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Case Report

Internal fixation for fragility fracture of pelvis in a patient with ankylosing spondylitis[☆]Hisanori Gamada^{a,b,*}, Yohei Yanagisawa^a, Yusuke Eda^b, Shotaro Teruya^a, Masashi Yamazaki^a^a Department of Orthopaedic Surgery, Faculty of Medicine, University of Tsukuba, Tsukuba, Japan^b Department of Orthopaedic Surgery, Ibaraki Western Medical Center, Chikusei, Japan

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ABSTRACT

Ankylosing spondylitis is a common inflammatory rheumatic disease with a prevalence of 0.1%–1.4%. The most common vertebral fractures associated with ankylosing spondylitis are cervical spine injuries due to low-energy trauma, whereas pelvic fractures are rare. Conversely, fragility fracture of the pelvis is a fracture of the pelvic ring caused by low-energy trauma with a background of bone fragility. In recent years, minimally invasive surgery for early mobilization of displaced fragility fracture of the pelvis has been reported. We report herein a case of a 91-year-old male with ankylosing spondylitis who underwent internal fixation for fragility fracture of his pelvis. He was brought to the emergency room with a complaint of pain in the right hip after a fall from a standing position at home. Computed tomography showed a fracture of the right pubis and a fracture, which crossed the sacrum from the ilium with a maximum dislocation. There was also extensive ossification of the anterior longitudinal ligament in the thoracolumbar spine and bony ankylosis of both sacroiliac joints. The diagnosis after the injury was fragility fracture of the pelvis, which complicated by ankylosing spondylitis. The fracture type did not match the Rommens and Hofmann classification criteria. Iliac intramedullary stabilization was performed in accordance with the treatment of Rommens and Hofmann classification type IIIa. The patient's pain reduced a day after the surgery, and he was able to use a wheelchair. He was able to walk with a cane two months after the surgery, and bone union was achieved four months postsurgery.

Iliac intramedullary stabilization was useful in patients with atypical fractures a) that did not fit the Rommens and Hofmann classification criteria owing to the presence of ankylosing spondylitis, and b) in cases wherein strong fixation was considered necessary.

Introduction

Ankylosing spondylitis (AS) is a common inflammatory rheumatic disease with a prevalence in the range of 0.1%–1.4% [1,2]. Owing to the long lever arm and fragility of the bone caused by ankylosis, even minor trauma can lead to fracture, and patients with

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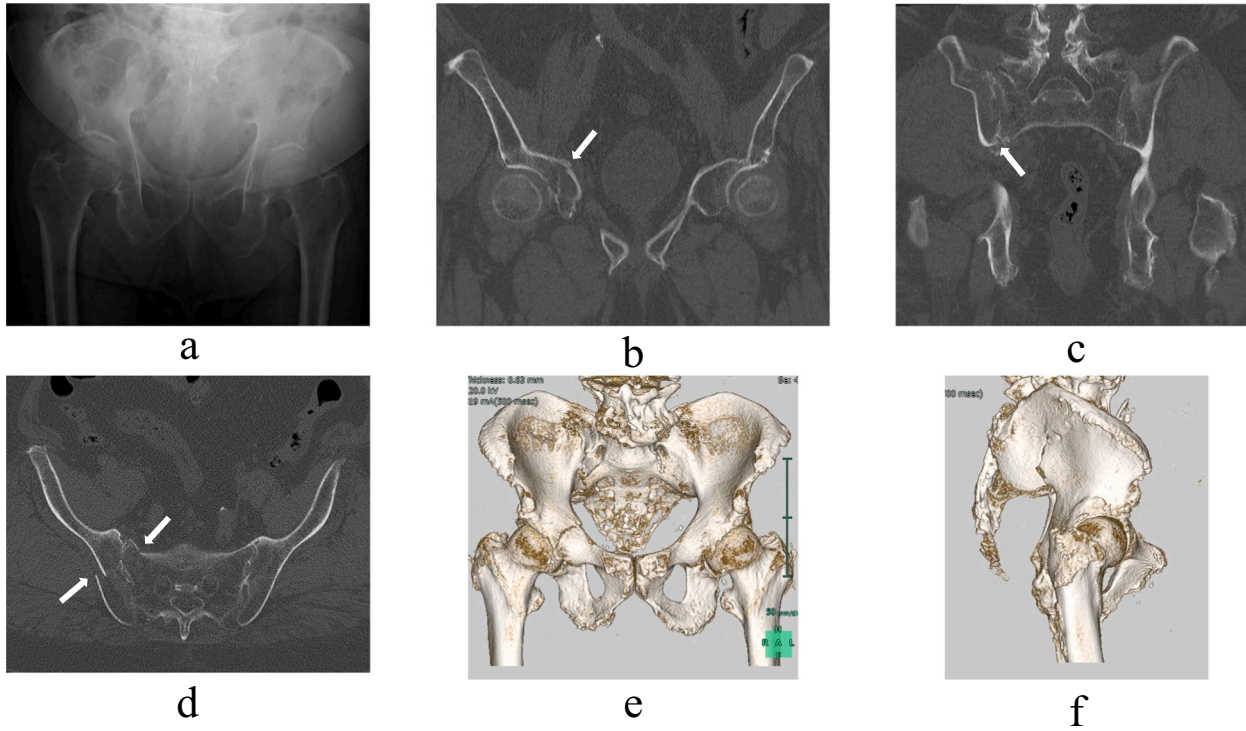


