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Clinical Results of Vertebral Fracture Related to Diffuse Idiopathic Skeletal Hyperostosis (DISH) Which Underwent Conservative Treatment: Three Case Reports

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ABSTRACT

Background: A vertebral fracture in a patient with diffuse idiopathic skeletal hyperostosis (DISH) is unstable due to larger moment via the long lever arm of an ankylosed spine. Therefore, surgical treatment is commonly recommended to avoid complications of nonunion and paralysis. In this report, we present 3 cases of vertebral fractures related to DISH which were primarily forced to undertake conservative treatment because of medical comorbidities and advanced age.

Case 1: A 93-year-old woman suffered from T10 vertebral fracture by a ground-level fall on her back. A trunk cast for 6 weeks was followed by brace wear for 3 months with administration of daily teriparatide. Then complete bone union was confirmed at 2 years after injury without back pain.

Case 2: An 84-year-old man suffered from T12 vertebral fracture by a fall on his back from a chair. A trunk cast for 12 weeks was followed by brace wear for 6 months with administration of daily teriparatide. Then acceptable bone union was confirmed at 1 year after the injury, and activities of daily living became independent.

Case 3: An 87-year-old woman suffered from T10 vertebral fracture due to a ground-level fall on her back when doing pruning work. Conservative treatment by trunk cast was first initiated with administration of daily teriparatide. However, delayed paralysis developed at 2 weeks after casting, so minimally invasive spinal stabilization (MISt) was performed. Bone union was obtained at 1 year after the injury without any neurological impairment.

Conclusions: Favorable clinical courses have been obtained in 2 cases, whereas MISt was required for delayed paralysis in 1 case. Although surgical stabilization is the first-line treatment for vertebral fracture with DISH, conservative treatment can also be one of the options in cases with high operative risk due to serious medical comorbidities. However, during conservative treatment, cautious observation is necessary not to overlook the occurrence of paralysis.

Level of Evidence: 4.

Clinical Relevance: Conservative treatment for vertebral fracture with DISH can be one of the options in cases with high operative risk due to serious medical comorbidities.

Other & Special Categories

Keywords: diffuse idiopathic skeletal hyperostosis (DISH), vertebral fracture, conservative treatment, trunk cast, minimally invasive spinal stabilization (MISt)

INTRODUCTION

Diffuse idiopathic skeletal hyperostosis (DISH), which was first proposed by Resnick et al[1] in 1975, is a systematic condition leading to ossification of ligaments and entheses. The criteria of DISH defined by Resnick and Niwayama[2] include the presence of flowing ligamentous calcification and ossification along the anterolateral aspects of at least 4 contiguous vertebral bodies, a relative preservation of intervertebral disc height without degenerative disc disease in the involved areas, and the absence of apophyseal joint bony ankylosis and sacroiliac joint erosion, sclerosis, or bony fusion. Vertebral fractures accompanying DISH lead to reverse chance fractures that can likely bring on spinal paralyses and are frequently resistant to conservative treatment.[3] In this report, we present 3 cases of vertebral fractures related to DISH in which patients were primarily forced to undertake conservative treatment because of medical comorbidities and advanced age (Table).

Case 1

A 93-year-old woman experienced severe back pain after a ground-level fall on her back. She visited a neighborhood orthopaedic clinic, and no

Table. Summary of cases.

Case	Age	Gender	Facture	Comorbidities	Cast	Brace	Surgery	Locomotion (Initial/Final)
1	93	Female	T10	HT	6W	3M	NA	Single crutch/single crutch
2	84	Male	T12	HT, DM, BC, PC	12W	9M	NA	Caster walker/caster walker
3	87	Female	T10	HT	1W	NA	Postinstr and fusion	Caster walker/single crutch

Abbreviations: BC, bladder cancer; DM, diabetes mellitus; HT, hypertension; M, month; NA, not applicable; PC, prostatic cancer, W, week.

bone injury was indicated on x-ray radiographs. However, her severe back pain continued, and she visited our orthopaedic department at 4 days after injury. At that time, she could not walk by herself due to back pain, but no neurological deficit was detected. On x-ray radiographs, ossification of the anterior longitudinal ligament (OALL) of the whole spine with thoracic hyperkyphosis was recognized, but no findings of vertebral collapse were confirmed. Computed tomography (CT) images demonstrated discontinuation of OALL and distension of the posterior wall of the vertebral body at T10 level. Magnetic resonance imaging (MRI) also identified high-intensity changes of the T10 vertebral body and the posterior elements that implied extensive fresh vertebral fracture (Figure 1). As a result of these imaging findings, T10 vertebral fracture related to DISH was diagnosed; however, conservative treatment was selected due to extremely advanced age. A trunk cast for 6 weeks was followed by brace wear for 3 months with administration of daily teriparatide. An ambulation exercise was initiated at 1 month after injury, and the brace was weaned at 4 months after injury. At 2 years after injury, complete bone union and vanishment of the cavity of the T10 vertebral body that remained at 1 year after injury were confirmed on CT images (Figure 2).

