WANG, Fuyun LIU, Weiming HU, Feipeng WANG

The Third Affiliated Hospital of Zhengzhou University, Department of Pediatric Orthopedics, Zhengzhou, China

Corresponding author: Bing XIA 🗵 1312755420@gg.com

# ABSTRACT

AIM: To analyze changes in the lumbosacral angle in children with tethered cord syndrome before and after spinal cord untethering surgery, and to determine the clinical value of such changes at the last follow-up.

MATERIAL and METHODS: We retrospectively analyzed 23 children over 5 years old who were treated with spinal cord untethering in our hospital from January 2010 to January 2021 and who had complete medical data. X-rays were used to examine the child's spine preoperatively, postoperatively, and at follow-up with frontal and lateral radiographs, and lumbosacral angle data were measured and analyzed.

RESULTS: A total of 23 children aged 5-14 years had their lumbosacral angles measured and analyzed with a postoperative followup of 12-48 months. The mean preoperative lumbosacral angle was 70.30 ± 9.04°, the mean postoperative lumbosacral angle was 63.34 ± 5.60°, and the mean lumbosacral angle at the last follow-up was 61.61 ± 9.14°. There was a statistically significant reduction in the lumbosacral angle in the children postoperatively and at the last follow-up compared to the preoperative period (p=0.002; p=0.001).

CONCLUSION: Spinal cord untethering can improve the inclination of the lumbosacral angle in children older than 5 years with tethered cord syndrome.

KEYWORDS: Children, Lumbosacral angle, Tethered cord syndrome, X-ray

# INTRODUCTION

he presence of physiological pronation of the lumbar segment of the normal spine leads the sacrum to assume a certain inclination angle, and this inclination angle promotes the formation of the lumbosacral angle (LSA). Changes in the LSA directly affect the physiological curvature of the lumbar spine and the lumbosacral tilt angle, causing the lumbar spine to lose its original stability, which in turn leads to lumbar degenerative disease and lower back pain.

In tethered cord syndrome (TCS), the spinal cord is stretched taut and the spine is twisted, and the lumbosacral vertebrae are compressed forward by the accumulation of subcutaneous fat in the child, which can lead to an increase in the concurrent LSA due to the effect of asymmetric mechanical muscle tension (3). The degree of sacral tilt relative to the horizontal will become more and more serious with the age of the child (8), which may lead to increased neuropathy such as anal/bladder dysfunction, sensorimotor dysfunction, and low back pain (9). The current treatment for TCS is spinal cord untethering, and patients with either secondary or recurrent TCS can also benefit from surgical untethering (17). However, changes in LSA after spinal cord untethering have rarely been reported. In this study, we analyzed changes in the LSA of children with TCS over 5 years of age after spinal cord untethering and at the last follow-up compared to their preoperative period, thus providing a theoretical basis for clinical prognosis.

Bing XIA

0 : 0000-0002-2501-5159 Hongqian WANG (): 0000-0001-7888-0261 Yingmei DONG 0: 0000-0003-2334-2954

Fuyun LIU Weiming HU

Wenjing WANG (D): 0000-0002-4785-1246 0000-0002-9570-5796 000-0002-7224-8975

Feipeng WANG (0: 0000-0002-1672-3823

This study was approved by the Ethics Committee of the Third Affiliated Hospital of Zhengzhou University (Approval number: 2021-008-02).

# Patients

We retrospectively analyzed the preoperative, postoperative, and final follow-up changes in the LSA in children aged 5-14 years with TCS who were diagnosed and treated with spinal cord untethering at our hospital from January 2010 to January 2021. The inclusion criteria were a diagnosis of spinal TCS confirmed by medical history, physical examination, preoperative MRI examination, whole spine frontal and lateral X-ray examination, and urodynamic examination; clinical symptoms of spinal cord tethering, contraindications to surgery were excluded, and spinal cord untethering was performed in our hospital; and postoperative clinical data were followed up for more than 12 months. The exclusion criteria were a history of lumbar spine trauma; abnormal spine development such as congenital scoliosis, anterior/retroflexion, spinal tumor, etc.; history of multiple surgeries on the lumbosacral region; and incomplete clinical data and imaging data.

