A case of adult onset tight filum terminale associated with ankylosing spondylitis

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Abstract

We report a case of adult onset tight filum terminale with ankylosing spondylitis. A 28-year-old man presented with persistent thigh pain on both sides. Since he was 10 years old, he had occasionally felt low back pain and the pain had gradually increased. Recently his height had decreased by 10 cm with increasing thoracic kyphosis. He felt pain in both thighs and middle back and urinated frequently. Spinal and pelvic X-ray films showed ankylosing spondylitis. Blood examination revealed an increased blood sedimentation rate and C-reactive protein. HLA-B27 was positive. Tight filum terminale with ankylosing spondylitis was diagnosed. After transection of the filum terminale, his thigh pain, middle back pain and frequent urination improved. The tightness of the filum terminale may have been induced by the increased kyphosis due to progressing ankylosing spondylitis.

Introduction

Tight filum terminale (TFT) is a rare spinal disorder thought to be a type of spinal cord traction caused by a tense filum terminale. Since the first report of Garceau in 1953, there have been many reports regarding TFT or filum terminale syndrome, but none has described TFT accompanied by ankylosing spondylitis.

Case report

Patient: A 28-year-old man was admitted with persistent pain in both thighs. Since age 10 he had occasionally felt low back pain, and visited a local physician who prescribed non-steroidal anti-inflammatory drugs; however, the pain gradually increased. Recently his height had decreased by 10 cm and his posture showed thoracic kyphosis. He felt pain in both thighs and middle back. The frequency of urination was about 20 times in the daytime and 4-5 times at night. When he first presented, plain radiography disclosed the characteristic findings of ankylosing spondylitis, such as bamboo spine, thoracic kyphosis and bony fusion of the bilateral sacroiliac joints (Fig. 1). Blood examination revealed an increased blood sedimentation rate and C-reactive protein. HLA-B27 was positive.

Clinical findings: Physical examination on admission disclosed marked limitations in spinal column movement and thoracolumbar kyphosis with disappearance of physiologic lordosis of the lumbar spine. In supine position, straight leg rising test indicated positive at 70 degrees bilaterally. The muscle strength of both lower limbs showed normal. Sensory disturbance was observed bilaterally under the L5 level. Left patellar tendon reflex deteriorated while the other tendon reflex
of the lower extremities showed normal. In prone position, no tenderness on pressure was recognized on the spine and paravertebral muscles. In standing position, his finger-floor distance was 35 cm (Fig. 2). Middle back pain and both thigh pain were provoked by lumbar anterior flexion and neck anterior flexion. These symptoms were reduced by neck extension only. (Positive provocation test)

On MRI, no neural compressive lesions such as spinal cord tumors or disc herniations were recognized (Fig. 3). Myelography revealed a straightened filum terminale (Fig. 4). Cystometry indicated a spastic bladder. According to these clinical findings, we diagnosed TFT with ankylosing spondylitis.

Operative findings: The operation was carried out under general anesthesia. Before the skin incision, an epidural electrode was inserted into the epidural space cranially at the thoracic spinal level. Then S1
laminectomy was performed and the dural tube was opened. The filum terminale was macroscopically recognized at the mid-line of the spine. It appeared redder than the nerve (Fig. 5). We confirmed the absence of spinal cord evoked potentials by electrical stimulation of the filum terminale and the presence of normal spinal cord evoked potentials by stimulation of the cauda equina. After coagulation with a bipolar coagulator, we transected the filum terminale with microscissors upon which the edge of the filum terminale retracted rapidly 20 mm in cranial direction. Three days after the operation, the patient could walk unaided with a soft corset. After transection of the filum terminale, not only the thigh pain but also middle back pain and frequent urination improved.

Discussion

Some authors have reported ankylosing spondylitis accompanied with neurological symptoms: atlantoaxial dislocation\(^3\), vertebral fracture\(^4\), multiple sclerosis\(^5\) and cauda equina syndrome\(^6\). In the present case, all the above diseases except cauda equina syndrome can be excluded on the bases of the clinical and imaging findings. A total of 71 cauda equina syndrome cases

![Sagittal View of the Thoracic Spine(T2)](image1)

![Sagittal View of the Lumbar Spine(T2)](image2)

Fig. 3 On MRI, no neural compressive lesions such as spinal cord tumors or disc herniations were recognized.

![Root sleeves show normal opacification](image3)

![Allow indicates filum terminale](image4)

Myelography

Fig. 4 Myelography revealed a straightened filum terminale.
have been reported. Of these, 56 were treated by oral medicine only and 15 were treated surgically. Laminectomy was performed in 11 cases and lumbo-peritoneal shunt in 4 cases. Improvement was obtained in 6 cases, 7 cases were unchanged and 2 cases worsened following operation.

