

Large aortic pseudoaneurysm after fusion surgery for hyperextension-type lumbar fracture in diffuse idiopathic skeletal hyperostosis: illustrative case

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BACKGROUND This study aimed to report an aortic pseudoaneurysm, a rare but lethal complication, after a spinal fracture in ankylosing spine.

OBSERVATIONS An 83-year-old obese woman presented with dementia and was nonambulatory after a fall. She was transported to the hospital, and imaging showed a hyperextension-type L1 fracture with diffuse idiopathic skeletal hyperostosis (DISH). After posterior fusion surgery using percutaneous pedicle screws, screw loosening was detected 10 days postoperatively. Fracture dislocation was reduced by changing to transdiscal screws and rodding while in the lateral position. However, the anterior opening persisted. Enhanced computed tomography performed at 6 weeks postoperatively showed a large aortic pseudoaneurysm extending into the vertebral fracture site without screw loosening. Neither endovascular aortic repair nor open surgery was applicable. The patient was transferred to a sanatorium and died of pneumonia 5 months postoperatively without aortic aneurysm rupture.

LESSONS An aortic pseudoaneurysm can occur in hyperextension-type spinal fractures in DISH, even after fusion surgery, when the edge of the fracture is in contact with the aortic wall. The anterior opening dislocation should be reduced as much as possible.

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KEYWORDS ankylosing spine; spinal fracture; hyperextension; aortic pseudoaneurysm; surgery complication

Spinal fractures in patients with ankylosing spinal disorders (ASD), such as ankylosing spondylitis or diffuse idiopathic skeletal hyperostosis (DISH), are often mechanically unstable. The fractures are usually of the severe types, such as AO spine injury type B or C, even if the cause of injury is a low-energy trauma, such as a fall.¹ These fractures may cause immediate- or late-onset spinal cord injury and neurological deficits after the trauma. Hence, operative management is usually recommended for all patients, except in rare cases of stable fractures.^{2,3} Patients with ASD who have fractures and have received both conservative and operative treatment have higher complication and mortality rates⁴ and require intensive management.

One of the most severe complications in the treatment of spinal fractures in ASD is aortic injury. In a systematic review of spinal fracture in patients with ASD, one patient with aortic pseudoaneurysm and four patients with aortic dissection (three of whom died)

were reported.¹ Meanwhile, in a Japanese nationwide multicenter study, spinal fractures of two patients (0.7%) with DISH caused aortic aneurysms.⁵ The vertebral fractures in ASD have a relatively higher frequency of anterior splaying, which may cause aortic wall injury. Therefore, spinal fractures in ASD are associated with an increased risk of aortic pseudoaneurysm development. Herein, we present a patient with ASD who suffered a spinal fracture accompanied by an aortic pseudoaneurysm, which is a rare but lethal complication.

Illustrative Case

An 83-year-old woman had become nonambulatory because of severe back pain after a fall while walking. She arrived at a nearby hospital, where a vertebral fracture of L1 was noted (Fig. 1A and B). Because of the large dislocation of the fracture, she was transferred to our hospital. Her medical history included dementia and osteoporosis. She walked independently at home using a wheeled walker

ABBREVIATIONS ASD = ankylosing spinal disorders; CT = computed tomography; DISH = diffuse idiopathic skeletal hyperostosis.

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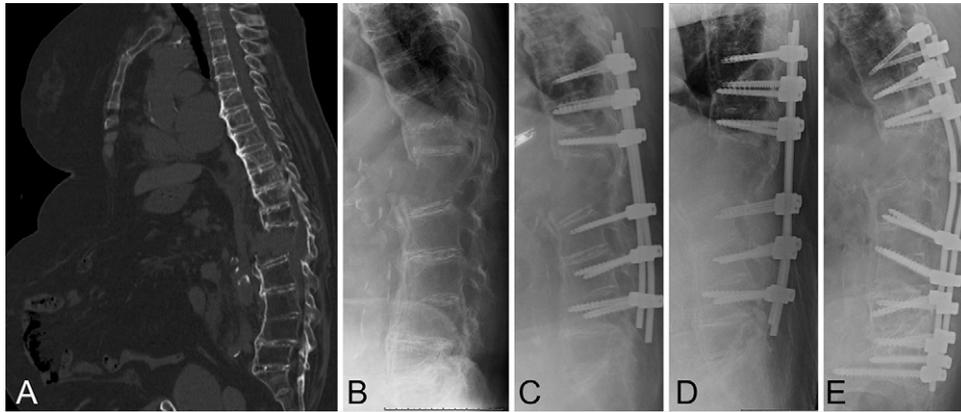


