

CASE REPORT

Elderly-onset degenerative “lumbar spondylotic myelopathy” in a patient with a low-placed spinal cord successfully treated by laminotomy: a case report

Ko Hashimoto^{1,2}, Takumi Tsubakino¹, Takeshi Hoshikawa¹, Tomowaki Nakagawa¹, Takashi Inawashiro¹, Shoichi Kokubun³, Eiji Itoi² & Yasuhisa Tanaka¹

¹Department of Orthopaedic Surgery, Tohoku Central Hospital, Yamagata, Japan

²Department of Orthopaedic Surgery, Graduate School of Medicine, Tohoku University, Sendai, Miyagi, Japan

³Research Center for Spine and Spinal Cord Disorders, National Hospital Organization Sendai-Nishitaga Hospital, Sendai, Miyagi, Japan

Correspondence

Ko Hashimoto, 1-1 Seiryō-machi, Aoba-ku, Sendai, Miyagi 980-8574, Japan.

Tel: +81 22 717 7245;

Fax: +81 22 717 7248;

E-mail: khashimoto@med.tohoku.ac.jp

Funding Information

No sources of funding were declared for this study.

Received: 15 June 2015; Revised: 31 August 2015; Accepted: 28 September 2015

Clinical Case Reports 2015; **3**(12): 1021–1025

doi: 10.1002/ccr3.424

Portions of this work were presented in a podium presentation at the 22nd Japanese-Korean Combined Orthopaedic Symposium, Nikko, Japan, 8 June 2012.

Introduction

Low-placed spinal cord is associated with various types of spinal dysraphism, such as spina bifida or lipomyelomeningocele [1]. These deformities can cause longitudinal traction of the spinal cord and a multitude of neurological deficits in childhood, known as “tethered cord syndrome (TCS)” [2]. Although TCS usually occurs in childhood, tethering of the spinal cord itself can be silent for decades until adulthood [3–5]. As for its pathomechanism, degenerative changes or trauma is assumed to play an important role in adult-onset TCS, while stretching of the spinal cord itself is the major cause of the neurological deficits in childhood-onset TCS [5]. The surgical strategy needs to be altered according to the pathology, that is, while release of the stretched spinal

Key Clinical Message

The authors report a rare case of elderly-onset “lumbar spondylotic myelopathy” occurred on a low-placed spinal cord compressed at multiple levels with thickened ligamenta flava. A posterior decompression surgery could alleviate neurological symptoms successfully instead of untethering of the spinal cord, a widely accepted surgery for tethered cord.

Keywords

Adulthood onset, decompression, laminotomy, nerve compression, spondylosis, tethered cord syndrome.

cord is widely performed for childhood-onset TCS [6–8], another surgical approach needs to be considered according to the pathomechanism [9].

A rare case of elderly-onset lumbar spinal canal stenosis causing “lumbar myelopathy” in a patient with a long-term, silent, low-placed spinal cord successfully treated by laminotomy alone is reported. The pathomechanism of adult-onset TCS and surgical options are discussed.

Case Report

History and presentation

A 70-year-old woman complaining of progressive numbness and muscle weakness of the lower extremities and inability to walk was referred to our clinic 5 months after

onset. She had no complaints related to the upper extremities, but she was not ambulatory and showed instability even on simple standing. A difficulty in urination was also noted (score 1 of 3 in the urinary scoring in Japanese Orthopaedic Association [JOA] score for thoracic myelopathy). Her symptoms did not change with posture. A pressure sore was found on her sacral skin. She had undergone an operation to remove a “fatty hump” on the sacral skin in her childhood.

Examination

Grade 3 or 4 muscle weakness in Medical Research Council (MRC) scale was found in the lower extremities, including the iliopsoas, quadriceps femoris, and tibialis anterior muscles, and the toe extensors and flexors predominantly on the left side. Deep tendon reflexes were normal in the upper extremities, but showed a brisk patellar tendon reflex (PTR) on the right and a decreased Achilles tendon reflex (ATR) on both sides. Pin-prick sensations were decreased throughout both lower extremities, including the perineal region. The JOA score for thoracic myelopathy was 3 out of the full marks of 11.

