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CASE REPORT

Proximal Junctional Failure After Long-Segment Instrumentation for Degenerative Lumbar Kyphosis With Ankylosing Spinal Disorder

Daisuke Ikegami, MD, PhD, Takashi Matsuoka, MD, Yuji Miyoshi, MD, Yoichi Murata, MD, and Yasuaki Aoki, MD, PhD

Study Design. Case report.

Objective. We report a case of proximal junctional failure at the ankylosed, but not the mobile, junction after segmental instrumented fusion for degenerative lumbar kyphosis with ankylosing spinal disorder.

Summary of Background Data. Proximal junctional failure (PJF) and proximal junctional kyphosis (PJK) are important complications that occur subsequent to long-segment instrumentation for correction of adult spinal deformity. Thus far, most studies have focused on the mobile junction as a site at which PJK/PJF can occur, and little is known about the relationship between PJK/PJF and ankylosing spinal disorders such as diffuse idiopathic skeletal hyperostosis.

Methods. The patient was an 82-year-old female with degenerative lumbar kyphosis. She had abnormal confluent hyperostosis in the anterior longitudinal ligaments from Th5 to Th10. The patient was treated operatively with spinal instrumented fusion from Th10 to the sacrum.

Results. Four weeks subsequent to initial surgery, the patient developed progressive lower extremity paresis caused by the uppermost instrumented vertebrae fracture (Th10) and adjacent subluxation (Th9). Extension of fusion to Th5 with decompression at Th9–Th10 was performed. However, the patient showed no improvement in neurological function.

Conclusion. PJF can occur at the ankylosing site above the uppermost instrumented vertebrae after long-segment instrumentation for adult spinal deformity. PJF in the ankylosed spine may cause severe fracture instability and cord deficit. The ankylosed spine should be integrated into the objective determination of materials contributing to the appropriate selection of fusion levels.

Key words: proximal junctional failure, proximal junctional kyphosis, adult spinal deformity, degenerative lumbar kyphosis, idiopathic skeletal hyperostosis, ankylosing spinal disorder, spinal instrumented fusion, fracture, subluxation, paresis.

Level of Evidence: 3

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Proximal junctional failure (PJF) is an important complication that occurs subsequent to long-segment instrumentation for correction of adult spinal deformity. The phenomenon of PJF is distinct from proximal junctional kyphosis (PJK), in that PJF includes mechanical failure and/or spinal instability.[1] Thus far, many studies have focused on mobile junctions as a site at which PJK/PJF can occur,[2,3] and little is known about the relationship between PJK/PJF and ankylosing spinal disorders such as diffuse idiopathic skeletal hyperostosis (DISH). We report a case of PJF at the ankylosed junction, but not at the mobile junction, after segmental instrumented fusion for degenerative lumbar kyphosis with DISH. This case report suggests that PJF in the ankylosing spine may cause severe fracture instability and cord deficit.

CASE REPORT

An 82-year-old female had been experiencing low back pain since 2007. She presented to the hospital in June 2014 complaining of increasing low back pain. Her neurological examination results were normal. Standing sagittal radiograph showed decreased lumbar lordosis (Figure 1A). A lateral radiograph of the thoracic spine and a sagittal reconstructed computed tomographic scan revealed abnormal confluent hyperostosis in the anterior longitudinal ligaments and local reduction of trabeculae in the anterior vertebral body above Th10 (Figure 1B, C). A dual-energy x-ray absorptiometry showed a right femoral neck T score of −0.6, which suggested an absence of general osteopenia. Therefore, the patient was found to have lumbar degenerative kyphosis in addition to DISH.

From the Department of Orthopaedic Surgery, Japanese Red Cross Society Himeji Hospital, Hyogo, Japan.

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Address correspondence and reprint requests to Daisuke Ikegami, MD, PhD, Department of Orthopaedic Surgery, Japanese Red Cross Society Himeji Hospital, 1-12-1 Shimoteno, Himeji, Hyogo 670-8540, Japan; E-mail: daikegami@gmail.com

