Author’s response to reviews

Title: Real spinal cord injury without radiologic abnormality in pediatric patient with tight filum terminale following minor trauma: a case report

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Author’s response to reviews:

Dear Prof. Zenon Pogorelić:

Thank you very much for your comments about our paper submitted to BMC Pediatrics.

We have carefully checked the manuscript and revised it as following.

(1) We have updated some of the references.

(2) We have rewritten the introduction and discussion sections of the manuscript.

Besides the above changes, we have corrected some expression errors. Thank you very much. If you have any question about this paper, please don’t hesitate to let me know.

Dear Prof. Dimitrios Christos Nikas:

Thank you very much for the excellent and professional revision of our manuscript.
1. In the second case, patient arrived in our hospital within 8 hours of injury. This patient was treated with high-dose methylprednisolone. Therapy at our institution followed the NASCIS II and NASCIS III protocols for dosing and administration, which consisted of methylprednisolone administration with a bolus dose followed by a 45-minute pause and then a 23-hour continuous infusion. In the first case, patient arrived in our hospital 30hrs past injury. In the third case, there were 3 days past injury when patient arrived in our hospital. In these two cases, peri-operative intravenous low-dose corticosteroids were used to prevent complications and mortality. Slow intravenous infusion of dexamethasone (0.2 mg/kg of real body weight 1 day before the surgery and at day +1, day+2 after the surgery).

2. Over the years, we have found some similar SCIWORA patients with occult spinal dysraphism in clinical practice. The trauma was potentially mild in all patients, but resulted in catastrophic damage of the cord. The injured portion involved the lower thoracic and lumbar spinal cord and the prognosis was poor after conservative treatment. But the data of these patients was incomplete. Based on the findings of previous clinical experience, we infer that preexisting spinal or neurological abnormalities, such as tight filum terminale may act as a predisposing factor for SCIWORA. Lysis of the filum terminale can release the spinal cord traction, which is beneficial to the recovery. We fully communicated with the family members of the children and got their consents before the operation. The aim of this case report is to bring this new finding to world attention, and might even spark international debates on this medical important subject.

In order to find possible preexisting spinal or neurological abnormalities, a thoroughly exploration of the conus and filum terminale at the same time was needed. The patients included in the study underwent an extended surgical exploration and lysis of the filum terminale through a multi-level laminoplasty. L2-L4 laminoplasty was performed in the operation. In all cases, neuroendoscope was also used to get a close look of the conus medullaris.

The term lysis means sectioning of the filum terminale. The filum terminale has clinical significance in its contribution to tethered cord syndrome (TCS), a form of occult spinal dysraphism. TCS refers to a collection of signs and symptoms of motor and sensory neuron dysfunction attributable to spinal cord traction. The basic underlying problem in tight filum terminale is abnormal longitudinal stretch on the conus by the filum. Sectioning of the filum terminale, cutting the arachnoid and fibrous bands, releasing the spinal cord and correction of the associated malformation was the main surgical technique that was performed in TCS patients.
3. The term lysis means sectioning of the filum terminale. In the operation, patients are positioned prone under general anesthesia with supporting rolls on each side. The dura was opened in the midline and tacked by four sutures bilaterally. Following the dural opening, filum terminale, arachnoid bands, and rootlets were first observed. Filum terminale and nerve roots were free from the surrounding tissues after the identification. Filum terminale was coagulated and cut. In addition, all connective tissues attached to the caudal part of the spinal cord and conus medullaris were released. A cut portion of the filum terminale was sent for pathologic review.

The surgical plan was tailored according to the preoperative radiological diagnosis and modified as necessary according to intraoperative pathological findings. The routine procedure is the L5 laminectomy, and additional partial or complete additional laminectomy is added in order to expose the dura and then to identify filum terminale. In order to find possible preexisting spinal or neurological abnormalities, a thoroughly exploration of the conus and filum terminale at the same time was needed. The patients included in the study underwent an extended surgical exploration and lysis of the filum terminale through a multi-level laminoplasty. L2-L4 laminoplasty was performed in the operation.

4. In the third case, preop MRI showed fibrolipoma of the filum terminale. Terminal filum with fibrous degeneration was found in the first case and terminal filum fibrolipoma was found in the second case and the third case in the operation. The fibrolipoma of the filum terminale was confirmed by histopathology.

5. See question answer 3.

6. See question answer 3.
The exact mechanism of tethered spinal cord remains unknown. Most scholars believed that the tethered cord syndrome (TCS) results from the fixation of the spinal cord in the vertebral canal due to the adhesions either of congenital or acquired origin. [1,2] In theory, the basic underlying problem in tight filum terminale is abnormal tension on the conus by the filum. The spinal cord and conus are secured to the surrounding spinal canal down to the level of T12 by the dentate ligaments. The conus is fixed to the canal caudally by the filum terminale, a fibro elastic structure that continues as far down as S2 intradurally and then continues extradurally. The normal filum is less than 2 mm in diameter, and the normal location of the conus tip is above the lower part of the body of L2. [3] The primary role of the filum terminale is to secure and stabilize the conus during movements, which can significantly lengthen or shorten the spinal canal. Some studies have shown as much as a 7% increase in length of the canal during flexion. [4] When applied to neural tissue, this stretch can result in metabolic derangements equivalent to ischemic injury. [5,6] It serves to reason that the filum is the structure responsible for accommodating these length changes. The failure of the filum to facilitate spinal cord movement is thought to be the pathologic mechanism that leads to symptoms of a tethered cord. [6] The spinal cord relies on oxidative metabolism to produce ATP and decreased elasticity of an abnormal filum transfers pathologic stretch to the distal cord and conus. The resultant reductions in oxidative metabolism mirror those seen in hypoxemia. [6] Mild to moderate stretch in experimental models results in transient reductions in metabolism, whereas more severe stretch causes persistent metabolic derangements that may not recover. This stretch leads to mechanical torsion and ischemia in distal spinal cord including conus medullaris thus motor-sensory deficits and associated urological and orthopedic complications are seen in TCS. [1,2]

References:


Dear Prof. Ivna Cvitković:

Thank you very much for the excellent and professional revision of our manuscript. After carefully studying your advice, we have made corresponding changes to the paper. The patients in our report had no signs or symptoms of tethered cord syndrome prior to the minor trauma. On admission to the hospital, all the patients had paralysis of the lower extremities and dysfunction of two bowel movements that preoperative urodynamic testing could not be done. Based on the patients’ medical history, neurological imaging and surgical evidence, the diagnosis of the three cases is tight filum terminale, which is a component of occult spinal dysraphism. The first and second cases have negative findings from lumbosacral MRI, the diagnosis is occult tight filum terminale. In the third case, preop MRI showed fibrolipoma of the filum terminale. The diagnosis of the patient is tight filum terminale, fibrofatty filum terminale.