An anteroposterior plain radiograph showed spina bifida at the sacrum. The physiological lumbar lordosis was preserved in the lateral view. No spinal instability was found in the dynamic lateral images (Fig. 1). T2-weighted sagittal magnetic resonance (MR) images showed tethering of the spinal cord to the fibrous tissue at the posterior aspect of the sacral spinal canal. A low-placed spinal cord was observed even at the L4/5 and L5/S1 disk levels. Furthermore, an intramedullary high-signal area was found at the L2/3 level (Fig. 2A). The T2-weighted axial MR images showed compression and deformity of the spinal cord at L1/2, L2/3, and L3/4 disk levels by bulged disks and thickened ligamenta flava (Fig. 2B–D). The spinal cord was present at the L5/S1 disk level without compression or deformity (Fig. 2E).

The patient was diagnosed as having a “lumbar myelopathy” due to a combination of tethered cord, degenerative lumbar disk protrusion, and thickening of the ligamenta flava.

Operation and postoperative course

Since the symptoms were progressive, bilateral laminotomy through a posterior approach was performed at the levels of L1/2, L2/3, and L3/4. The laminae of L1, 2, and 3 were fenestrated by preserving the spinous processes and surrounding ligaments (Fig. 3). The numbness of the lower extremities was alleviated, and the urinary problems disappeared shortly after surgery (score 3 of 3 in the urinary scoring in JOA score for thoracic myelopathy). Six weeks after surgery, she became ambulatory with a cane. Decompression of the low-placed spinal cord was confirmed on the postoperative MR images (Fig. 4). The JOA score for thoracic myelopathy improved to 7/11 after surgery and was maintained at 30-month postoperative follow-up.

Discussion

Symptoms and pathophysiology

Adult-onset TCS is rare compared to congenital or childhood TCS, and many cases are asymptomatic until adulthood [8]. Symptoms of adult-onset TCS are similar to those of childhood-onset TCS, for example, low back pain, sensory disturbance and/or motor weakness of the lower extremities, and bowel/bladder dysfunction [1].

In terms of pathology, childhood- and adult-onset TCS are also similar, that is, tight terminal filum, myelomeningocele, lipomyelomeningocele, split cord malformation, or arachnoidal adhesions are major causes of neurological symptoms [10]. The present case also had

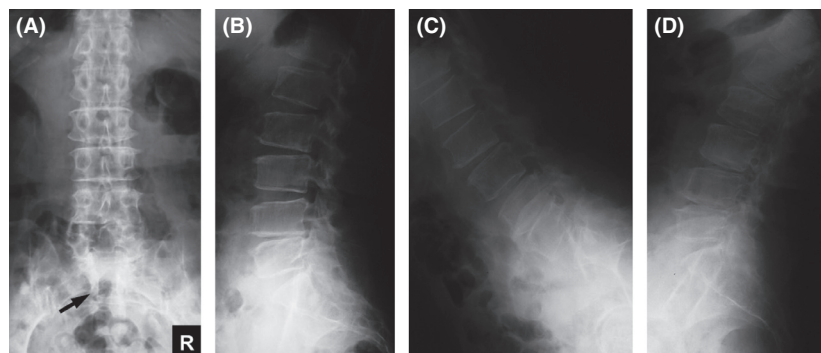


Figure 1. Preoperative plain radiographs. (A) An anteroposterior view reveals spina bifida occulta (arrow). Dynamic lateral images in neutral (B), flexion (C), and extension (D) positions.