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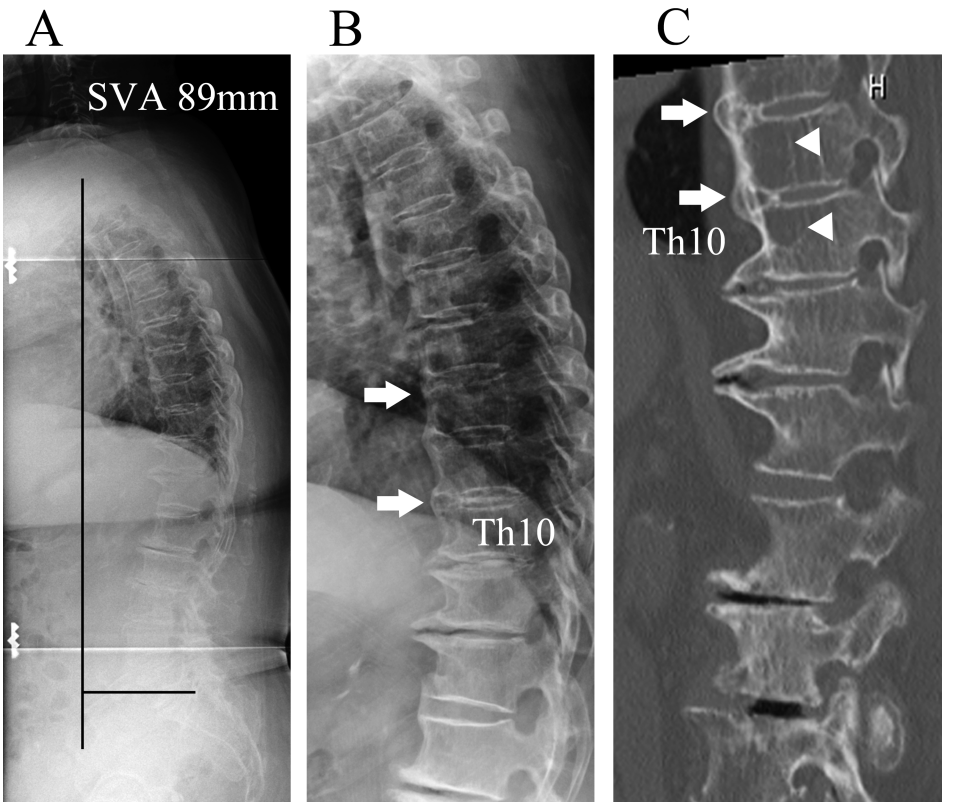

Figure 1. A, Preoperative standing sagittal radiograph demonstrating decreased lumbar lordosis and C7 plumb line shifted to the anterior by approximately 9 cm (lumbar lordosis = 30°, pelvic tilt = 31°, sagittal vertical axis = 89 mm, pelvic incidence-lumbar lordosis = 27°). **B,** Preoperative lateral radiograph of the thoracic spine revealing abnormal confluent hyperostosis in anterior longitudinal ligaments from Th5 to Th10 (arrows). **C,** Preoperative sagittal reconstructed computed tomographic scan demonstrating abnormal confluent hyperostosis in the anterior longitudinal ligaments (arrows) and local reduction of trabeculae in the anterior vertebral body (arrowheads) above Th10. SVA indicates sagittal vertical axis.

In October 2014, we performed spinal instrumented fusion from Th10 to the sacrum. Immediately postoperatively, a lateral radiograph showed increased lumbar lordosis (Figure 2A, B). Two weeks subsequent to the operation, severe thoracic back pain occurred while the patient attempted to rise from a bed. Four weeks subsequent to the initial operation, the patient developed progressive lower extremity paresis and loss of sensation distally from the midthorax. Lateral radiograph and lateral computed tomographic reconstruction revealed junctional fracture of Th10 and adjacent subluxation of Th9 (Figure 3A, B). These physical and image-based findings confirmed the diagnosis of spinal cord injury caused by fracture of the uppermost instrumented vertebra and adjacent subluxation.

The patient underwent extension of fusion to Th5 with decompression at Th9–Th10 (Figure 4). We chose Th5 as the uppermost instrumented vertebra in the revision surgery because local reduction of trabeculae in the anterior vertebral body was slightest at this level. After this surgery, the patient began to show progressive relief of thoracic back pain but no improvement in neurological function.

DISCUSSION

This case suggested 2 clinical issues. First, PJF can occur at the ankylosing site, adjacent to the uppermost instrumented vertebra, after long-segment instrumentation for adult spinal deformity. Two primary ankylosing spinal disorders, DISH and ankylosing spondylitis, lead to ankylosis of the spinal column. DISH was first described as "senile ankylosing hyperostosis of the spine" by Forestier and Rotes-Querol[4] and is characterized by an ossifying diathesis with spinal and peripheral enthesopathy. The prevalence of DISH in patients aged 50 years or older is reported to be greater than 15%.[5,6] Therefore, it seemed reasonable to think that many patients with adult spinal deformity have an ankylosing spine. Many risk factors for PJF, such as older age, large abnormal preoperative sagittal parameters, greater curvature correction, and fusion to the lower lumbar and sacrum, were identified.[1,7–10] However, a relationship between the ankylosing spine and PJF has not been reported to date. To the best of our knowledge, this is the first reported case in which PJF occurred at the ankylosing site of the vertebral column.