Case 2

An 84-year-old man had difficulty with ambulation due to severe back pain after a fall on his back from a chair. He was raced to the emergency hospital and diagnosed with an usual T12 osteoporotic vertebral fracture on x-ray radiographs. First, brace treatment was initiated, but his severe back pain was not relieved, and he visited our orthopaedic department at 2 weeks after injury. At the first visit, he could not rise from his bed due to back pain, even though no neurological deficit was verified. On x-ray radiographs, OALL of the thoracolumbar spine was recognized, but neither

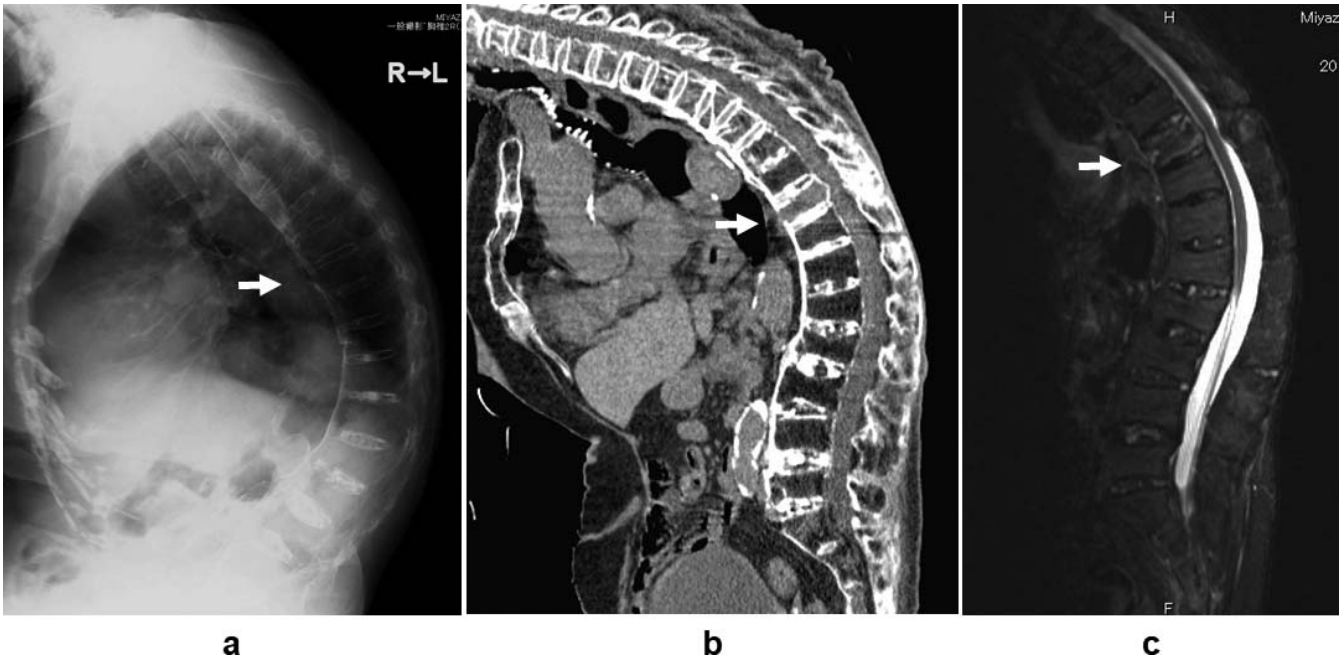

Figure 1. Case 1: Pretherapeutic x-ray, computed tomography (CT), magnetic resonance imaging (MRI). (a) X-ray lateral view. No obvious vertebral collapse was observed; although, thoracic hyperkyphosis with the ossification of anterior ligament of the whole spine was identified. (b) CT sagittal view. Ossification of anterior longitudinal ligament was discontinued, and the posterior wall of the vertebral body was distended at the T10 vertebral level. (c) MRI STIR (short T1 inversion recovery) sagittal view. High-intensity changes of the T10 vertebral body and the posterior elements were confirmed.

