Original Article

Scoliosis may be the first symptom of the tethered spinal cord

Mustafa Barutçuoğlu, Mehmet Selçuki, Ahmet Sukru Umur, Mesut Mete, Seren Gulsen Gurgen[1], Deniz Selcuki[2]

Abstract

Background: Tethered cord syndrome (TCS) is a progressive clinical entity that arises from abnormal spinal cord tension. Scoliosis may be a unique symptom in TCS. The aim of this study is to investigate prognosis after releasing the filum terminale in scoliosis due to TCS with/without findings in magnetic resonance imaging (MRI) and to draw attention to the importance of somatosensorial evoked potentials (SSEP) on the differential diagnosis of idiopathic scoliosis versus scoliosis due to TCS with normal appearance of filum terminale and conus medullaris.

Materials and Methods: Eleven female and seven male patients with progressive scoliosis were included in the study. They were evaluated radiologically, SSEP and urodynamical studies. Preoperative and postoperative anteroposterior full spine X-rays were obtained for measuring the Cobb's angle. MRI was performed in all cases for probable additional spinal abnormalities. All patients underwent filum terminale sectioning through a L5 hemilaminectomy. The resected filum terminale were subjected to histopathological examination.

Results: The mean Cobb angle was 31.6° (range 18°–45°). Eight patients (44.45%) had a normal appearance of filum terminale and normal level conus medullaris in MRI, but conduction delay and/or block was seen on SSEP. In the histopathological examination of filum terminale dense collagen fibers, hyaline degeneration and loss of elastic fibers were observed. Postoperatively none of the patients showed worsening of the Cobb angle. Three patients showed improvement of scoliosis.

Conclusion: In TCS presented with scoliosis, untethering must be performed prior to the corrective spinal surgery. Absence of MRI findings does not definitely exclude TCS. SSEP is an important additional guidance in the diagnosis of TCS. After untethering, a followup period of 6 months is essential to show it untethering helps in stopping the progress of the scoliotic curve. In spite of non progression (curve stopped lesser than 45°) or even improvement of scoliosis, there may be no need for major orthopedic surgical intervention.

Key words: Cobb angle, scoliosis, somatosensorial evoked potentials, tethered cord syndrome
MeSH terms: Spinal cord, scoliosis, tethered cord syndrome, neural tube defects

Introduction

Tethered cord syndrome (TCS) is a well-known clinical entity and is characterized by motor and sensorial changes in the legs, back pain, foot deformities, urinary dysfunction and spinal deformity. Scoliosis can be a unique and leading sign of TCS.[1] In the classic description of TCS by Hoffman *et al*.,[2] scoliosis was a prominent manifestation in 10 of 31 patients. Recent reports have suggested that scoliosis is more frequent than thought in TCS and additionally McLone *et al*. demonstrated that scoliosis may be the first sign of TCS.[1-4]

Pathological entities of the neural tube like meningoceles, lipomyelomeningoceles, myelomeningoceles, split cord malformation, dermal sinus tract, thickened filum and others may be the cause of tethering.[5] Yamada *et al*. showed that longitudinal stretching of the spinal cord interferes with the mitochondrial oxidative metabolism and can result in motor and sensory changes in the lower limbs, incontinence and musculoskeletal deformities including neuromuscular scoliosis.[6]

In the simplest form, the spinal cord can be tethered by a thickened filum (>2 mm) with a low conus medullaris (at or below L2–L3 level) without other forms

Department of Neurosurgery, Celal Bayar University School of Medicine, Turkey, [1]Department of Histology and Embryology Celal Bayar University Vocational School of Health Services, Turkey, [2]Department of Neurology, Celal Bayar University School of Medicine, Turkey

Address for correspondence: Dr. Mustafa Barutçuoğlu, Department of Neurosurgery, School of Medicine, Celal Bayar University, Turkey.
E-mail: barutcum@yahoo.com

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of spinal dysraphism. However, the spinal cord can even be tethered by a filum terminale with normal appearance and a normal level conus medullaris.[7-9]