#### **Clinical Manifestations**

All children had preoperative MRI examinations that confirmed low tethered conus medullaris, and whole spine X-ray examinations were performed to exclude spinal deformities. Clinical symptoms and signs included lumbosacral hair, masses, and skin pits (7 cases), decreased muscle strength or muscle atrophy in the lower limbs bilaterally or unilaterally (7 cases), decreased sensation or loss of sensation in the lower limbs bilaterally or unilaterally (3 cases), abnormal urination and defecation (10 cases), clubfoot bilaterally or unilaterally (6 cases), and pain in the lumbosacral region or lower limbs (3 cases).

#### **Study Methods**

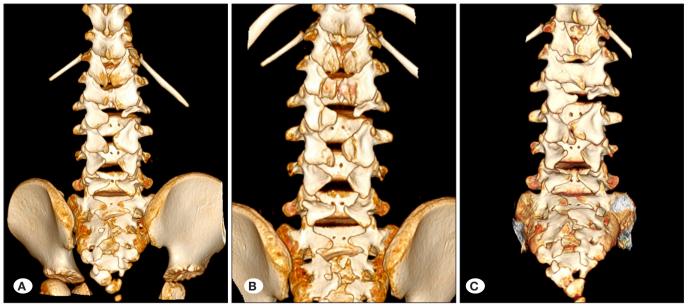
After admission to the hospital and completion of relevant investigations, spinal cord untethering was performed by the same experienced doctor. The spinal cord tethering was released by opening the laminae of the diseased segment using the double-opening/single-opening technique, and the laminae were repositioned and fixed with an absorbable internal fixation system after surgery to maintain the integrity of the posterior spinal column and the incision was closed layer by layer (Figure 1). Changes in LSA in lateral spinal radiographs were measured and analyzed preoperatively, postoperatively, and at the last follow-up.

#### **LSA Measurements**

The LSA was measured by drawing a straight line perpendicular to the tangent to the anterior surface of the third lumbar vertebra and drawing a straight line perpendicular to the sacral line, which is perpendicular to the line connecting the midpoint of the anterior edge of the first sacral vertebra and the anterior edge of the second sacral vertebra, and the angle formed by the two lines was the LSA (15,16).

#### **Statistical Analysis**

The measurement data were expressed as mean  $\pm$  standard deviation. SPSS 25.0 software was used for statistical data analysis. The Wilcoxon signed rank test and Mann–Whitney *U*-test were used to test the significance of the variables, and Spearman correlation analysis was used to assess the correlation of variables. Differences were considered statistically significant at p<0.05.



**Figure 1:** Case 10, male, 7 years old. Preoperative **(A)**, postoperative **(B)**, and 36-month postoperative follow-up **(C)** CT 3D reconstructions. Intraoperatively, the lamina below L1 was seen to be unfused, the T12-L1 lamina was opened with a double door, the right side of L2 and the left side of L3 were opened with a single door, and the dura was seen to be pulled by abnormal fiber bundles between the T12-L1 lamina. After spinal cord untethering the T12-L3 lamina was shaped and fixed with the L1 absorbable nail plate system.

## RESULTS

We studied a total of 23 children with TCS (10 males and 13 females) with a mean age of 9.00  $\pm$  3.09 years and a mean postoperative follow-up of 19.26  $\pm$  9.02 months (Table I). The mean preoperative LSA was 70.30  $\pm$  9.04°, the mean postoperative LSA was 63.34  $\pm$  5.60°, and the mean postoperative LSA at the last follow-up was 61.61  $\pm$  9.14°. The angles were significantly different (p=0.002; p=0.001) both postoperatively and at the last follow-up compared to preoperatively (Table II). There was no statistical difference between postoperative and final follow-up (p=0.346).

We divided the children into two groups according to the age of puberty at around 10 years, including 13 cases younger than 10 years and 10 cases 10 years of age and older. Statistical analysis of the change in LSA in both groups showed that the angle in the group less than 10 years of age was statistically significant after surgery and at the last follow-up compared with the angle before surgery (p=0.013; p=0.041). There was no statistical difference in the group older than 10 years of age after surgery compared with the angle before surgery (p=0.058), but there was a significant difference at the last follow-up compared to preoperatively (p=0.012) (Table II). Statistical analysis of the postoperative LSA correction rates in the two groups separately showed no statistically significant difference between the two groups (U=55.000; p=0.535). Figure 2 shows the LSA measurements (preoperative, postoperative, and final follow-up) of a 7-year-old boy with TCS.