FIG. 1. Reconstructed sagittal CT in the lateral position (A) and lateral radiographs immediately after trauma (B), after the primary surgery (C), after loosening of the caudal pedicle screws (D), and after the second surgery (E).

before the injury. She was lying on her side because of severe back pain in the supine position. She had no sensory disturbance or numbness in the lower extremities. Weakness of the iliopsoas and quadriceps femoris muscles was detected (manual muscle testing 2 level), and it was accompanied by back pain. She weighed 75 kg with a height of 150 cm. Her body mass index was 33.3 kg/m². Radiographs showed an anterior opening-type vertebral fracture and ankylosing spine united by ossified anterior longitudinal ligaments (Fig. 1B). Computed tomography (CT) showed severe anterior opening of the fracture site with an angle of 31° and ankylosis from the T2 vertebra to the sacrum. It also showed a distance of 30 mm between the edges of the anterior wall, whereas the translation of the spinal canal was relatively small (Fig. 1A). The patient was diagnosed with an L1 vertebral fracture classified as AO type B3 accompanied with DISH.⁶ Three days later, she underwent posterior instrumentation surgery using a percutaneous pedicle screw from the T10 to L4 vertebrae. The patient was placed prone on an operative table with the craniocaudal interval of support pads, which were narrowed to reduce fracture site opening. The anterior opening of the fracture site could not be reduced, despite multiple postural changes. Because almost all pedicle screw insertion torques were low and the primary stability of the screws was poor, we abandoned the reduction technique using pedicle screws. Accordingly, the fracture displacement, or anterior opening, remained (Fig. 1C) and was slightly more enlarged than in the initial radiograph.

Although the patient's back pain was relieved to some extent postoperatively, she remained bedridden. If the patient lay supine on the flat floor, her craniocervical part became ungrounded and an extension force would be applied to the fracture site because she had a severe kyphotic ankylosing spine. Consequently, the flat supine position was avoided; instead, a head-up tilt above 20° was maintained. A hard corset could not be applied because of severe obesity. She complained of severe back pain after a CT scan was obtained 8 days postoperatively to check the implant and fracture displacement. Ten days postoperatively, a radiograph revealed loosened pedicle screws on the caudal side (Fig. 1D). It was assumed that the pedicle screws were removed by extension force when she was placed supine for the CT scan. One day later, the patient developed paraplegia, despite not having any trauma. The deterioration was due to a cauda equina injury resulting from implant

loosening and instability of the fracture site. Emergency reoperation was performed on the same day.

First, while the patient was in the prone position, the loosened screws were removed and replaced with transdiscal screws^{7,8} using a navigation system for prolonged fusion to the pelvis. Next, the patient was in the right lateral position and bent forward as much as possible. Afterward, the rods were bent to a contour that was more kyphotic than the natural lateral position. The rods were placed into the screw heads while a surgical assistant out of the operative field attempted to draw the patient's knee to her chest to reduce the anterior opening of the fracture. In addition, the kyphotic angle was increased using in situ benders. The anterior opening of the fracture site was not completely reduced but was narrower than that before the surgery (Fig. 1E). The wound was closed, and the surgery was completed.