It is known that the majority of patients with scoliosis (80–90%) are "idiopathic" because an underlying clinical or radiological cause has not been determined. According to the scoliosis research society, the prevalence of adolescent idiopathic scoliosis is 2% to 3% in the general population.[10]

Tethered cord may be the most important and relatively easily treatable primary disease for associated scoliosis. Releasing the tethered spinal cord in these patients may stabilize or even reverse the progression of scoliosis. In addition, untethering aims to improve neuromuscular function even in patients that need surgical correction of scoliosis.[11-14]

The aim of this study is to evaluate scoliosis due to TCS with and without tethering, findings on magnetic resonance imaging (MRI) and also to draw attention to the importance of somatosensorial evoked potentials (SSEP) on the differential diagnosis between idiopathic scoliosis and TCS due to filum terminale with normal appearance and normal level conus medullaris. In addition, we also focused to investigate the progression of scoliosis and need for corrective spinal surgery in patients who underwent release of tethering.

Materials and Methods

Eleven female and seven male patients with a mean age of 14.72 years (range 3–42 years) were included in the study [Table 1]. Mean duration of symptoms was 9 months (range 1–36 month). Full spine standing X-rays were obtained for calculating the Cobb's angle on coronal curves. Postoperative curve was defined as stable if less than 10° of progression occurred. Decrease of Cobb's angle up to 10° was defined as improvement.

MRI was performed in all patients for the diagnosis of TCS and for probable additional spinal abnormalities. Normal conus medullaris level was accepted as between the lumbar 1st and 2nd disc spaces and the normal thickness of filum as taken as <2 mm. Developmental abnormalities of neural tissue such as myelomeningocele, lipomyelomeningocele, split cord malformations etc., which caused secondary tethering and scoliosis were excluded from the study.

We performed SSEP for spinal cord conduction in 16 patients. There was poor cooperation for the examination in two toddlers. Greater than 50% decrease in amplitude or a >10% increase in latency was considered as pathological. All SSEP recordings were done at lumbal, thoracal, cervical levels and the somatosensorial cortex simultaneously.

Urodynamic studies were also performed in all patients to investigate detrussor and sphincter activity evaluations.

All untethering procedures were performed at the department of neurosurgery. The study was approved by the hospital's Institutional Review Board Committee.

All cases underwent a standard surgical procedure of a single level lumbar hemilaminectomy at the fifth lumbar vertebrae on the left. After opening the dura mater, we used a nerve stimulator to differentiate the filum terminale from other neural tissue. Following the untethering procedure a part of the filum was taken for histopathological examination in all patients. The patients were discharged on the third postoperative day.

Results

All patients presented with progressive scoliosis and low back pain. Some patients mentioned weak sensorial symptoms such as numbness in the perineum or non radicular pain in the legs. Four patients had frequent urinary infections. There were no objective, additional complaints or findings. Scoliosis was defined as a coronal deformity of up to 10°. Mean value of Cobb angle in the study was 31.6° (range 18°–45°). Eleven patients (61.1%) had thoracolumbar scoliosis and 7 (38.9%) had thoracic scoliosis. One patient had a right curved scoliosis and 17 (94.45%) patients had left.

Besides scoliosis, 10 of 18 patients had thick and/or fatty filum terminale, low level of conus medullaris and developmental abnormalities such as block vertebrae. Other eight scoliotic patients (44.45%) had a normal appearance of filum terminale and normal level conus medullaris without different forms of spinal dysraphism on MRI.

We clinically suspected the tethered cord in scoliotic patients, if there was a late start of the scoliotic curve, in particularly during adolescence. We did not suspect TCS when the spinal deformity progresses in spite of appropriate treatment. On the contrary, progress of spinal deformity after appropriate treatment means untethering did not help to improve or stop the bending. It is for this reason we recommend 6 months of followup period to see the results of untethering. Should there be a progress of the curve than we refer the patient to orthopedics for surgical intervention. We found pathological SSEP results in 12 of 16 patients (75%) of which there was total block in nine (75%), conduction delay in three (25%). SSEP was significant in all cases