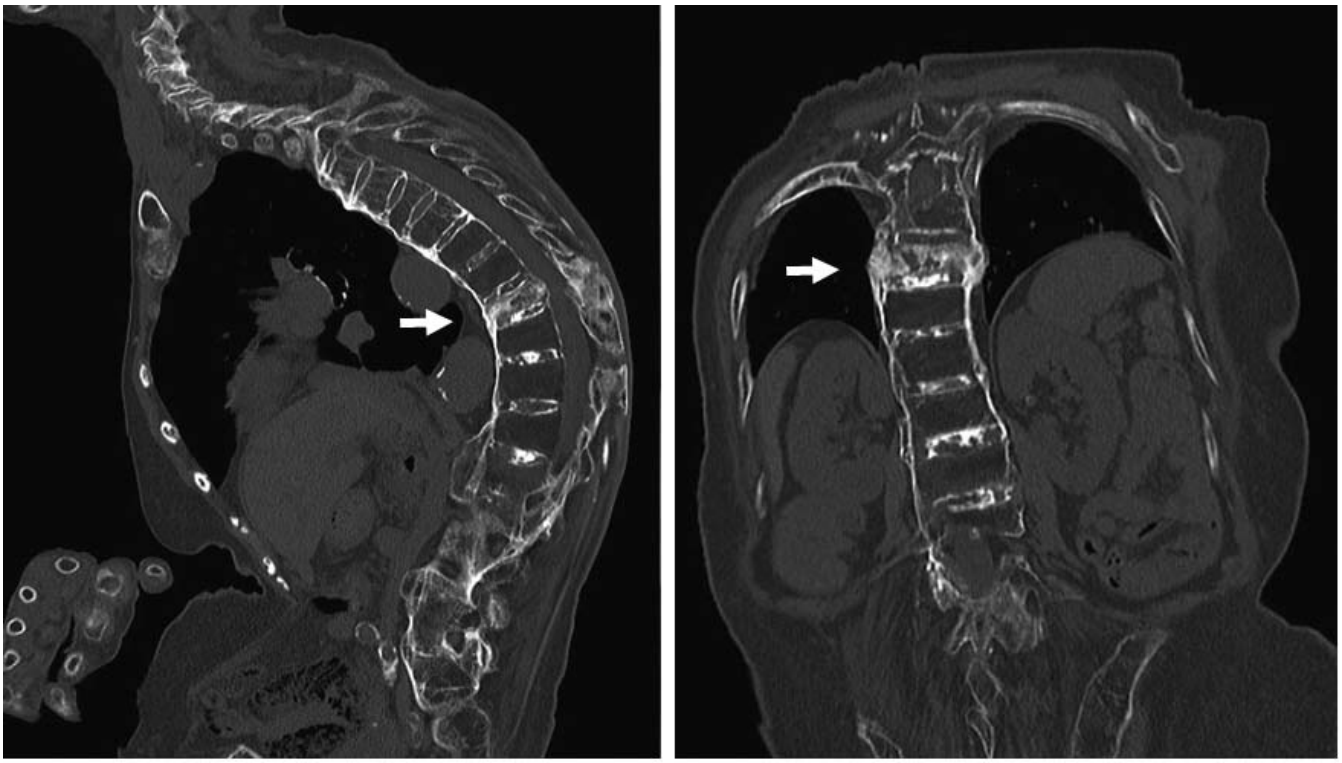

Figure 2. Case 1: Computed tomography (CT) at 2 years, 1 month after injury. (a) Sagittal view. Bone union of both the T10 vertebral body and the spinous process was achieved. (b) Coronal view. Callus formation was recognized at not only the central part but also the lateral sides of the T10 vertebral body.

findings of vertebral collapse nor kyphotic deformity were confirmed. CT images demonstrated discontinuation of OALL and distension of the posterior wall of the vertebral body at the T12 level. MRI indicated high-intensity changes of the T12 vertebral body and the posterior elements that implied extensive fresh vertebral fracture (Figure 3). As a result of these imaging findings, T12 vertebral fracture related to DISH was determined, and a trunk cast was applied accompanied with administration of daily teriparatide because of advanced age and cardiovascular comorbidity. Six weeks after casting, back pain persisted; therefore, another 6 weeks of external fixation by trunk cast was carried out. At 3 months after injury, cane walk was made possible, and activities of daily living became independent. Although the cavity of the T12 vertebral body remained at 5 months after injury, bone augmentation and size reduction were confirmed on CT images at 1 year after injury. At this point, brace removal was permitted (Figure 4).