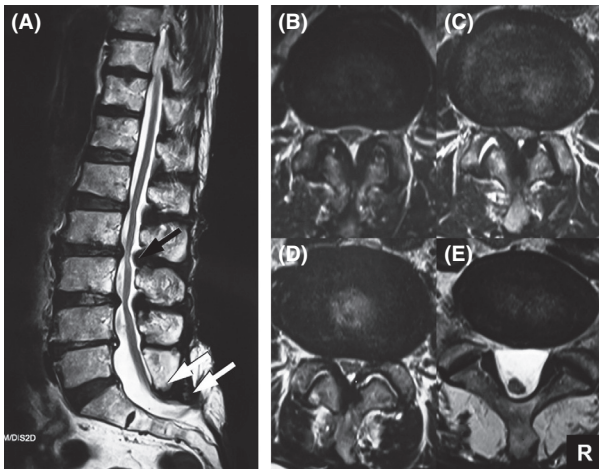


Figure 2. Preoperative T2-weighted magnetic resonance images. (A) A midline sagittal section demonstrates tethering of the spinal cord (white arrows) and an intramedullary high-intensity area at L2/3 (black arrow). Axial images at L1/2 (B), L2/3 (C), L3/4 (D), and L5/S1 (E) disk levels.

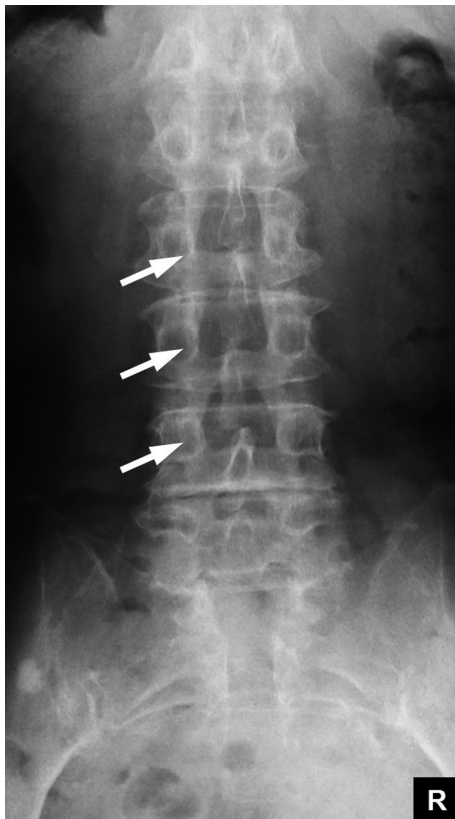


Figure 3. Postoperative plain radiograph. Bilateral laminotomies have been performed at L1/2, L2/3, and L3/4 (arrows).

typical neurological symptoms as previously reported, and she was also suspected to have had a sacral spinal lipoma on MRI with a history of resection of a sacral fatty mass lesion.

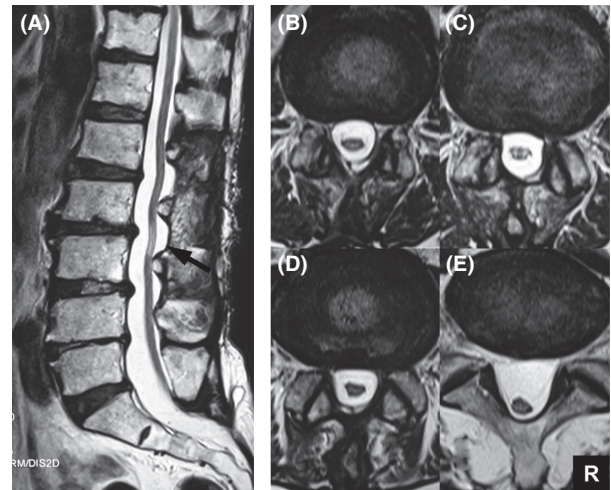


Figure 4. Postoperative T2-weighted magnetic resonance images. (A) Sagittal section. Axial images demonstrating the decompressed spinal cord at L1/2 (B), L2/3 (C), and L3/4 (D) and no obvious change at L5/S1 (E).