Table 1: Clinical details of patients

Patient	Gender	Age (in years)	Complaint	Duration of symptom	MRI	Urodinamy	SSEP	Cobb		Followup (in years)
								Before (in degree)	After (in degree)	
1	♀	14	Bending of spinal column	12 months	Rotoscoliosis (left), fatty filum	Sensorial delay, increased capacity	Normal	42	40	11
2	♀	6	Bending of spinal column	3 months	Scoliosis (left), L1 hemivertebrea, thick filum	Normal	Thoraco-lumbar conduction delay	29	30	4
3	♀	13	Bending of spinal column	1 month	Scoliosis (left)	Normal	Right lumbar block	31	30	3
4	♀	14	Bending of spinal column	3 months	Scoliosis (left)	Normal	Total lumbar block	22	12	3
5	♂	5	Bending of spinal column	5 months	Scoliosis (left), low level conus medullaris	Normal	Normal	28	20	17
6	♀	42	Bending of spinal column	5 months	Scoliosis (left), thoracal 6-7 hydromyelia, fatty filum	Normal	Total lumbar block	35	30	9
7	♂	3	Bending of spinal column	3 months	Rotoscoliosis (right)	Normal	N/A	34	30	4
8	♂	8	Bending of spinal column	3 months	Scoliosis (left), thick filum	Normal	N/A	38	26	1
9	♂	17	Bending of spinal column	6 months	Scoliosis (left)	Neurogenic bladder	thoracal-lumbar block	18	8	2
10	♀	23	Bending of spinal column	18 months	Scoliosis (left)	Normal	Total lumbar block	45	45	2
11	♂	14	Bending of spinal column	12 months	Scoliosis (left)	Neurogenic bladder	Right thoracal-lumbar conduction delay	32	34	13
12	♂	21	Bending of spinal column	36 months	Scoliosis (left), cervical syrinx	Neurogenic bladder	thoracal-lumbar block	27	25	11
13	♂	18	Bending of spinal column	18 months	Scoliosis (left)	Normal	Thoracal lumbar block	32	26	9
14	♀	12	Bending of spinal column	12 months	Scoliosis (left), low level conus medullaris	Normal	Right conduction delay	45	45	1
15	♀	12	Bending of spinal column	3 months	Scoliosis (left), thick filum	Normal	Normal	19	20	1
16	♀	17	Bending of spinal column	6 months	Scoliosis (left)	Neurogenic bladder	Right thoracal-lumbar block	18	18	1
17	♀	13	Bending of spinal column	12 months	Scoliosis (left), low level conus medullaris	Normal	Normal	32	N/A	17
18	♀	13	Bending of spinal column	6 months	Scoliosis (left), low level conus medullaris	Normal	Thoracal block (left) right thoracal-lumbar conduction delay	42	42	0.25

NA=not available, MRI=Magnetic resonance imaging, SSEP=Somatosensorial evoked potentials, ♀=Female, ♂=Male

with normal filum and conus medullaris seen on the MRI [Figure 1].

In urodynamical studies, only four patients had increased detrussor activity with sphincteric dyssynergia defined as neurogenic bladder abnormality. In one patient, there was sensorial delay, both increased bladder capacity and compliance. There were no pathological bladder function findings in the other 13 patients (72.22%).

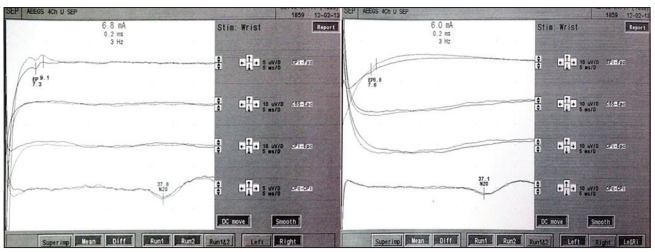

Figure 1: Somatosensorial evoked potentials results, bilaterally lumbar-thoracal total block

In the histopathological examination of the fila in all patients (including those with normal appearance of the fila on MRI), we observed dense collagen fibers and hyaline degeneration in Masson's trichrome staining [Figures 2 and 3],

furthermore there were no elastic fibers seen in the Verhoeff's Van Gieson staining in these fila [Figure 4].