Second, PJF in the ankylosing spine may cause severe fracture instability and cord deficit. PJF includes both kyphosis and structural failure of the vertebral body and/or posterior ligament complex.[1] A fracture of the uppermost instrumented vertebra, followed by adjacent vertebral subluxation, has a strong possibility of neurological deficit *via* spinal cord compression.[11] In patients with DISH, ankylosed spinal segments can act on the long lever arm and result in increased fracture instability.[12,13] These prior reports led to the presumption that, in our case report, a preoperatively existent ankylosed spinal segment above Th10 and long fusion with instrumentation from Th10 to the sacrum increased junctional stress concentration synergically, which resulted in severe fracture instability and cord deficit.

In our case, local reduction of trabeculae was identified in the anterior vertebral body above the uppermost instrumented

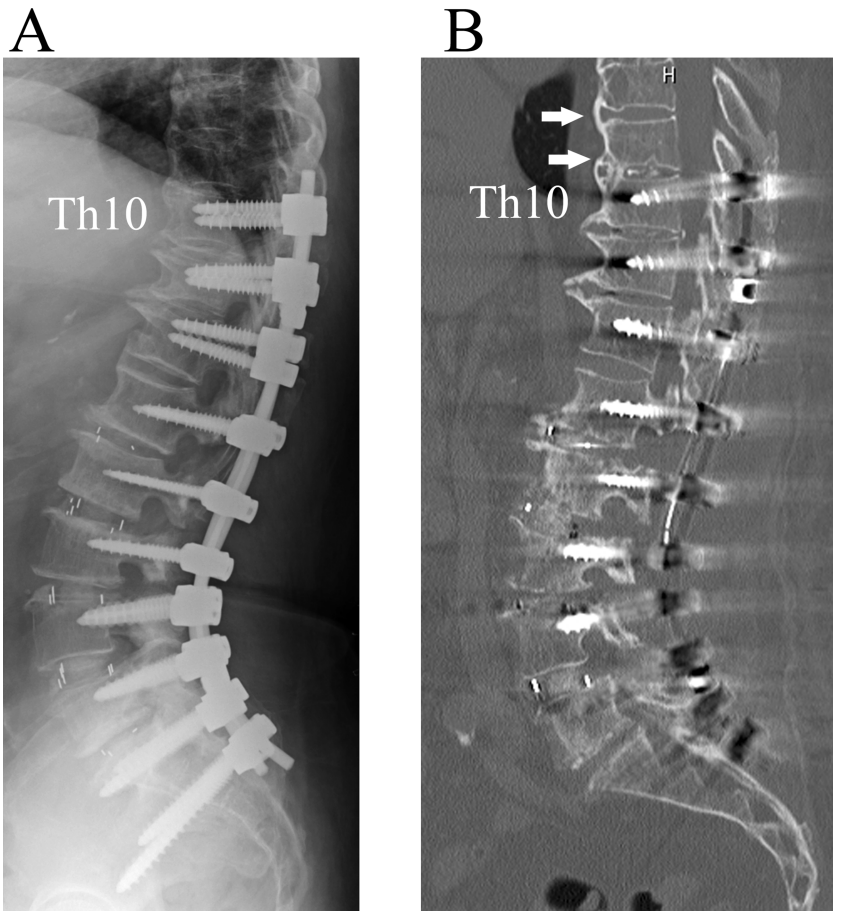

Figure 2. A, Postoperative lateral radiograph demonstrating increased lumbar lordosis and posterior instrumentation from Th10 to the sacrum. **B,** Postoperative sagittal reconstructed computed tomographic scan revealing confluent hyperostosis in the anterior longitudinal ligaments (arrows) above the uppermost instrumented vertebra (Th10).

vertebra (Th10). Stress shielding, which occurred as a result of load transfer *via* ossified ligaments, leads to weakness of the vertebral body.[13] These biomechanical properties may also have contributed to the proximal junctional fracture observed in our case, despite the absence of general osteoporosis.

In conclusion, PJF can occur at the ankylosing site above the uppermost instrumented vertebra after long-segment instrumentation for adult spinal deformity, and PJF in the ankylosing spine may cause severe fracture instability and cord deficit. Selection of fusion levels in adult spinal deformity