The reason why the lesions remain silent for decades until the onset of symptoms in adult- or elderly-onset TCS is still controversial. Stetler et al. [11] reported that the tethering of the conus medullaris itself is not enough to cause symptoms until there is a precipitating factor that increases traction directed to the conus medullaris. Ratliff et al. [1] noted that 60–70% of patients with adult TCS had clear triggering events correlated with a significant worsening of symptoms. Pang et al. hypothesized that the mechanisms were (1) transient stretching of the spine; (2) mechanical narrowing of the spinal canal; and (3) spinal trauma to the pre-existing tethered cord [5, 11]. In the present case, “transient stretching of the spine” is less likely, because the symptoms were not worsened with a specific posture or motion stretching the spinal cord. In addition, the patient had no obvious traumatic episode at onset. Considering the fact that the patient was asymptomatic for more than six decades, the etiology of the present case was assumed to have been gradual, mechanical narrowing of the spinal canal due to protruded intervertebral disks and thickened ligamenta flava, that is, the same as that of spondylotic myelopathy.

Degenerative disk disease following an asymptomatic tethered cord has been reported previously [5, 9, 12–14]. Sostrin et al. [14] and Srinivas et al. [9] reported elderly-onset TCS without traumatic events showing progressive symptoms. Although the spinal cord tethering is conspicuous on imaging studies, the possibility of spinal cord compression, not only tethering itself, should be considered as a potential cause of the neurological deficits, especially in elderly-onset cases. On the other hand, it is difficult to specify the level of the nerve damage as the

precise myelomere of the elongated spinal cord is unclear. In this regard, the level of decompression was determined by the narrowed spinal canal and the compressed spinal cord in T2-weighted MRI in the present case.

Surgical options

The surgical options for symptomatic tethered cords are generally untethering [13, 15] or spinal shortening [16–19]. The purpose of these operations is to relieve the mechanical tension of the tethered cord, that is, stretching of the spinal cord itself needs to be the pathology to justify these surgical methods. Meanwhile, the present case did not show neurological deficits for decades while repetitive mechanical tensions would have been applied to the tethered cord. Moreover, the MRI demonstrated disk protrusions and thickening of ligamenta flava that compressed the tethered cord at multiple levels. These findings indicated that the mechanism of the nerve damage in the present case was the compression of the tethered cord due to degenerative changes in the spinal canal, not tethering of the spinal cord itself. In this situation, release or untethering of the spinal cord may not eliminate the pathomechanism. Instead, decompression of the compressed spinal cord is mandatory and sufficient.

The tethered spinal cord is usually located posteriorly in the dural sac with a terminal filum that clings to the posterior aspect of the sacral dura mater. For this reason, as long as the lumbar lordosis is preserved, the spinal cord is able to shift posteriorly after the removal of ligamenta flava at the stenotic levels. In the present case, posterior decompression without discectomy rescued the compressed spinal cord successfully and alleviated the patient's neurological symptoms. In the considerable number of case series about adult TCS, only Sostrin *et al.* [14] and Srinivas *et al.* [9] reported similar cases of elderly-onset TCS treated successfully only by lumbar laminectomy of the affected tethered cord. The lack of necessity for discectomy makes this operation easier because discectomy by retracting the dural sac at the spinal cord level has a great risk of spinal cord injury. Furthermore, as shown in our case, laminotomy has the benefit of preserving the posterior elements of the lumbar spine, such as spinous processes and surrounding ligaments, so as to avoid postoperative instability compared to wide laminectomy reported in the literature. In this regard, laminotomy is feasible to relieve the pathophysiology.

Understanding the pathophysiology of the neurological symptoms in adult-onset TCS is important to choose the surgical options. As shown in the past case series, untethering or spinal shortening can be indicated when tethering or stretching of the spinal cord itself is the main pathology. On the other hand, as shown in the present case, the

concomitant spinal cord compression due to degenerative changes can have a greater effect on neurological deterioration. In such cases, the less invasive laminotomy can be a feasible surgical option for adult-onset TCS.

Conflict of Interest

None declared.

