Scoliosis may be the first symptom of the tethered spinal cord

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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 somatosensory 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 that 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, somatosensory evoked potentials, tethered cord syndrome

MeSH terms: Cobb angle, scoliosis, somatosensory evoked potentials, tethered cord syndrome

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
of spinal dysraphism. However, the spinal cord can even be tethered by a filum terminale with normal appearance and a normal level conus medullaris.\textsuperscript{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.\textsuperscript{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.\textsuperscript{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 somatosensory 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.

\textbf{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 1\textsuperscript{st} and 2\textsuperscript{nd} disc spaces and the normal thickness of filum as taken as <2 mm. Developmental abnormalities of neural tissue such as myelomeningocele, lipomyelomingocele, 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 somatosensory cortex simultaneously.

Urodynamic studies were also performed in all patients to investigate detrusor 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.

\textbf{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.
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In urodynamical studies, only four patients had increased detrusor 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%).

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],

Table 1: Clinical details of patients

| Patient | Gender | Age (in years) | Complaint | Duration of symptom | MRI | Urodinamy | SSEP | Cobb Before (in degree) | Cobb After (in degree) | Followup (in years) |
|---------|--------|----------------|-----------|---------------------|-----|-----------|------|------------------------|----------------------|---------------------|
| 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 hemivertebra, 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) | Normal | Thoraco-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 thoraco-lumbar conduction delay | 32 | 34 | 13 |
| 12      | ♂      | 21             | Bending of spinal column | 36 months | Scoliosis (left), cervical syrinx | Neurogenic bladder | Thoraco-lumbar block | 27 | 25 | 11 |
| 13      | ♂      | 18             | Bending of spinal column | 18 months | Scoliosis (left) | Normal | Thoracic 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 thoraco-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 | Thoracic block (left) right thoraco-lumbar conduction delay | 42 | 42 | 0.25 |

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

Figure 1: Somatosensorial evoked potentials results, bilaterally lumbar-thoracal total block
Scoliosis may be the first symptom of the tethered spinal cord. Furthermore, there were no elastic fibers seen in the Verhoeff’s Van Gieson staining in these filia [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

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 thick 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.\textsuperscript{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.\textsuperscript{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.\textsuperscript{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\textsuperscript{2} clearly defined radiological criteria on TCS, but Selçuki et al.\textsuperscript{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.\textsuperscript{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 somatosensory 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.\textsuperscript{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.,\textsuperscript{1} 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.
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Table 2: Probability of progression: Magnitude of curve versus age

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

[Figures 2 and 3]. These properties of histopathologically abnormal fila terminalia have already been explained in by Selçuki et al. 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 filament 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 time24 [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.

Conclusions

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 filament 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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