

Diffuse Idiopathic Skeletal Hyperostosis with Severely Displaced Spine Fracture Managed with Posterior Approach Alone: Case Reports and Literature Review

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Abstract

Diffuse idiopathic skeletal hyperostosis (DISH) is a condition in which minor trauma can cause extremely unstable vertebral fractures. Spinal fractures associated with DISH are prone to instability due to the large moment of lever arm and secondary neurological deterioration; hence, surgical internal fixation is considered necessary. On the other hand, some reports suggest that patients with DISH have a high osteogenic potential. In this report, we describe three patients with DISH. These patients had spinal injuries that resulted in a large gap, for which anterior fixation with bone graft would generally be considered due to comminuted fractures. However, we achieved good bony fusion with posterior fixation alone, without forcible correction.

Keywords: diffuse idiopathic skeletal hyperostosis, spinal injuries, vertebral fractures, osteogenic potential

Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) is a non-inflammatory systemic condition characterized by progressive calcification and ossification of ligaments and entheses.^{1,2)} To date, the etiology and pathogenesis of DISH remain poorly understood; however, several authors have described associations with advancing age, hypertension, obesity, and type 2 diabetes mellitus.²⁾ The spinal column of patients with DISH tends to fracture in a pattern similar to that observed in a long bone. Furthermore, patients with DISH are at a greater risk of developing unstable spine fractures due to a stiff ankylosing spine with long lever arms.³⁾ Surgical treatment is generally recommended based on the incidence of neurological complications and in case of poor prognosis of fractures. Although various surgical approaches have been reported for treating fractures in patients with DISH, including anterior, posterior, and combined approaches,⁴⁾ additional anterior fixation

procedures are generally necessary for vertebral fractures with a large bony gap.

In this report, we present patients with DISH who had severely dislocated spine fractures. Therein, bony fusions were achieved with posterior fixation alone.

This study was approved by the local Institutional Review Board of Iseikai Hospital (No. 2022-7), and informed consent was obtained from the patients for the use of their data.

Case Report

Case 1

An 83-year-old female fell from her bed and sustained a dislocation fracture at the C6 vertebral body corresponding to type C in the AO classification and compression fracture at the T7 vertebral body. She underwent laminoplasty from the C3 to the T1 levels for cervical ossification of the posterior longitudinal ligament (OPLL) at another institute