References

- Ratliff, J., P. S. Mahoney, and D. G. Kline. 1999. Tethered cord syndrome in adults. *Southern Med. J.* 92:1199–1203.
- Yamada, S. 2004. Tethered cord syndrome in adults and children. *Neurol. Res.* 26:717–718.
- Iskandar, B. J., B. B. Fulmer, M. N. Hadley, and W. J. Oakes. 2001. Congenital tethered spinal cord syndrome in adults. *Neurosurg. Focus* 10:e7.
- Klekamp, J. 2011. Tethered cord syndrome in adults. *J. Neurosurg. Spine* 15:258–270.
- Pang, D., and J. E. Jr Wilberger. 1982. Tethered cord syndrome in adults. *J. Neurosurg.* 57:32–47.
- Bowman, R. M., A. Mohan, J. Ito, J. M. Seibly, and D. G. McLone. 2009. Tethered cord release: a long-term study in 114 patients. *J. Neurosurg. Pediatr.* 3:181–187.
- Duz, B., S. Gocmen, H. I. Secer, S. Basal, and E. Gonul. 2008. Tethered cord syndrome in adulthood. *J. Spinal Cord Med.* 31:272–278.
- Huttmann, S., J. Krauss, H. Collmann, N. Sorensen, and K. Roosen. 2001. Surgical management of tethered spinal cord in adults: report of 54 cases. *J. Neurosurg.* 95:173–178.
- Srinivas, S., R. Shetty, and I. Collins. 2012. Symptomatic lumbar disc protrusion causing progressive myelopathy in a low-lying cord. *Global Spine J.* 2:115–118.
- Lee, G. Y., G. Paradiso, C. H. Tator, F. Gentili, E. M. Massicotte, and M. G. Fehlings. 2006. Surgical management of tethered cord syndrome in adults: indications, techniques, and long-term outcomes in 60 patients. *J. Neurosurg. Spine* 4:123–131.
- Stetler, W. R. Jr, P. Park, and S. Sullivan. 2010. Pathophysiology of adult tethered cord syndrome: review of the literature. *Neurosurg. Focus* 29:E2.
- Kawamura, I., Y. Ishido, M. Zenmyo, T. Yamamoto, Y. Kagawa, S. Komiya, *et al.* 2010. Pedicle subtraction osteotomy for adult tethered cord syndrome with lumbar canal stenosis: report of two cases. *Int. J. Neurosci.* 120:735–737.
- Rajpal, S., R. S. Tubbs, T. George, W. J. Oakes, H. E. Fuchs, M. N. Hadley, *et al.* 2007. Tethered cord due to spina bifida occulta presenting in adulthood: a tricenter review of 61 patients. *J. Neurosurg. Spine* 6:210–215.
- Sostrin, R. D., J. R. Thompson, S. A. Rouhe, and A. N. Hasso. 1977. Occult spinal dysraphism in the geriatric patient. *Radiology* 125:165–169.

15. Lapsiwala, S. B., and B. J. Iskandar. 2004. The tethered cord syndrome in adults with spina bifida occulta. *Neurol. Res.* 26:735–740.
16. Kanno, H., T. Aizawa, H. Ozawa, T. Hoshikawa, E. Itoi, and S. Kokubun. 2008. Spine-shortening vertebral osteotomy in a patient with tethered cord syndrome and a vertebral fracture. Case report. *J. Neurosurg. Spine* 9:62–66.
17. Kokubun, S. 1995. Shortening spinal osteotomy for tethered cord syndrome in adults. *Spine Spinal Cord* 8.
18. Kokubun, S. 2003. Shortening spinal osteotomy through posterior approach and its applications. *Seikei Saigai Geka* 46.
19. Kokubun, S., H. Ozawa, T. Aizawa, N. M. Ly, and Y. Tanaka. 2011. Spine-shortening osteotomy for patients with tethered cord syndrome caused by lipomyelomeningocele. *J. Neurosurg. Spine* 15:21–27.