Fig. 1. Plain radiograph on initial examination (a). Computed tomography (CT) showed a fracture of the right pubis (b: coronal view, arrow) and a fracture crossing the sacrum from the ilium with a maximum dislocation of 6 mm (c: coronal view, d: axial view, arrows). Three-dimensional CT renderings of the pelvic area (anterior–posterior and right–left views) (e, f).

AS are associated with a four times higher likelihood to suffer fractures compared with patients without AS [3]. The most common vertebral fractures associated with AS are cervical spine injuries owing to low-energy traumas, whereas sacral and pelvic fractures are rare [3]. Conversely, fragility fracture of the pelvis (FFP) is a fracture of the pelvic ring caused by low-energy traumas in cases wherein there is a background of bone fragility. FFP incidents have been increasing with the aging of the population [4]. In recent years, minimally invasive surgery performed for the early mobilization of displaced FFP has been reported based on the Rommens and Hofmann classification criteria [4]. We report herein the successful outcome of iliac intramedullary stabilization (ILIS) in a patient with AS and atypical FFP that did not fit the Rommens and Hofmann classification criteria [5].

Case presentation

A 91 year old man (height: 157 cm, weight: 78.3 kg, body mass index [BMI] 31.8) was brought to the emergency room with a complaint of pain in the right hip after a fall from a standing position at his home. When he arrived at the hospital, he had difficulty moving, owing to the pain. No neurological symptoms were observed. Computed tomography (CT) showed a fracture of the right pubis and a fracture crossing the sacrum from the ilium with a maximum dislocation of 6 mm (Fig. 1). There was extensive ossification of the anterior longitudinal ligament as well in the thoracolumbar spine and bony ankylosis of both sacroiliac joints (Fig. 2). He also experienced limited lumbar spine motion, which met the New York criteria for AS [6].

We diagnosed the patient with FFP, which was complicated by AS (after the injury). The fracture type was a displaced unilateral posterior injury, type III of the Rommens and Hofmann classification. However, due to the presence of bony ankylosis of the sacroiliac joint, which did not function as a joint, and the atypical shape of the iliac fracture, we considered that the fracture type was neither type IIIb nor type IIIa but a variant of type III that did not fit the Rommens and Hofmann classification criteria [4].

At the time of admission, the mean value of dual-energy X-ray absorptiometry of the left femur in young adults was 71 %. The patient had a medical history of hypertension and asthma, and his physical status classification based on the American Society of Anesthesiologists was equal to two. He had difficulty rolling over, owing to the pain, and surgery was performed on the fourth day of admission. ILIS was performed in accordance with the treatment of Rommens and Hofmann classification type IIIa [5].

Surgery was performed with general anesthesia in a prone position. An 8 cm skin incision was made along the posterior superior iliac spine bilaterally, and two long and thick iliac screws were inserted on each side. The left and right screws were then connected with rods, and the rods were connected with transverse connectors. The operation time was 162 min, and the blood loss was 690 g. The diameters and lengths of the iliac screws were in the range of $\Phi 10$ –100 mm distally on both sides, and $\Phi 9$ –80 mm and 90 mm proximally (Fig. 3).

The postoperative weight bearing was 1/2 PWB immediately after surgery in consideration of the obese patient status (BMI 31.8). Thereafter, the weight increased according to the pain, and the full weight was applied 4 weeks postsurgery. Weekly teriparatide injections were introduced postoperatively. His pain improved the day after surgery, and he was able to use a wheelchair. He was able to walk with a cane 2 months after surgery, and bone union was achieved 4 months postsurgery (Fig. 4). At 15 months postoperatively, he had no complaints of pain, and he returned to his preinjury life. The modified Majeed score (maximum 76 points as the items “work” and “sexual intercourse” were omitted) [7] was 76 points at 15 months postoperatively.