The cases were followed up postoperatively for a mean of 6 years (range 3–204 months). No patient showed worsening of the Cobb angle. Three patients showed a slight improvement of the scoliotic curve (10°–12°).

Discussion

Scoliosis is defined as a lateral curvature of the spine on the coronal plane that is Cobb's angle greater than 10°. This deformity may include lateral inter-vertebral tilting and rotation of the vertebral bodies across the apex toward the convexity of the curve in the axial plane.[15,16]

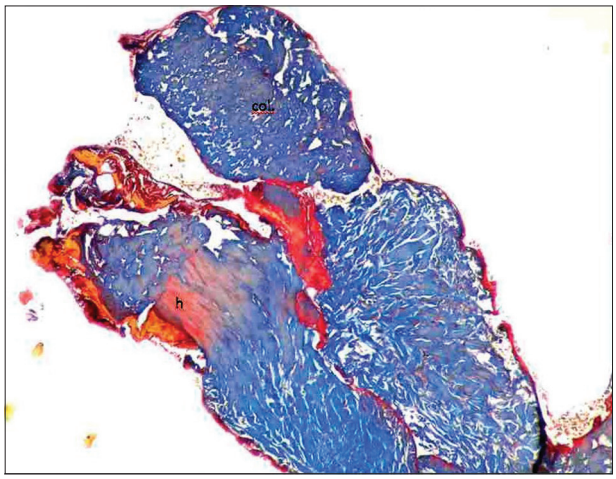

Figure 2: Histological view of a filum terminale in a scoliosis patient with normal appearance. It is clear that microscopical cut view is not normal with dense collagen tissue and hyaline degeneration. ×10 (Col: Collagen, h: Hyaline [Masson's trichrome])

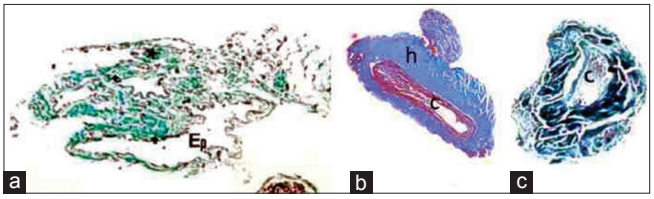

Figure 3: (a) Loose and rich collagen tissue in normal filum terminale. (b) Dense collagen (light blue) tissue and large vessel in the middle of a filum of a scoliosis patient. (c) Dense collagen and large capillary (c) in the middle of filum terminale of a patient with incontinence (histological sections stained with Masson's tricrome and the pictures "a" and "c" are taken from Lit. 17)

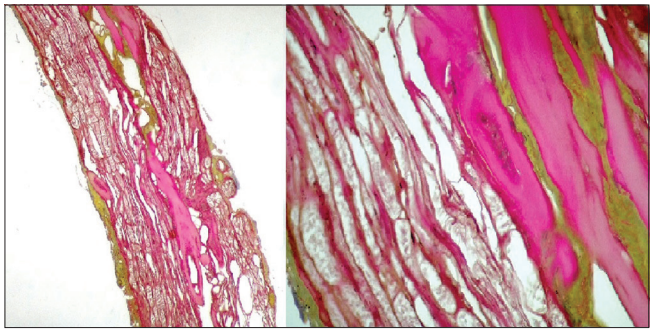

Figure 4: Histological view of filum terminale staining with Verhoeff ×10, seen loose of elastic fibers (black) and dense collagen tissue (red)

Neuromuscular scoliosis is a deformity difficult to treat. It may progress even after skeletal maturity. The understanding of the pathophysiology of neuromuscular scoliosis in TCS continues to evolve. This clinical syndrome seems purely the result of spinal cord traction. Development of scoliosis may be body's attempt to minimize the abnormal tension placed on the spinal cord because of the tethering effect of the tight filum. Scoliosis and kyphoscoliosis have limited mobility of the spinal column and flexion increases spinal cord traction. The concave side of the spinal canal is shorter than the convex side and rotation is always to the concave side which shortens the distance travelled by the spinal cord.