The patient experienced no motor recovery of the lower extremities postoperatively. Enhanced CT was performed 1 week following the second operation after ultrasonography detected deep venous thrombosis on both sides of lower extremities. No pulmonary embolism or screw deviation was confirmed. Furthermore, the patient remained undynamic with reduced appetite for more than 1 month after the surgery because of low-activity delirium. Another enhanced CT scan was obtained at 6 weeks postoperatively to identify the cause of melena. CT showed no cause for melena. However, pooling of the contrast agent in the L1 vertebral body was observed, which meant that the aortic pseudoaneurysm extended into the fracture site (Fig. 2A–C). The maximum diameter of the aneurysm was 117 mm in transverse. A review of previous CT showed no aneurysm, but the aortic wall was shown to be in contact with the anterior opening edge of the L1 vertebral body fracture (Fig. 2D). Repeated rubbing of the edge of the fracture site against the aorta since the time of the injury caused an aortic wall injury and development of a large pseudoaneurysm. Because no screw loosening was seen on the latest CT, the fracture edge continued to rub against the aortic wall due to rod deflection after the second surgery. Endovascular aortic repair with stent grafts was not applicable because the location of the aneurysm was close to the bifurcations of major arteries, namely, the superior mesenteric artery. Aortic replacement, which had a high risk of life-threatening complications for elderly patients with dementia and malnutrition, was also not

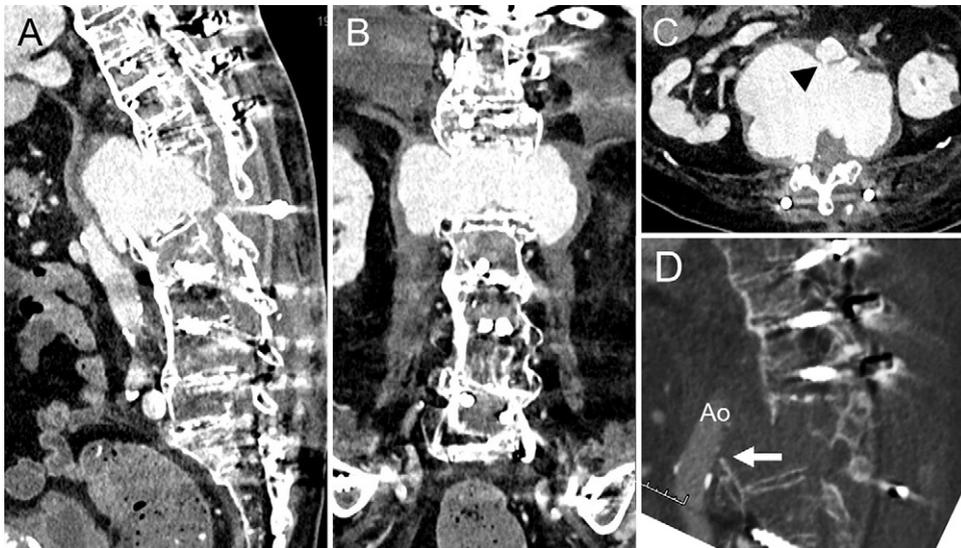


FIG. 2. Aortic pseudoaneurysm and the entry point (*black arrowhead*) on the enhanced CT at 6 weeks postoperatively (**A–C**). The *white arrow* indicates contact between the fracture edge and the aortic wall on the CT at 1 week postoperatively (**D**).

applicable. Therefore, only blood pressure was monitored and an antihypertensive drug was administered. Still bedridden, the patient was then transferred to a sanatorium for long-term care at 8 weeks postoperatively. She gradually experienced wasting from malnutrition due to loss of appetite and died of pneumonia 5 months postoperatively. There was no evidence of aortic aneurysm rupture.

Discussion

Observations

Aortic injury accompanied by spinal fracture is usually caused by high-energy trauma.^{9,10} However, in patients with ASD, aortic injury in association with thoracolumbar fracture can occur after minor trauma, such as a fall.^{11,12} The current patient had an ankylosed spine due to DISH and sustained a lumbar spine fracture from a fall. There was repeated minor traumatic damage to the aortic wall caused by the edge of the hyperextension-type fracture. This resulted in the development of a pseudoaneurysm, even after reduction and fusion surgery. Two previous case reports demonstrated pseudoaneurysm accompanied by a spinal fracture in ASD. There was only one case of a pseudoaneurysm appearing after fusion surgery.^{13,14} In addition, the reported aneurysm was not massive with a diameter larger than that of the vertebral body, such as in our case.