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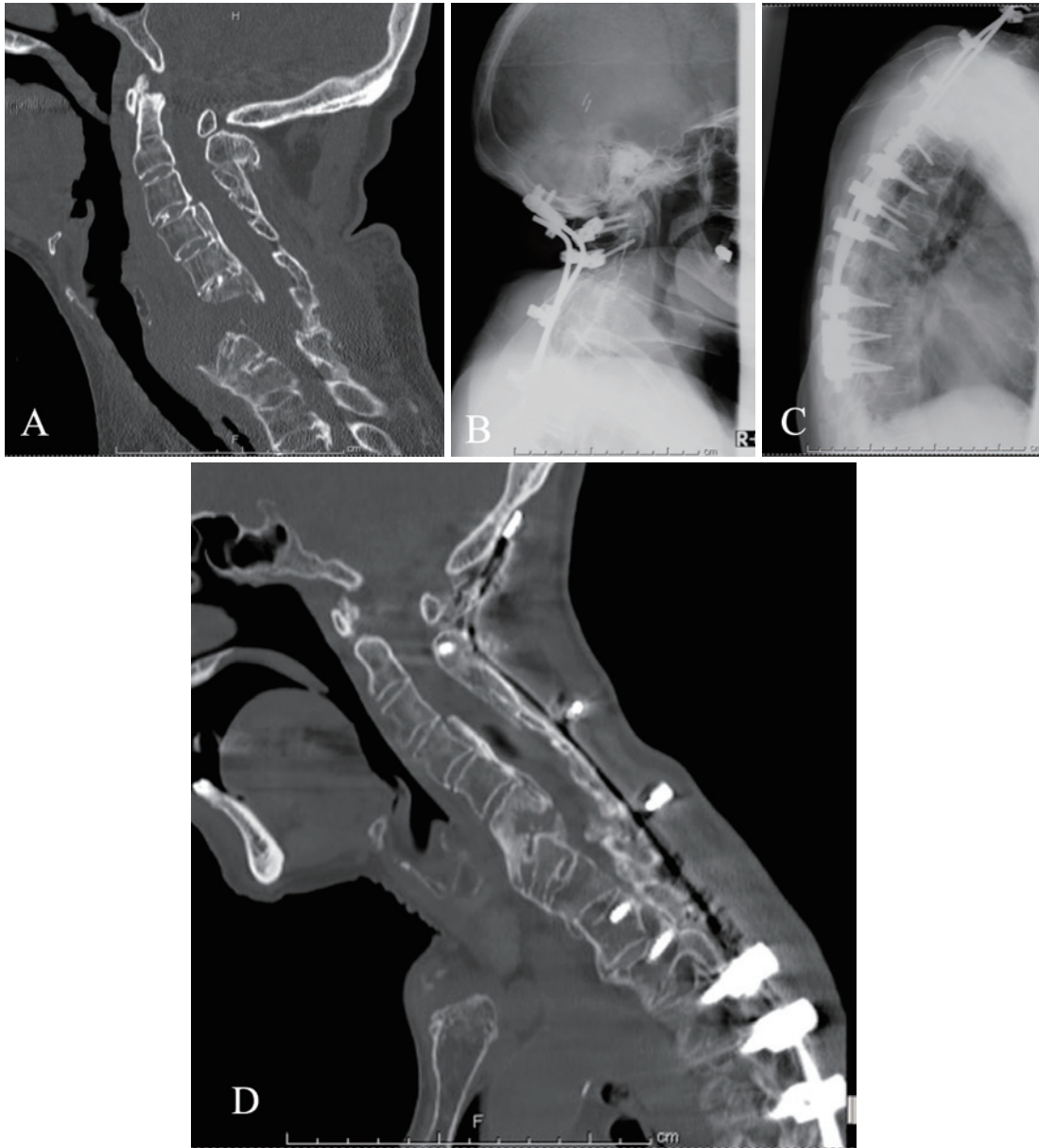


Fig. 1 Pre- and postoperative radiological studies of case 1.

Preoperative computed tomography (CT) reveals a markedly displaced vertebral body fracture at the C6 level by hyperextension injury (A). Postoperative cervical (B) and thoracic (C) X-rays reveal the operative construct of O-Th10 posterior lateral fixation with occipital bone plate and pedicle screws. The follow-up CT at 1 year after the operation (D) showed bony fusion between the fractured ends at the C6 level.

more than 15 years earlier. Computed tomography (CT) revealed a markedly displaced vertebral body fracture at the C6 level, presumably due to hyperextension injury (Fig. 1 A). The patient underwent external fixation using a halo vest as the primary treatment and received *in situ* posterior fixation from the occiput to the T10 level using occipital bone plate and pedicle screws 1 week later (Fig. 1B, C). She began rehabilitation on the 3rd day after surgery and became ambulant with light support at the 1-month mark. The follow-up CT at the 1-year mark revealed bony fusion

bridging the dislocated vertebral fracture (Fig. 1D).

Case 2

A 59-year-old female had been suffering from severe pain in the right hip and thigh after tumbling down on the floor. She underwent posterior decompression and posterolateral fusion from the C2 to the T12 levels for OPLL and compression fracture of the Th2 vertebral body 2 years before. CT revealed a dislocation fracture at the L1 vertebral body (Fig. 2A, B). Posterior lateral fusion was performed

