



하부천골 제2,3구역의 부전골절에 의한 말총증후군 환자: 증례 보고

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Cauda Equina Syndrome Caused by a Sacral Insufficiency Fracture in the Lower Part of Zones 2 and 3: A Case Report

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Sacral insufficiency fractures (SIFs) often cause referred low back pain, and are rarely accompanied by cauda equina syndrome (CES). We report a case of CES caused by SIF in the lower part of zones 2 and 3, which is not a typical location. A postmenopausal woman often sat on a chair for long periods, and pain in the coccyx and perianal area occurred. After 4 years, during which time the pain worsened and she developed urinary and fecal incontinence, magnetic resonance imaging showed a SIF involving the S3-S5 vertebrae. Prolonged latency of the bulbocavernosus reflex was confirmed through an electrodiagnostic study, and detrusor underactivity was confirmed through a urodynamic study, leading to a CES diagnosis. Unlike typical cases of osteoporotic SIF, the fracture occurred at an atypical location. We believe that long periods of sitting caused a different weight-bearing pattern of the sacrum from that found in typical cases.

Keywords: Fractures, stress; Cauda equina syndrome; Osteoporosis

Introduction

Sacral insufficiency fractures (SIFs) occur when the sacral bone deteriorates and becomes too weak for the stress of weight bearing. With an incidence rate of only 1.0% to 1.8%, they are often missed due to nonspecific symptoms and variable X-ray findings. Since most SIF patients complain of low back pain, their exams are often focused on the lumbar region rather than the sa-

crum, which leads to misdiagnosis [1].

Cauda equina syndrome (CES) is a disease in which impairment occurs in the cauda equina, a bundle of axons leading to the distal end of the spinal cord. Symptoms include low back pain, unilateral or bilateral sciatica, reduced sensation in the saddle area, reduced sexual function, fecal incontinence, bladder dysfunction, and lower limb weakness. The most common cause is a herniated lumbar intervertebral disc; various conditions such

as epidural abscess, spinal epidural hematoma, diskitis, tumor, spinal stenosis, and aortic obstruction can also cause CES. However, SIF is generally not included in the list of etiologies. It is rarely considered when assessing the cause of CES [2].

We herein report a case CES caused by SIF in a middle-aged woman who complained of sphincter dysfunction as a main symptom.

Case Report

A 56-year-old woman visited our outpatient clinic with coccyx and perianal area pain accompanied by urinary and fecal incontinence. She had gone through menopause 4 years prior, just before the pain occurred, and had no history of pelvic radiation or rheumatoid arthritis. Also 4 years prior, she began sitting in a chair for long periods to study for exams; from that point on, she experienced pain in the area where her hips touched the chair. She received 12 steroid injections, including several caudal epidural injections, to improve her symptoms. When an injection was given, the pain was relieved. However, the pain returned in less than a week, and the injection was repeated. At the time, lumbar spine magnetic resonance imaging (MRI) revealed no specific findings. Two months before visiting the hospital, she reported an open feeling in her anus, and, when severe, she rated her pain at 9 on the visual analog scale. According to the patient, there was no history of trauma, and a physical examination revealed hypoesthesia in the S3 and S4 dermatomes. Voluntary anal contraction also revealed a hypotonic pattern, and the bulbocavernosus reflex (BCRL) was decreased.

Due to suspicion of CES, electromyography, nerve conduction

study and BCRL study were conducted. Electrodiagnostic study showed abnormal spontaneous activities in bilateral tensor fascia lata, gluteus maximus, tibialis anterior, peroneus longus, extensor hallucis longus, gastrocnemius and bulbocavernosus muscles. Peripheral nerves conducted normally in both motor and sensory nerve fibers of bilateral lower extremities. BCRL study showed prolonged latencies for both sides. In addition, an urodynamic study showed a correlation between CES and 100 mL postvoid residual urine and detrusor underactivity. A lumbar spine MRI with contrast enhancement showed SIF involving the S3-S5 vertebrae and presacral soft tissue edema. According to the Denis classification, SIF had occurred not only in bilateral zone 1 but also in bilateral zones 2 and 3 (Fig. 1) [3]. The lumbar spine MRI was not clear at the coccyx level, so an additional sacrum computed tomography scan with contrast enhancement was performed, revealing permeative bone destruction and sclerosis involving the S3 and S4 vertebrae and the coccyx with ill-defined enhancing presacral soft tissue (Fig. 2). The patient's bone mineral density was evaluated using dual energy X-ray absorptiometry; the femoral neck T-score was -0.7, and the lumbar spine L1-L4 T-score was -3.3. The patient's 25-hydroxy vitamin D was 32.3 ng/mL (25-80 ng/mL), and her parathyroid hormone was 39 pg/mL (15-65 pg/mL), both of which were within the normal range.