Case 3

An 87-year-old woman realized severe back pain due to a ground-level fall on her back when doing pruning work. She visited our orthopaedic department at 5 days after injury because her back pain was not alleviated. At the first visit, she could walk with a walker, and no neurological deficit was observed. On x-ray radiographs, multiple-vertebral collapse with thoracic hyperkyphosis was recognized. CT images revealed angular displacement of fracture and expansion of the cavity of the T10 vertebral body. MRI indicated high-intensity changes of the T10 vertebral body and the posterior elements that implied extensive fresh vertebral fracture (Figure 5). First, we diagnosed a usual T10 osteoporotic vertebral fracture, and brace treatment was initiated. However, her back pain deteriorated. CT images demonstrated discontinuation of OALL and expansion of the vertebral body at T10 level. At this point, T10 vertebral fracture related to DISH was diagnosed; however, conservative treatment was continued due to refusal of surgical treatment because of advanced age. Then a trunk cast was applied with administration of daily teriparatide, although incomplete paraplegia occurred at 1 week after casting. On MRI, mild compression and meandering of the spinal cord due to spinal canal stenosis created by advanced angular

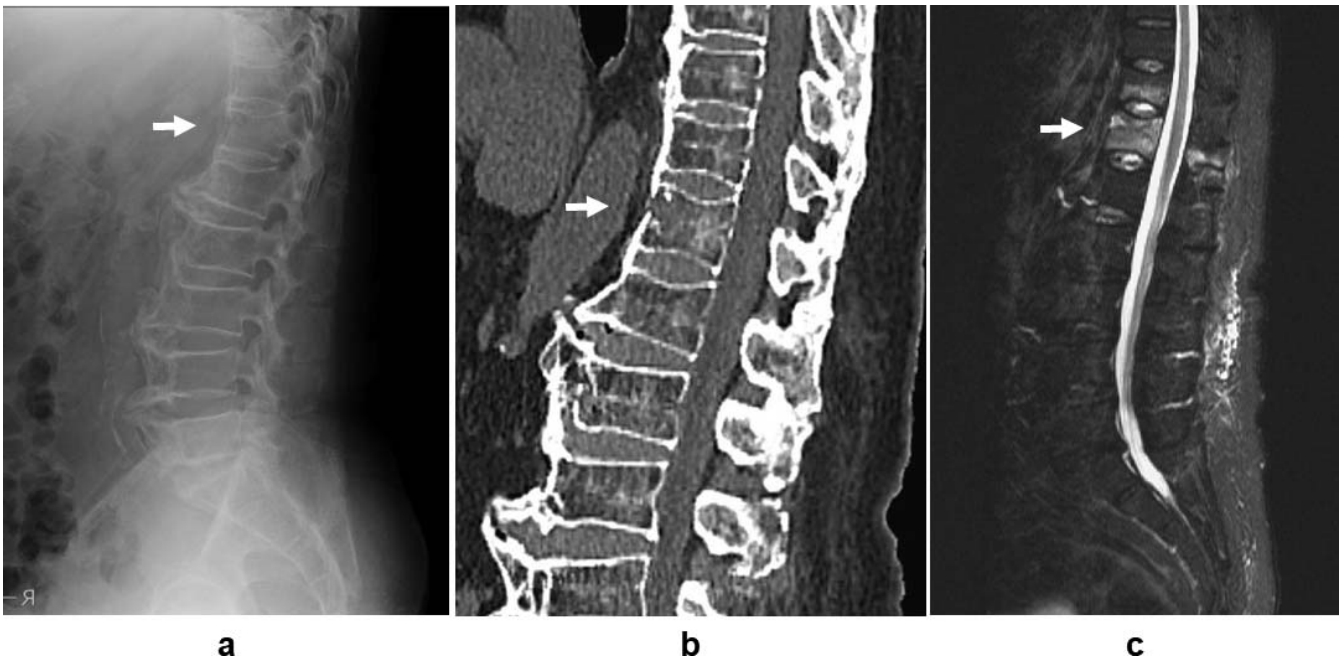

Figure 3. Case 2: Pretherapeutic x-ray, computed tomography (CT), magnetic resonance imaging (MRI). (a) X-ray lateral view. Massive osteophyte formation of the anterior portion of the spine was observed; although, neither abnormality of spinal alignment nor collapse of vertebral bodies existed. (b) CT sagittal view. Ossification of anterior longitudinal ligament was discontinued at the thoracolumbar level. (c) MRI STIR (short T1 inversion recovery) sagittal view. High-intensity changes of the T12 vertebral body and the posterior elements were confirmed.