Table I: Clinical Data of the Included Patients

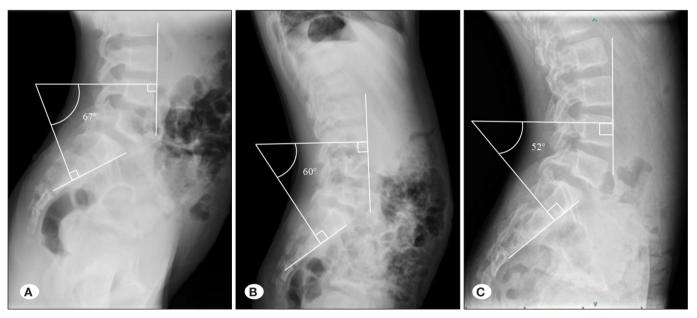
Case	Age (years)	Sex	Preoperative LSA (°)	Postoperative LSA (°)	LSA at the last follow-up (°)	Follow-up time (months)	Vertebroplasty segment
1	11	F	72	69	59	36	L4-S2
2	14	М	66	64	56	16	L3-S2
3	13	F	71	66	60	12	L1-L3
4	13	F	71	73	68	21	L2-L5
5	5.5	М	65	58	53	14	L5-S1
6	5.5	М	59	57	49	12	L4-S2
7	6.7	F	76	65	55	14	T12-L1
8	7	М	54	57	59	13	L4-S3
9	7	М	61	69	68	17	L4-S1
10	7	М	67	60	52	48	T12-L3
11	7	F	70	60	68	21	L4-S4
12	8	F	79	61	69	13	S1-S2
13	8	F	81	69	84	15	L5-S2
14	14	М	55	64	59	24	L4-L5
15	7	F	68	60	65	25	T11-L5
16	5	F	61	61	61	14	L4-S2
17	12	М	82	65	70	17	L3-S2
18	11	F	72	70	44	13	L3-L5
19	5	М	78	54	55	16	S1-S3
20	6.5	F	65	54	60	33	L2-L3
21	13	F	73	70	70	22	L5-S2
22	11	F	88	71	76	15	T11-L1
23	10	М	83	60	57	12	S1-S2

M: Male, F: Female.

Age (years)	Preoperative	Postoperative	The last follow-up	p-value (preoperative vs. postoperative)	p-value (preoperative vs. postoperative)	p-value (preoperative vs. postoperative)
All cases	70.30 ± 9.04	63.34 ± 5.60	61.61 ± 9.14	0.002	0.001	0.346
<10	68.00 ± 8.43	$60.38 \pm 4.84$	61.38 ± 9.45	0.013	0.041	0.694
≥10	73.30 ± 9.36	67.2 ± 4.02	61.90 ± 9.23	0.058	0.012	0.064

Table II: The means ± Standard Deviations for LSAs (°)

P<0.05 was statistically significant.



**Figure 2:** Case 10: A 7-year old male patient. Preoperative (**A**), postoperative (**B**), and follow-up (**C**) lateral spine x-rays were taken, and the LSA was measured and recorded. The preoperative LSA was 67°, the postoperative LSA was 60°, and at the end of 48 months of follow-up the LSA was 52°.

# DISCUSSION

The LSA is an important indicator of sacral tilt angle, and changes in the LSA will lead directly to changes in lumbosacral spine morphology resulting in a compensatory change in the lumbar anterior convexity angle and a decrease in lumbar spine stability. Bailey et al. found that sacral spine morphology was highly positively correlated with the size of the LSA, and the greater the LSA the greater the sacral curvature and the degree of lumbar tilt (2). Kocyigit and Berk found that the correlation between the LSA and the lumbar anterior convexity angle tended to decrease with increasing age (6). Abitbol's measurements showed that the LSA in healthy children up to 11 months of age is generally between 20° and 45° (1), and the earlier they generally learn to stand and walk, the earlier their LSA will increase accordingly, stabilizing at 5 years of age independent of age, height, and weight, with continued increases likely related to neurological deformities (13). Thus the LSA remains relatively stable before puberty. Statistical analysis of the angles of the children in this study group postoperatively and at the final follow-up showed no significant difference (p=0.346), which is consistent with this