Discussion

SIFs are less likely to be accompanied by neurological complications than sacral fractures caused by high-energy trauma.

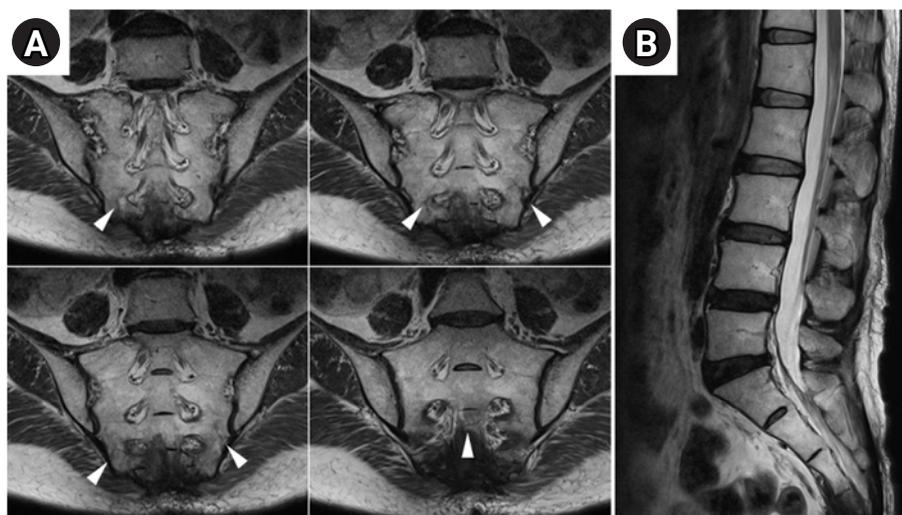


Fig. 1. (A) Lumbar-spine magnetic resonance imaging (MRI) (coronal view) shows multiple sacral insufficiency fractures including all bilateral zones 1, 2, and 3 (arrowheads). (B) Lumbar-spine MRI (sagittal view) shows no specific findings except for a mild degree of disc bulging at L5-S1.

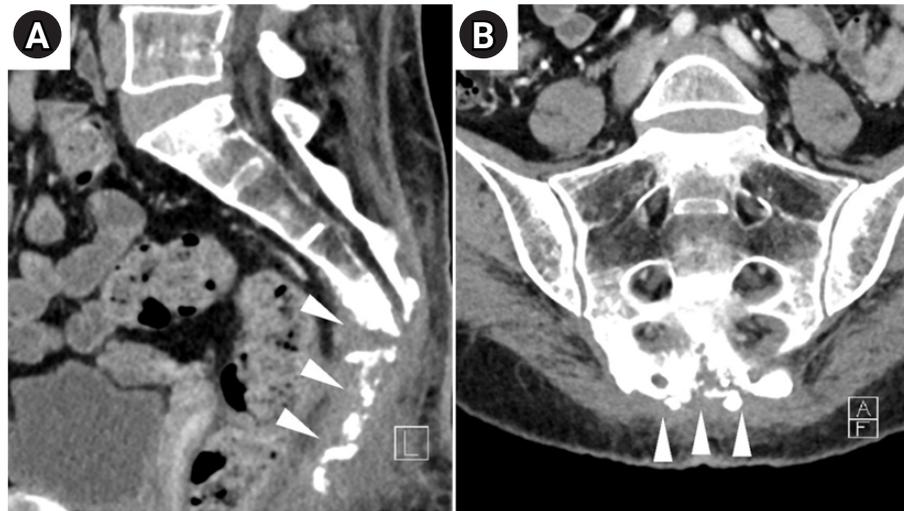


Fig. 2. (A) Sacrum computed tomography (CT) (sagittal view) shows permeative bone destruction and sclerosis involving S3, S4, and the coccyx with ill-defined enhancing presacral soft tissue (arrowheads). (B) Sacrum CT (coronal view) shows bilateral sacral fractures (arrowheads).