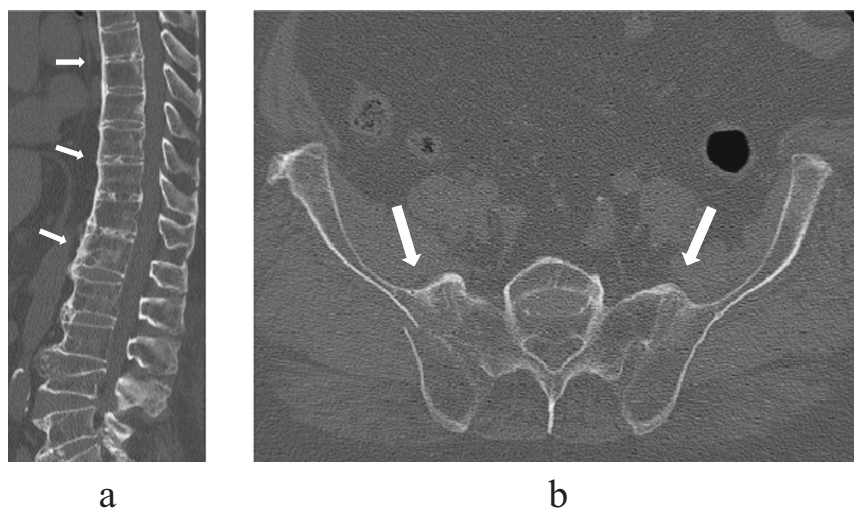


Fig. 2. Computed tomography showed extensive ossification of the anterior longitudinal ligament in the thoracolumbar spine (a: sagittal view, arrow) and bony ankylosis of both sacroiliac joints (b: axial view, arrow).

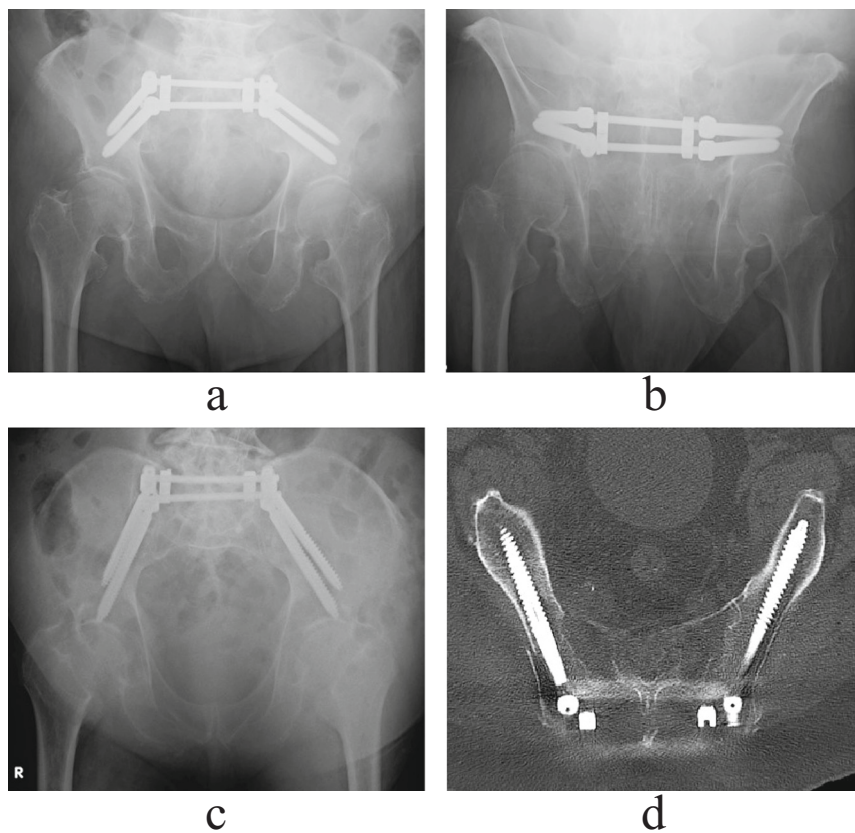


Fig. 3. Postoperative pelvic anteroposterior, outlet, and inlet views (a, b, c). Extremely long iliac screws (sizes in the range of $\Phi 10$ – 100 mm) were inserted bilaterally in postoperative computed tomography axial view (d).