The pathophysiology of TFT is thought to involve traction myelopathy caused by tense filum terminale, but details are still unclear. Tightness of the filum terminale is considered to be a congenital anatomic malformation, and many juvenile cases of TFT have been reported. On the other hand, not a few authors reported TFT with adults onset. This disease has been considered a rare disorder of the spinal cord diagnosable only on an exclusionary bases.

In our observations of the early several patients of TFT, we had noticed that their low back pain or leg pain were provoked by lumbar anterior flexion and neck flexion.

**Fig. 5** Operative findings. The filum terminale can be recognized macroscopically at the mid-line of the spine, because it is redder than the nerve.

**Fig. 6** Provocation test.
anterior flexion and this pain was reduced by neck extension only. We considered that this phenomenon reflects the reinforcement and relaxation of the spinal cord tension. On the bases of these considerations, we have utilized this phenomenon as a TFT provocation test (Fig. 6). Since 1994 we have employed this provocation test for the early diagnosis of this disease and have found that this disorder is not as rare as previously believed. From 1991 to 2001, we encountered 52 cases of TFT all of which were treated by transecting the tense filum terminale and 98% of these patients were relieved of their low back pain or leg pain or both. Clinical symptoms included low back pain in 45 cases (87%), leg pain in 37 (71%), leg numbness in 35 (67%), and bladder-bowel dysfunction in 46 (90%). We established 5 diagnostic criteria for TFT: 1) low back pain, 2) non-dermatomal leg pain, 3) bladder-bowel dysfunction, 4) spinal stiffness, 5) positive provocation test. The present case satisfied all 5 criteria. Spinal stiffness is the main symptom of ankylosing spondylitis. After transecting the filum terminale, the finger floor distance decreased to 20 cm from 35 cm preoperatively. In this case, the onset of neurological symptoms is extremely unusual. In our review of the literature on TFT and ankylosing spondylitis, we were not able to find any reports of cases similar to this case. In cases with back pain, leg pain, frequent urination and progressing spinal kyphosis, tightening of the filum terminale should be included in the differential diagnosis.

Conclusion

We reported a case of TFT with ankylosing spondylitis. After transection of the filum terminale, the patient's symptoms of thigh pain, middle back pain and frequent urination were improved. We considered the cause of TFT to have been increased kyphosis due to ankylosing spondylitis. TFT should be considered in cases of ankylosing spondylitis accompanied by leg pain or frequent urination, and transection of the filum terminale should be considered while the neural dysfunction is still reversible.

References

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強直性脊椎炎に合併した脊髄終末症候群の1例

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【要旨】Tight Filum Terminale (以下 TFT) は、脊髄終末の異常な緊張により腰痛や下肢痛、膀胱直腸障害などの症状を呈する疾患である。本症は特徴的画像所見に乏しく、従来より診断は除外診断とされ術前の診断は困難であったが、我々は独自に誘発テストを考察した際に、診断率向上に成果を上げている。今回成人男性の強直性脊椎炎の症例で脊柱後弯の進行とともに TFT と思われる下肢神経症状が出現した症例を経験した。症例は28才男性、最近の数年で伸長が約10cm短縮し胸椎後弯を呈するようになった。その頃から背部痛と両下肢痛および頻尿を自覚するようになった。近医にて消炎鎮痛剤の投与を受けるも軽快せず当科を受診した。初診時脊柱単純 X 線正面像で明らかな Banboo spine を呈し、側面像では胸椎後弯の増強を認めた。骨盤正面像では仙腸関節の強直を認め血液生化学的検査では ESR 41mm/h、CRP 2.1mg/dl、HLA－B27 隣性で強直性脊椎炎として矛盾のない所見であった。神経学的には下肢筋力低下はないが両下肢に知覚障害を認め体幹前屈にて背部痛と両大腿部痛が誘発された。また日中約20回夜間4～5回の頻尿を認めた。画像では脊髄圧迫所見は見られず、TFT 誘発テストが陽性であることなどから TFT と診断し終末切離術を施行した。術後背部痛と両下肢痛および膀胱直腸障害ともに著明に軽快し患者は満足している。内外の文献検索上強直性脊椎炎に合併した TFT の報告は見られず極めて稀な症例と考えられたため、症例報告とともに診断に至った根拠について考察し報告する。

〈Key words〉脊髄終末症候群、成人発症、強直性脊椎炎