After the second surgery, in which reduction of the dislocation or anterior opening of the fracture was attempted in the lateral position, repeated damage by the fracture edge caused an aortic wall injury without screw loosening. Thus, it is important to anatomically reduce fracture dislocation or anterior opening. However, the reduction of a hyperextension fracture in a patient with ASD is difficult for the following reasons: (1) the anterior opening is usually enlarged in prone position; (2) for reduction of fracture dislocation in an ankylosing spine, thorax and pelvis need to be aligned; and (3) the primary stability of the screws is poor due to osteoporosis and insufficient for fracture reduction. In addition, the large abdomen of

severely obese patients, such as in our case, may interrupt body flexion and fracture reduction. Although adjustment of the alignment is most important in reducing hyperextension-type fracture in ASD, as in our case, the procedure in the lateral position is appropriate because anterior opening was enlarged in our case by initial surgery, in which we made efforts not to open in the prone position. On the other hand, the trajectory of the transdiscal screw is beneficial in increasing the primary anchorage strength of screws.^{7,8}

Despite measures to reduce the displacement of the fracture, if an anterior opening remains and the edge of the fracture site contacts the aortic wall, it is unclear how much displacement is permissible and how additional surgery should be conducted. In our case, the anterior opening angle was approximately 30° and the distance was approximately 30 mm, as measured in the midline sagittal view of the reconstructed CT when the pseudoaneurysm was detected. Fracture dislocation in a patient with a calcified aorta should be reduced further. Posterior shortening osteotomy is a procedure that can correct the alignment and close the gap. This operation, however, is expected to cause massive bleeding, which may result in unstable hemodynamics or coagulopathy, and is associated with a high risk of neurological deterioration. Although anterior strut bone grafting may be performed as an alternative method, it cannot correct the alignment and does not necessarily increase stability as the placement on a fragile bone bed. In addition, considering the risk in approaching a stretched aortic wall, using this method is not recommended. In the future, similar cases should be reported to further discuss appropriate management.

Endovascular aortic repair for a traumatic aortic injury or aneurysm is effective and less invasive,^{15,16} but it is limited to the aneurysm site or its spread. Moreover, performing endovascular aortic repair for an aneurysm that is accompanied by a spinal fracture may be difficult if the aneurysm site is similar to the location seen in our case. On the other hand, open surgery is much more invasive and is limited by the patient's general condition and age. When treating similar fractures in patients with ASD, it is important to

consider the possibility of pseudoaneurysm and perform enhanced CT for its identification. Management procedure to avoid late-onset pseudoaneurysm development is also important, and precautions should be taken during both surgery and postoperative care. An anterior opening dislocation should be reduced in surgery. When the anterior opening remains in patients with an extended ankylosing spine, a mechanical load on the fracture site needs to be minimized. A supine position on a flat floor should be avoided in particular patients with a kyphotic spine because the supine position splay the fracture site, and minor movement of the edge of fracture may injure the aortic wall.

Lessons

An aortic pseudoaneurysm can occur in a hyperextension-type spinal fracture in DISH, even after fusion surgery, when the fracture edge is in contact with the aortic wall. The anterior opening dislocation should be reduced as much as possible in this type of fracture. Further research is required to determine an acceptable displacement range and recommended additional surgical methods.

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Disclosures

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper.

Author Contributions

Conception and design: Shoji. Acquisition of data: Shoji. Analysis and interpretation of data: Shoji. Drafting the article: Shoji. Critically revising the article: Shoji, Sawakami, Ishikawa, Segawa. Reviewed submitted version of manuscript: Shoji, Tanaka. Approved the final version of the manuscript on behalf of all authors: Shoji. Administrative/technical/material support: Wakabayashi. Study supervision: Sawakami, Ishikawa, Segawa.

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