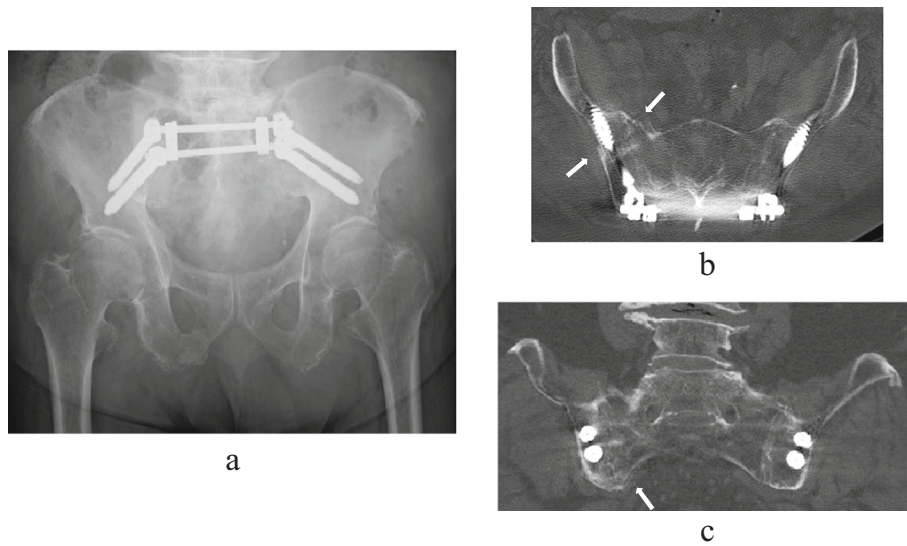


Fig. 4. Plain radiograph 4 months after surgery (a). Computed tomography showed that the bone union was achieved with bony callus (b: axial view, c: coronal view, arrows).

Discussion

In vertebral fractures associated with AS, cervical spine injuries are the most common fractures [3]. By contrast, sacral spine fractures are very rare, and account for 0.3% [3] of all the fractures. In surgery for thoracolumbar vertebral fractures associated with AS, it is important to deal with long lever arms owing to ligament ossification and bone fragility associated with stress shielding. Long fixation, such as three above and three below, and penetrating endplate screws, are reported to be effective ways for the improvement of the strength of fixation [8].

In this case, the sacrum and bilateral ilium were in complete continuity owing to bony ankylosis of the bilateral sacroiliac joints, which was similar to thoracolumbar vertebral fractures, which occurred within the range of ankylosis. In this case, the fracture line passed from the ilium to the sacrum owing to bony ankylosis of the sacroiliac joint. FFP complicated by unilateral sacroiliac joint injury is the Rommens and Hofmann classification criterion IIIb [4]. The fracture in this case was not a sacroiliac joint injury but a fracture at the part at which the iliac and sacral bones were one bony mass owing to the ankylosis of the sacroiliac joint. It was considered an atypical case—a variant of type III that did not fit the Rommens and Hofmann classification criteria. There were no reports of similar fractures in the past, and we struggled with the choice of the surgical technique. Referring to the treatment of thoracolumbar vertebral fractures complicated by AS, we thought that strong fixation to counteract a long lever arm and bone fragility were necessary. Although iliosacral screws and transsacral bar osteosynthesis are commonly used in FFP surgery and are minimally invasive, neither iliosacral screws nor transsacral bar osteosynthesis could be applied in this case because they should have been inserted through the fracture site [4]. Recently, there have been several reports on a technique of complete fixation in the pelvis for sacral fractures in FFP, avoiding spinopelvic fixation. These techniques are suitable for elderly patients, combining the S1 pedicle screw with the S2-alar-iliac screw or iliac screw for strong fixation in the pelvis [9,10]. However, there has been no report of its application to patients with AS. In our case, we were unsure if it could have been applied with one screw on each side against the ilium.

The fracture type in this case was atypical and did not fit the Rommens and Hofmann classification criteria. Thus, we carefully considered the surgical technique used. Considering the pathological condition and the patient's background, including unilateral instability, dislocation of ~6 mm, ossification of the sacroiliac joint due to AS, and osteoporosis, we considered that ILIS proposed by Okazaki was appropriate. In this procedure two iliac screws are inserted bilaterally and the left and right iliac screws were connected with rods [5]. The ILIS was originally designed for Rommens and Hofmann classification criterion IIIa. The ILIS is a minimally invasive technique with closed reduction, and the long iliac screws provide strong fixation by applying compressive forces to the fracture when the affected limb is loaded, thus allowing the application of the full load after surgery, which is suitable for elderly patients.