In our series, mean age was 14.72 years. 10 of 18 patients were in the adolescent group, four patients were under 10 years old and the others were adult. More than half the patients in our series had idiopathic scoliosis with 5 of these 8 adolescent having a normal appearance of the filum terminale and conus medullaris. It's not always difficult to diagnose TCS if there is an apparent developmental anomaly such as a split cord malformation, dermal sinus tract, neural tube closure defect and the other skin stigmata. In imaging studies too thick, fatty, short filum terminale and low-lying conus medullaris are quite easy to recognize tethered spinal cord for the diagnosis.[6,9,15,17-22] Problem arises in the patients who have scoliosis without any other pathology or finding to explain the reason of this deformity.

After having untethered, the patient is safer for scoliotic curve correction procedure. Since 1979, the concept of tethered cord and its effects almost totally changed. It is 2003, when Selçuki *et al.*,[9] showed that filum terminale with normal appearance is not normal, for the first time in the literature. Many other investigations, including scanning electron microscopy studies followed Selçuki's histological study. These papers have taken their places in the literature and can easily be found elsewhere. The first paper of "Protean manifestations of tethered spinal cord" by Hoffman *et al.*,[2] was in 1976, and manifestations were pain, incontinence and scoliosis.The prevalence of idiopathic scoliosis has been reported to be 0.9–12% in the general population and minimum 10% of patients require treatment. Weis and Moramarco reported that the majority of the patients with scoliosis (80–90%) are "idiopathic" because an underlying cause can not been determined.[10] Many scoliosis patients who may have spinal cord tethering without MRI findings may be included in the idiopathic scoliosis group.

Conservative treatments like bracing and many others are designed to prevent curve progression during the growing years and is not indicated in patients who are already skeletally mature.[23] Surgical treatment is generally indicated if the curve reaches 50° at completion of growth. Curves <50° in skeletally immature patients and/or progressive double curves are also indications for corrective surgery.[16] The goals of surgery are to correct the deformity producing a balanced spine with a leveled pelvis and a solid spinal fusion to prevent or delay secondary respiratory complications.[10,12,13] These corrective spinal surgical interventions are usually difficult, expensive and may carry increased risk of morbidity. Limited instrumentation options, persistent anterior spinal growth after posterior fusion and the potential for thoracic insufficiency when a kind of fusion is performed in children or young patients, this may require repetitive spinal surgery. On the other hand, a corrective orthopedic procedure may cause irreversible neurological deficits unless untethering of the spinal cord has been performed previously.[19,24]

Since the concept of tethered spinal cord was first proposed, the finding of low-lying conus medullaris has been widely accepted as evidence of cord tethering. The easiest way to diagnose this condition is by a lumbar spine MRI. Hoffman[2] clearly defined radiological criteria on TCS, but Selçuki *et al*.[9] showed that normal appearance of the filum terminale does not mean that it really is totally normal. They reported that an abnormal filum terminale with a normal appearance has dense collagen fibers, wide and numerous capillaries and hyaline formation. The high amount of dense collagen fibers and hyalin cause the filum terminale to lose its elastic properties, ultimately leading to conduction of a tethering effect to the conus medullaris [Figures 2-4].

Somatosensory evoked potentials are not routinely used in spinal dysraphism or idiopathic scoliosis. The sensory stimulus has often been used in clinical evoked potential studies as a way of measuring the spinal cord function.[25] SSEPs are performed by stimulating a peripheral nerve at the wrist or the ankle and recording the responses from the popliteal fossa, cervical spine/brainstem and somatosensory cortex. SSEPs give direct feedback from the posterior column. Abnormal SSEPs provide clinical evidence of posterior column dysfunction. In our series, 8 of 18 patients were with normal appearance of filum terminale and conus medullaris seen in the MRI [Figure 1]. When normal appearance of filum terminale and normal level of conus medullaris are confusing findings, in case of delayed scoliosis then we performed SSEP to consider the surgical intervention. We have found that in 12 patients SSEPs were pathological both in conduction time and amplitude of the wave recorded. While it is not uncommon to see pathological lumbal or thoracic conduction blocks or delays although the SSEP results that recorded from cental cerebral lead shows normal waves that can easily be considered as normal, all SSEP recordings were done at lumbal, thoracic, cervical levels and at somatosensorial cortex simultaneously.