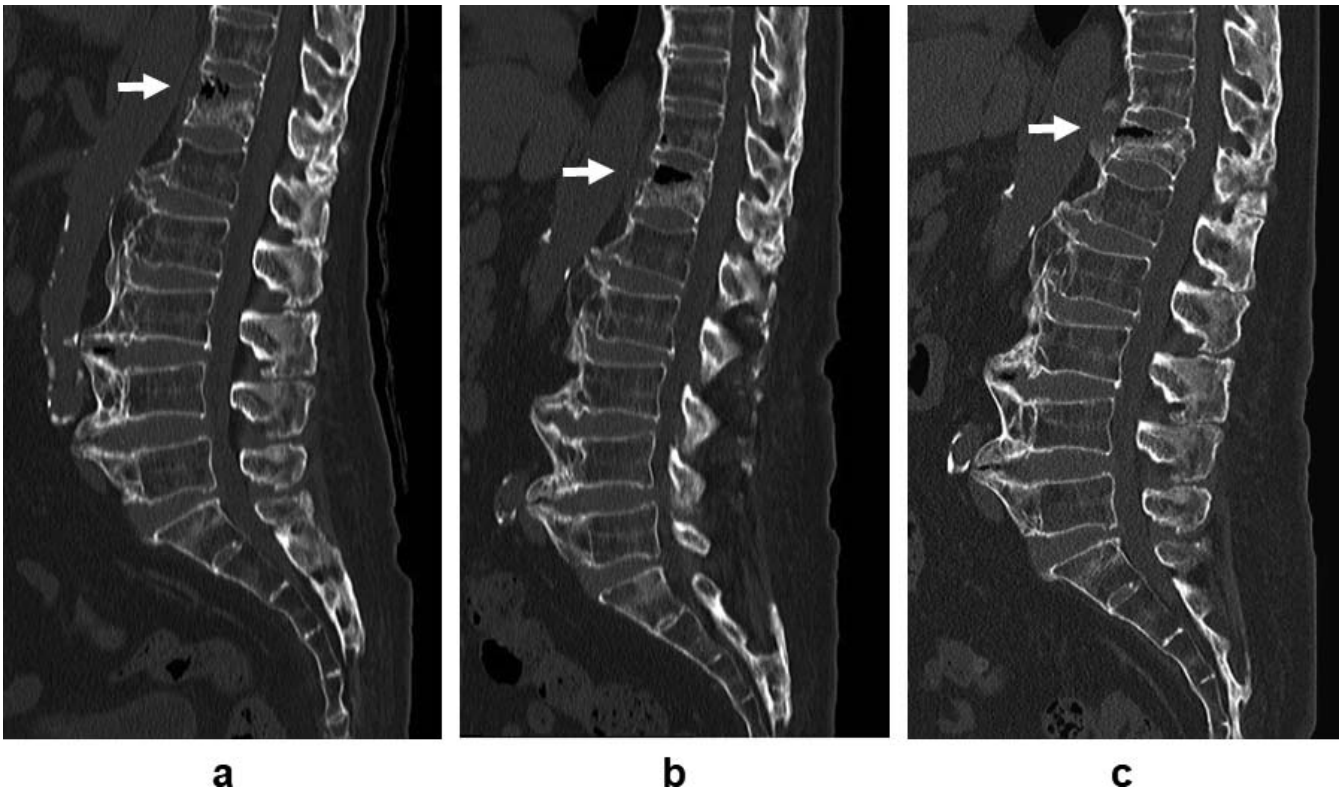

Figure 4. Case 2: Sequential changes of computed tomography (CT). (a) 3 months after injury. The cavity in the anterior part of the T12 vertebral body was recognized. (b) 5 months after injury. The cavity in the anterior part of the T12 vertebral body was enlarged. (c) 1 year after injury. In the T12 vertebral body, collapse has progressed, but the cavity was diminished in size.