The advantages of ILIS in this case were the avoidance of screw insertion through the fracture site, the insertion of a long iliac screw, and the expectation of a strong gripping force on the right ilium, as well as the strong fixation of the right and left ilia because the sacrum was also fractured. Although it was not certain whether ILIS—a type IIIa technique—would be effective in this case, we were able to insert a long, thick screw ($\Phi 10$ –100 mm) and able to provide pain relief and early mobilization immediately after surgery.

Postoperatively, there was no screw backout or wound infection, and bony union was achieved 4 months after surgery. The patient did not complain of pain at the final follow-up, and his progress was good.

The present case is a rare report of surgical treatment in a patient with AS and atypical FFP. The simultaneous presence of AS and FFP, with a possible atypical fracture type, is a particularly noteworthy aspect of this report.

To the best of our knowledge, there is no other surgical report of FFP complicated with AS. Sacral fractures are found in 0.3 % of vertebral fractures associated with AS [3], but the incidence and prevalence of FFP in AS are unknown. We spent considerable thought and consideration for the choice of the surgical technique because there were no previous reports on this topic. We chose ILIS and were fortunate that the outcome was acceptable. Similar to the case of thoracolumbar vertebral fractures in patients with AS, we believe that the choice of the surgical technique based on considerations of the long lever arm and bone fragility, was a critical factor in achieving a good outcome.

The progress of this case is still short—approximately only six months—and mid- to long-term follow-up is essential. In addition, it is necessary to study the surgical technique in more cases. We hope that the treatment of this case will help in the treatment of similar cases in the future.

Conclusion

We reported a FFP case in a patient with AS in which early pain relief and bone union were achieved by internal fixation. In this case, ILIS was also useful in patients with atypical fractures that did not fit the Rommens and Hofmann classification criteria owing to the presence of AS, and wherein strong fixation was considered necessary.

Consent

The patient provided consent for the publication and use of the attached pictures.

Declaration of competing interest

The authors declare that there is no conflict of interest regarding the publication of this article.

Data availability

The datasets generated and analyzed during the current study are available from the corresponding author on reasonable request.

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References

- [1] J. Braun, J. Sieper, Ankylosing spondylitis, *Lancet* 369 (2007) 1379–1390.
- [2] J. Zhao, C. Huang, H. Huang, et al., Prevalence of ankylosing spondylitis in a Chinese population: a systematic review and meta-analysis, *Rheumatol. Int.* 6 (2020) 859–872.
- [3] L.A. Westerveld, J.J. Verlaan, F.C. Oner, Spinal fractures in patients with ankylosing spinal disorders: a systematic review of the literature on treatment, neurological status and complications, *Eur. Spine J.* 18 (2009) 145–156.
- [4] P.M. Rommens, A. Hofmann, Comprehensive classification of fragility fractures of the pelvic ring: recommendations for surgical treatment, *Injury* 44 (2013) 1733–1744.
- [5] S. Okazaki, M. Shirahama, R. Hashida, et al., Iliac intramedullary stabilization for type IIIA fragility fractures of the pelvis, *Sci. Rep.* 10 (2020) 20380, <https://doi.org/10.1038/s41598-020-77560-7>.
- [6] S. van der Linden, H.A. Valkenburg, A. Cats, Evaluation of diagnostic criteria for ankylosing spondylitis. A proposal for modification of the New York criteria, *Arthritis Rheum.* 27 (1984) 361–368.
- [7] S.A. Majeed, Grading the outcome of pelvic fractures, *J. Bone Jt. Surg. Br.* 71 (1989) 304–306.
- [8] T. Hishiya, T. Ishikawa, M. Ota, Posterior spinal fixation using penetrating endplate screws in patients with diffuse idiopathic skeletal hyperostosis-related thoracolumbar fractures, *J. Neurosurg. Spine* (2021) 936–941.
- [9] R. Hartensuer, N. Grüneweller, M.F. Lodde, J. Evers, O. Riesenbeck, M. Raschke, The S2-alar-iliac screw for pelvic trauma, *Z. Orthop. Unfall.* 159 (2021) 522–532.
- [10] Y. Yanagisawa, Y. Eda, S. Teruya, H. Gamada, M. Yamazaki, 'Within ring' concept treatment for displaced h-shaped type ivb fragility fractures of the pelvis, *Case Rep. Orthop.* 2021 (15) (2021) 2021, <https://doi.org/10.1155/2021/6864910>. eCollection.