view. After grouping by age, there was no significant difference in the postoperative LSA correction rate when comparing the two groups, and the change in LSA in the group older than 10 years was not significantly different postoperatively compared with preoperatively, but there was a significant difference at the last follow-up compared with preoperatively. The lack of statistical significance is likely due to the small number of cases, and there is a need for longer-term followup to increase the reliability of the study results. In addition, differences in gender body image mainly appear after the second sexual characteristics at puberty, thus there was no significant difference in LSA between the genders (4).

Low tethered conus medullaris is a relatively common intraspinal deformity (12), and according to the literature 90% of children with spinal dysraphism develop spinal deformities of varying degrees (10), including scoliosis, kyphosis, and pronation, and often compound deformities, before the age of 10 years. Tubbs et al. suggested that the LSA of children with spinal bulge increases when the symptoms of TCS are exacerbated (15-17). The current treatment of choice for TCS is spinal cord untethering (14), but the perception of postoperative LSA changes is controversial and relatively poorly studied. Tubbs et al. showed that patients with spinal cord tethering who presented with clinical symptoms had a larger LSA than controls and that their signs and symptoms corresponded to an increase in LSA (15,17). However, the LSA did not change significantly after the untethering procedure was performed, and this suggested that the horizontal sacrum could be used as a marker for TCS and that measurement of LSA could be used as an adjunct. Cornips et al. found no significant difference in the LSA measurements in children with clinically progressive TCS compared to children with clinically stable TCS, and they concluded that LSA could not help them determine the need for surgical intervention (5). Because the LSA is still undergoing physiological changes before the age of 5 years, the above reported studies do not include age as a factor, so these findings are somewhat limited. Reigel et al. reported that spinal cord untethering slowed the progression of anterior convexity in patients with upper lumbar lesions but had little effect on the progression of anterior convexity in lower lumbar or sacral lesions, and they concluded that early untethering could reduce the incidence or slow the progression of spinal deformities (11). We previously treated an 8-year-old child with TCS who had low back pain and an enlarged LSA for more than 20 days and performed a surgical untethering after excluding contraindications to surgery, after which we found that the symptoms of lower back pain disappeared, the LSA decreased on radiographs, and the lumbar lordosis was significantly reduced (18). The results of the analysis of LSA changes before and after surgery in this group of children support these previous conclusions.

Structural stability of the posterior spine after spinal cord untethering in children is an important factor in preventing secondary deformities of the spine. Because children have more lax ligaments, immature vertebrae, and higher cancellous bone content, failure to reposition the laminae and reconstruct the posterior spinal structure after laminectomy will increase the risk of postoperative spinal deformity and spinal instability (7), which will inevitably have an impact on the LSA. After spinal cord untethering, the laminae were repositioned and vertebroplasty was performed according to the deformity of the laminae, and the spine was fixed with an absorbable internal fixation system to maintain the structural integrity of the posterior spine, and the mechanical structure of the spine was mostly unaffected.

A shortcoming of this study is that full spinal frontal and lateral radiographs are not routinely obtained after spinal cord untethering in children with TCS, resulting in a small sample size and a short follow-up period in some cases, so the significance of this study still needs to be analyzed with a large number of cases and longer follow-up.

# CONCLUSION

Compared with MRI, which is time-consuming and expensive, we believe that X-rays are more suitable for drawing vertebral osteotomies to measure the LSA because of the different imaging principles and shooting positions, which are economical and suitable for multiple reviews in children. In this study, we measured and analyzed changes in the LSA in sagittal lumbosacral radiographs of 23 children with TCS over 5 years of age before and after surgery and at the last follow-up, and the results showed that the LSA of the children after spinal cord untethering and at the last follow-up were statistically significant compared with the LSA before surgery, indicating that spinal cord untethering can improve the LSA tilt and delay the progression of lumbar lordosis in children with TCS over 5 years of age. There was no significant difference in the angle at the follow-up, indicating that the LSA remained relatively stable during the follow-up period.