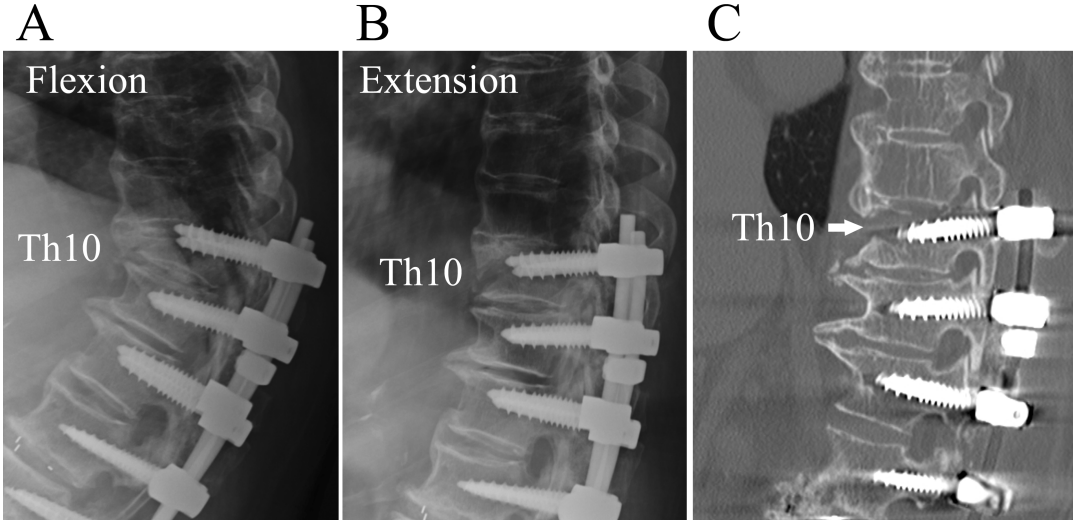

Figure 3. A and B, Flexion-extension radiographs of the thoracic spine demonstrating junctional fracture of Th10 and adjacent subluxation of Th9. **C,** Sagittal reconstructed computed tomographic scan revealing junctional fracture through the anterior superior body and superior endplate of Th10 (arrow).

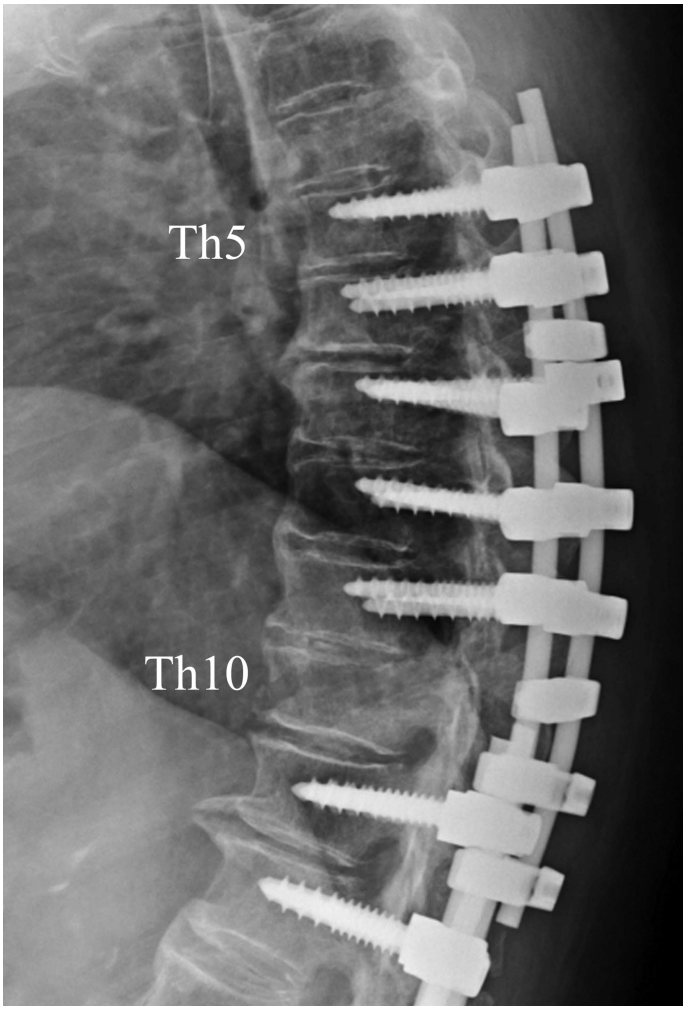

Figure 4. Lateral radiograph after revision surgery showing extension of fusion to Th5.

is an important consideration in planning surgery to prevent PJK and PJF. We must be aware that ankylosing sites above the uppermost instrumented vertebra may lead to serious PJF and permanent neurological deficit. The ankylosed spine should be integrated into the objective determination materials contributing to the appropriate selection of fusion levels. To prevent this complication, we recommend fusing more than 3 segments to ankylosing levels if fusion to ankylosing sites is necessary. More importantly, proximal extension to the ankylosing site should not be limited to the vertebra in which local reduction of trabeculae occurs as a result of stress shielding. Further reports should be accumulated to determine the prevalence and prognosis of PJF in the ankylosing spine.

➢ Key Points

- ❑ PJF can occur at the ankylosing site above the uppermost instrumented vertebrae subsequent to long-segment instrumentation for adult spinal deformity.
- ❑ PJF in ankylosing spine may cause severe fracture instability and cord deficit.
- ❑ The ankylosed spine should be integrated into the objective determination of materials contributing to the appropriate selection of fusion levels.

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