However, contrary to what we expected, SSEP results did not match with the urodynamic studies in tethered cord patients with urinary incontinence. In our previous studies we felt that the hyperreflexive, hypertonic bladder is the main indicator of the tethering of the conus medullaris, which is still very important in decision making to untether the spinal cord in patients with urinary incontinence.[21] It is interesting that in the majority of scoliosis patients who recorded SSEP's as pathological, the urodynamic studies revealed normal values. Most of the patients in the study had normal urodynamic evaluation, and significant part of them had normal MRI investigation as far as level of conus medullaris and thickness of filum terminale were concerned. While our impression about the hyperreflexive and hypertonic bladder has not changed so far vis-a-vis patients with bladder and sphincter dysfunction but after seeing these fifteen scoliosis patients with normal urodynamic study scores, we decided that SSEP might be a better indicator of tethering of spinal cord especially in normal level conus medullaris and normal thickness filum terminale in scoliosis.

After evaluating the results in our series, we noticed that tethered cord induced late onset scoliosis always in the thoracic area with a compensatory curve at the lumbar level. Thoracic curves were almost always directed to the left side. Hence, we came to a conclusion whether this left side directed thoracic late onset curve could be a sign of cord tethering. In order to answer this question, we believe accumulation of more data is needed indeed.

The tethered cord is vertically tethered spinal cord that is caused by tethering of filum terminale in majority of patients. Interestingly, despite the level of the scoliotic curve, untethering of distal spinal cord helps in most of the cases. Furthermore, it is reported by Milhorat *et al*., that untethering, in diagnosed tethered spinal cord cases, also helps in Chiari malformation of type I. There is, unfortunately, no clear cut explanation for this.

Histopathological analysis of the filum terminale with normal appearance revealed either dense collagen tissue or hyaline degeneration. All of the fila with macroscopically normal tissue showed either dense collagen tissue or some hyaline degeneration and markedly wide vessels

Table 2: Probability of progression: Magnitude of curve versus age

Magnitude of curve at the admission (°)	Age at the admission		
	10-12 years %	13-15 years %	16 years %
<19°	25	10	0
20°-29°	60	40	10
30°-59°	90	70	30
>60°	100	90	70

[Figures 2 and 3]. These properties of histopathologically abnormal fila terminalia have already been explained in by Selçuki *et al*.[9,14] Elastic tissue fibers were seen when stained with Verhoff's Van Gieson. This stain is useful in demonstrating the presence of elastic tissue. We did not obtain positive Verhoeff elastic fibers staining [Figure 4] in both normal appearing of fila and others. This histological result may explain stretching of normal level conus medullaris also with normal appearance of filum terminale on MRI.

In 1982, Nachemson *et al*. calculated the probability of curve progression before skeletal maturity based on known prognostic factors available at the time[24] [Table 2]. The results show that risk of curve progression decreases with increasing skeletal maturity. However, with larger magnitude curvatures, there may be a considerable risk of progression despite maturity.[26]

Scoliosis surgery is performed by both orthopedic surgeons and neurosurgeons. Since tethered cord syndrome is a neurosurgical issue, the orthopedic surgeons must keep in mind this important problem. This is why we wanted to draw attention to this very important point as far as we could. We believe that repetitive impulses (stimulations) on this important issue would be beneficial for true understanding of the intriguing problem.

Conclusion

Scoliosis may be the first sign of tethered spinal cord. At late onset cases of scoliosis must be assessed for the existence of TCS. In the presence of normal urodynamic studies and MRI; SSEP seems to be very helpful in considering surgical untethering. Filum terminale with normal appearance in MRI does not rule out cord tethering. It has been shown that filum terminale with normal appearance is not histologically normal. In diagnosed tethered cord cases who presented themselves with scoliosis, untethering of the spinal cord must be performed previous to the orthopedic corrective spinal surgery. After untethering, a followup period of 6 months allows the physician to evaluate the effects of untethering towards the evolution of scoliosis. In spite of improvement or cessation of progression of scoliosis there may be no need for major orthopedic surgical intervention.

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