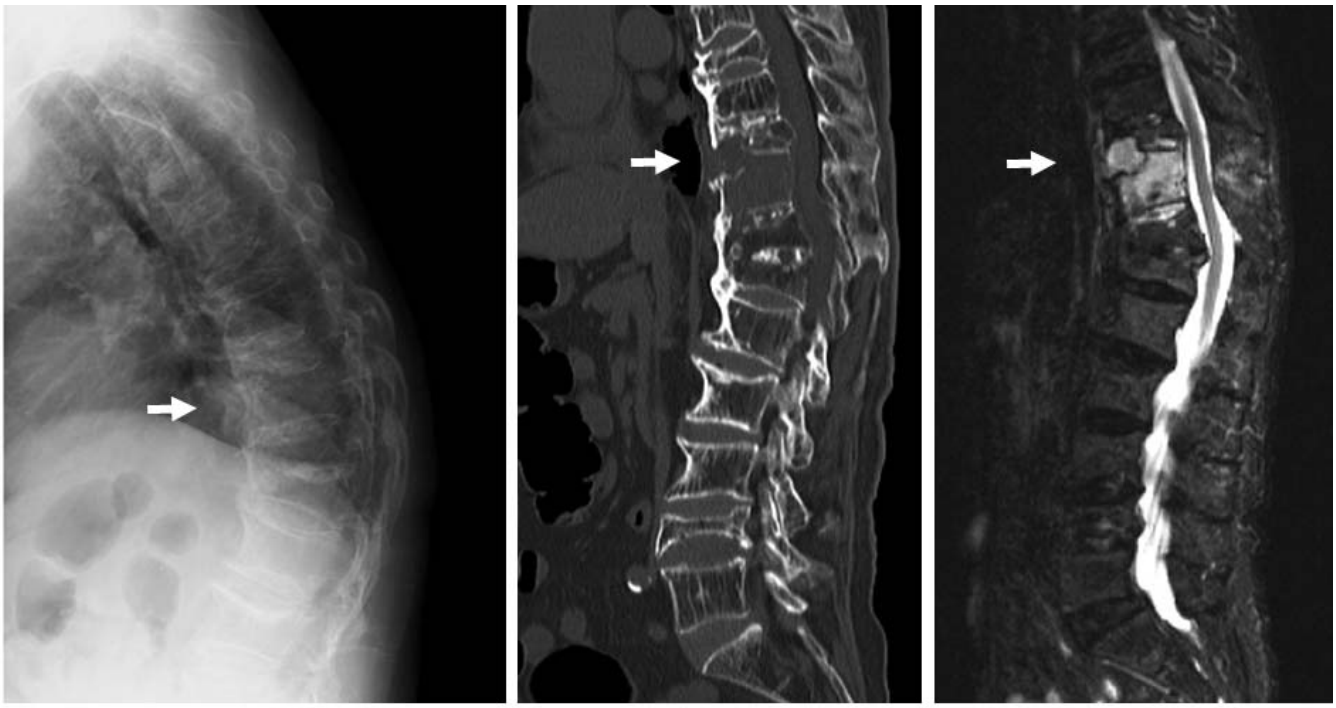

Figure 5. Case 3: Pretherapeutic x-ray, computed tomography (CT), magnetic resonance imaging (MRI). (a) X-ray lateral view. Multiple-vertebral collapse with thoracic hyperkyphosis was recognized. (b) CT sagittal view. Ossification of anterior longitudinal ligament was discontinued at the T10 vertebral level, and the spinous processes of T9, 10, and T11 were spontaneously fused. (c) MRI STIR (short T1 inversion recovery) sagittal view. High-intensity changes of the T10 vertebral body and the posterior elements were confirmed.

displacement of the fracture were identified (Figure 6). Therefore, minimally invasive spinal stabilization (MISt) was performed. At 2 weeks after surgery, her paralysis improved, and ambulation exercise was initiated. Although no major postoperative complication was encountered, delayed superficial wound infection occurred at 1 month after surgery, and prolonged wound treatment was necessitated. Finally, the sagittal spinal alignment was restored to normal, and bone union was confirmed on x-ray radiographs and CT images at 1 year, 4 months after injury (Figure 7). At this point, she could walk with a single crutch in the absence of back pain and any neurological deficit.