Finiels et al. [4] found neurological complications in 14 of 493 SIF patients; however, in all of them, initial symptoms involved some kind of pain, and neurologic symptoms were identified only in the form of delayed complications. Later, Muthukumar et al. [5] introduced four CES cases diagnosed with SIF, and these patients commonly showed unilateral acute insufficiency fractures involving zone 1 from the S1 to S3 vertebrae, extending from the sacroiliac joint to the lateral margin of the sacral foramen. This was the first published report of CES as a presenting feature of SIF [3]. Therefore, the present case is unique in that, unlike typical SIF cases, the fracture does not involve the sacral ala and invades only the S3-S5 vertebrae, the lower part of the sacrum. In addition, CES was diagnosed more objectively than in previous cases; the BCRL was numerically evaluated using an electrodiagnostic study, and an urodynamic study confirmed the urinary incontinence pattern to be overflow incontinence.

Insufficiency fractures are stress fractures that happen in everyday life without a high-energy trauma. They are more likely to occur to sacra with reduced elastic resistance [6]. The sacroiliac joint acts as a stress reliever between the trunk and the lower limb, and it buffers the torsional stress caused by alternating swing and stance phases while walking to prevent direct stress to the sacrum [7].

Osteoporotic SIFs usually start from the sacral ala, which has the highest ratio of trabecular to cortical bone [8]. According to the Denis classification, this corresponds to zone 1, located between the neural foramina and the sacroiliac joint, and it usually does not invade the neural foramina [6]. It is uncommon for SIFs to first occur in zones 2 or 3, reflected in the rare occurrence

of neurological complications in SIF [9].

Linstrom et al. [8] confirmed the fracture locations in 108 SIF patients and most of them showed typical vertical parasagittal plane fractures starting at the unilateral or bilateral sacral ala and continuing to the sacroiliac joint, and the fracture progressed in a characteristic “H” pattern over time, crossing at the upper S2 or lower S1 levels. However, a few cases showed atypical fracture patterns such as isolated transverse only sacral fractures. These were explained by confounding factors such as extreme amounts of sacral lordosis, unusual stress, or advanced osteoporosis.

In the present case, the fracture occurred at a location different from typical osteoporotic SIF. In addition, a hip and lumbar spine MRI taken 5 months before the start of sphincter dysfunction showed no fracture, but when the images were retaken after only 7 months, both vertical and horizontal fractures were found (this differs from the general course of SIF). Therefore, it can be assumed that stress was caused by a mechanism different from that caused by walking. This patient did not have severe sacral lordosis or severe osteoporosis, and the fracture was not accompanied by the confounding factors mentioned by Linstrom et al. [8] such as Tarlov cysts, sacral masses, or history of pelvic radiation. As to the characteristic history of this patient, over the course of about four years, she studied for more than 8 hours a day while sitting on a hard chair. This may have caused the weight of her upper body to be applied downward instead of being distributed to the bilateral lower extremities. Also at that time, she went through menopause and received 12 steroid injections, including repetitive caudal epidural injections, which may have increased her risk of fracture by lowering the bone marrow

density in the coccyx [10]. In this situation, we believe SIF occurred after coccyx fracture, as stress was applied vertically to the coccyx and sacrum. This would explain the vertical and horizontal fractures that occurred at the S3-S5 level, unlike most SIFs that usually start at the sacral ala.

The implications of this case are as follows. When patients at high risk of osteoporosis complain of back and buttock pain, care providers should check if there have been recent long periods of sitting. If so, they should include the sacrum in the lumbar spine MRI. CES must be differentiated because, as shown in this case, unlike typical SIF, fracture can occur at the S3-S5 level including zones 2 and 3. It should also be noted that, if multiple caudal epidural steroid injections are inevitably performed, the patient's local and systemic bone marrow density may be reduced. Careful attention should be given to the occurrence of osteoporotic fractures.

Conflict of Interest

No potential conflict of interest relevant to this article was reported.

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