#### **AUTHORSHIP CONTRIBUTION**

Study conception and design: BX Data collection: HW, YD Analysis and interpretation of results: BX, HW Draft manuscript preparation: HW Critical revision of the article: WW, FL, WH, FW All authors (BX, HW, YD, WW, FL, WH, FW) reviewed the results and approved the final version of the manuscript.

#### REFERENCES

- Abitbol MM: Evolution of the lumbosacral angle. Am J Phys Anthropol 72(3):361-372, 1987
- Bailey JF, Sparrey CJ, Been E, Kramer PA: Morphological and postural sexual dimorphism of the lumbar spine facilitates greater lordosis in females. J Anat 229(1):82-91, 2016
- 3. Bui CJ, Tubbs RS, Oakes WJ: Tethered cord syndrome in children: A review. Neurosurg Focus 23(2):E2, 2007
- 4. Cho Y: Evaluation of global sagittal balance in Koreans adults. J Korean Neurosurg Soc 60(5):560-566, 2017
- Cornips EM, Razenberg FG, van Rhijn LW, Soudant DL, van Raak EP, Weber JW,Robben SG, Fock JM, Catsman-Berrevoets CE, Vles JS: The lumbosacral angle does not reflect progressive tethered cord syndrome in children with spinal dysraphism. Childs Nerv Syst 26(12):1757-1764, 2010
- Kocyigit BF, Berk E: Comparison of lumbosacral alignment in geriatric and non-geriatric patients suffering low back pain. Pak J Med Sci 34(2):282-287, 2018
- McGirt MJ, Constantini S, Jallo GI: Correlation of a preoperative grading scale with progressive spinal deformity following surgery for intramedullary spinal cord tumors in children. J Neurosurg Pediatr 2(4):277-281, 2008
- 8. Oakes WJ: The borderlands of the primary tethered cord syndrome. Clin Neurosurg 43:188-202, 1996
- Paradiso G, Lee GY, Sarjeant R, Fehlings MG: Multi-modality neurophysiological monitoring during surgery for adult tethered cord syndrome. J Clin Neurosci 12(8):934-936, 2005
- 10. Piggott H: The natural history of scoliosis in myelodysplasia. J Bone Joint Surg Br 62-B(1):54-58, 1980
- Reigel DH, Tchernoukha K, Bazmi B, Kortyna R, Rotenstein D: Change in spinal curvature following release of tethered spinal cord associated with spina bifida. Pediatr Neurosurg 20(1):30-42, 1994

- Robbins JW, Lundy PA, Gard AP, Puccioni MJ: Perineal pain secondary to tethered cord syndrome: Retrospective review of single institution experience. Childs Nerv Syst 31(11):2141-2144, 2015
- Rotenstein D, Reigel DH, Lucke JF: Growth of growth hormone-treated and nontreated children before and after tethered spinal cord release. Pediatr Neurosurg 24(5):237-241, 1996
- Thuy M, Chaseling R, Fowler A: Spinal cord detethering procedures in children: A 5 year retrospective cohort study of the early post-operative course. J Clin Neurosci 22(5):838-842, 2015
- Tubbs RS, Bui CJ, Loukas M, Shoja MM, Oakes WJ: The horizontal sacrum as an indicator of the tethered spinal cord in spina bifida aperta and occulta. Neurosurg Focus 23(2):E11, 2007

- 16. Tubbs RS, Naftel RP, Rice WC, Liechty P, Conklin M, Oakes WJ: The patient with symptoms following resection of a lipomyelomeningocele: Do increases in the lumbosacral angle indicate a tethered spinal cord? J Neurosurg 105(1 Suppl):62-64, 2006
- Tubbs RS, Wellons JC 3rd, Bartolucci AA, Blount JP, Oakes WJ: Horizontal sacrum as an indicator of a tethered spinal cord. Pediatr Neurosurg 36(4):209-213, 2002
- Zhang Y, Xia B, Liu F, Niu X, Hu W, Wu H: Tethered cord syndrome with lower back pain and lumbosacral angle increase: Case report. Childs Nerv Syst 36(1):219-221, 2020