DISCUSSION

DISH patients are unequally distributed between males and females, and DISH prevalence rapidly increases with age.[4,5] The ankylosed spine is prone to vertebral fracture from trivial trauma, such as a fall from a standing or sitting position or a low-speed motor vehicle collision, because of decreased bone mineral density due to altered biomechanical loading of vertebral bodies.[6] Moreover, since hyperextension is the most frequent

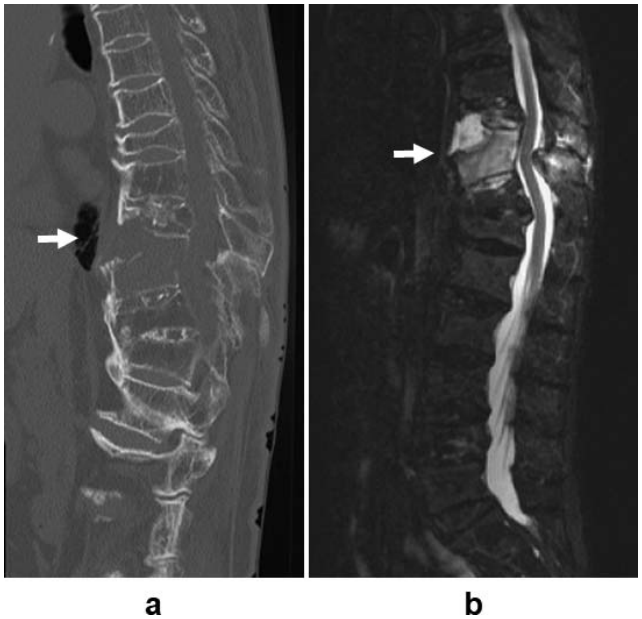

Figure 6. Case 3: Computed tomography (CT) and magnetic resonance imaging (MRI) at the advent of paralysis. (a) CT sagittal view. Angular displacement of fracture has been progressed, and the cavity of the T10 vertebral body was spread. (b) MRI STIR (short T1 inversion recovery) sagittal view. Mild compression and meandering of the spinal cord due to spinal canal stenosis created by advanced angular displacement of fracture was identified.

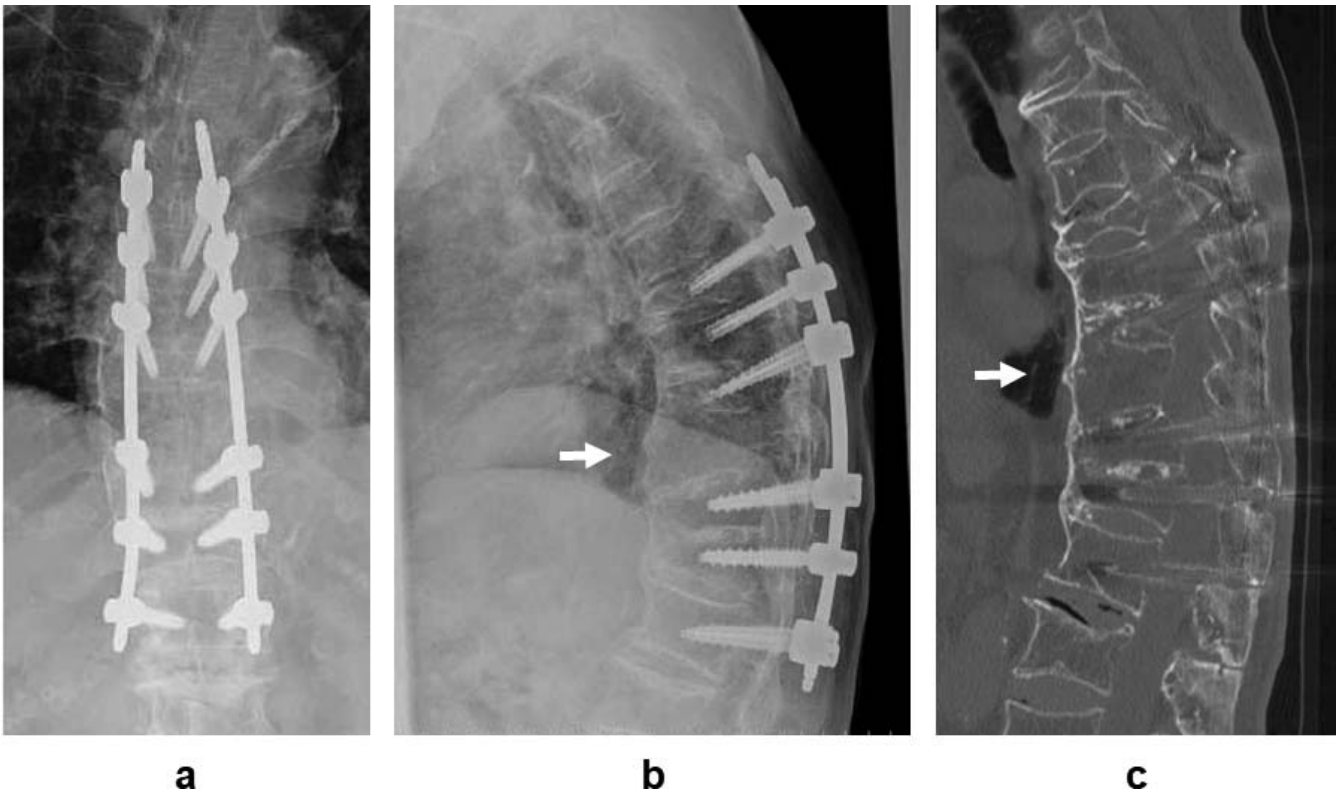

Figure 7. Case 3: X-ray and computed tomography (CT) at 1 year, 4 months after surgery. (a) X-ray anterior-posterior view. (b) X-ray lateral view. (c) CT sagittal view. Bone union of the T10 vertebral body with proper sagittal spinal alignment was confirmed.

cause of vertebral fracture, a delay in diagnosis often occurs on account of absence of obvious collapse of the vertebral body.[7] Caron et al[8] reported that almost one-fifth of patients with ankylosing spinal disorders (DISH and ankylosing spondylitis) had a delay in diagnosis of their spine fracture, and they were associated with an 81% likelihood of decline in neurologic function. Vertebral fractures on the immobile fused spine may eventually result in 3-column injuries that are extremely unstable just like a long-bone fracture, and late onset paralyses are probably happened along with the progression of displacement.[7] For these reasons, when elderly patients suspected with vertebral trauma are encountered, mechanism of injuries must be foremost confirmed. If the spinal trauma is suspected in DISH patients, proactive exploration via CT or MRI should be conducted to boost the chance of early detection of vertebral fracture, even though obvious findings of bone injury are undetectable on x-ray radiographs. Further, when a fresh vertebral fracture has been diagnosed, careful posture support by strict internal or external fixation should be implemented before deterioration of displacement to avoid complication of nonunion, spinal deformity, spinal paralysis, and death.[9] Presently, MISt seems to be the first choice of treatment for vertebral fracture with DISH. However, conservative treatment is occasionally necessary to choose in cases with high operative risk due to serious medical comorbidities or advanced age.

There has been much controversy in this field, and no definitive treatment strategy has emerged for treating spinal fractures in patients with DISH. Tanishima et al[10] recommend posterior fusion with vertebroplasty just after injury for vertebral fracture in elderly patients of advanced age with DISH to extend their healthy lifespan. In contrast, Whang et al[11] recommended conservative treatment with immobilization for stable fractures that were not associated with neurologic deficits.

In these situations, several reports have recently suggested that teriparatide would exhibit efficacy to promote healing of bone fracture and pseudoarthrosis by aggressively stimulating osteoblasts to promote bone formation.[12–15] Transcending fresh fractures, the effective treatment of delayed union of a lumbar spine fracture with daily administration of

teriparatide in patients with DISH was also reported.[16] Teriparatide is a recombinant bioactive fragment (1-34) of human parathyroid hormone and is indicated for the treatment of postmenopausal osteoporosis. Also, it has been proven efficacious for acceleration of fracture healing in animals.[17] Iida et al[18] stated that teriparatide could be a medicine that will fundamentally change the treatment method concept for vertebral fractures related to DISH.

The vertebral fractures in all 3 cases in the current series have also been caused by minor trauma. In 2 cases, bone union was obtained with conservative treatment of a trunk cast followed by brace wear with daily teriparatide administration. However, 1 case needed surgical treatment because of delayed incomplete paraplegia due to displacement of fracture despite strict immobilization with a trunk cast. Fortunately, no major postoperative complication was developed, except delayed superficial wound infection that occurred at 1 month after surgery.

Along with the advent of a society aging more than ever in Japan, an increase in elderly vertebral fracture patients with DISH is expected in the future. When elderly patients suspected to have vertebral trauma are encountered, screening of the entire spinal column with CT or MRI is recommended. Although surgical stabilization is the first-line treatment for vertebral fracture in the DISH patient, conservative treatment can also be one of the options in such cases with high operative risk due to serious medical comorbidities or extreme advanced age. When conservative treatment is implemented, strict external fixation by a rigid trunk cast under careful observation and the patient's understanding of the uncommon refractory vertebral fracture are imperative to achieve bone union. However, a switch of the management plan to operative treatment should not be feared in the event of an outbreak of neurological symptoms after confirming the full cooperation of anesthesiologists and the explicit consent of the patients and their families.

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