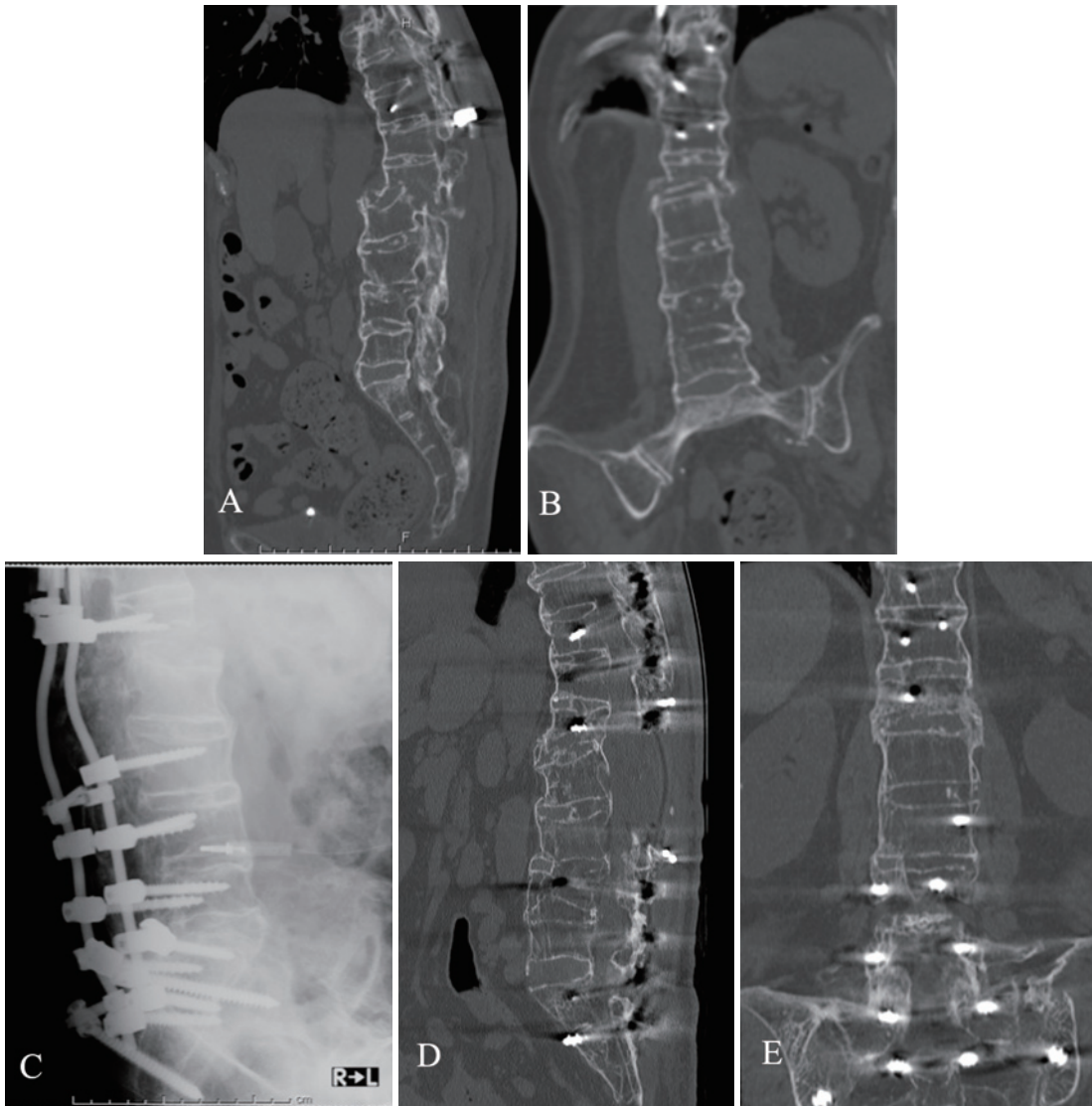


Fig. 2 Pre- and postoperative radiological studies of case 2.

Preoperative computed tomography (CT) reveals chance fracture at the L1 level with severe translation (A, B). Postoperative lumbar X-ray (C) reveals the operative construct of the caudal part of posterior fixation with pedicle screws and S2 alar iliac screws demonstrating slight reduction of the fracture. The follow-up CT at 1 year postoperatively reveals bony fusion between the fractured ends (D, E).

from the T12 to the S2 levels using pedicle screws and S2 alar iliac screws (Fig. 2C). She was relieved from pain in the right leg postoperatively. The follow-up CT after the operation revealed bone synthesis between the fractured ends along the anterior longitudinal ligament at 6 months and almost complete bony fusion at 1 year postoperatively (Fig. 2D, E).

Case 3

A 78-year-old male had a dislocation fracture at the C7-T1 levels after falling down the stairs (Fig. 3A). He had no neurological deficit upon arrival. After external fixation using halo vest upon admission, posterior lateral fixation from the C5 to the Th2 levels was performed (Fig. 3B). The

follow-up CT at the 1-year mark revealed solid bony fusion between severely dislocated vertebral bodies (Fig. 3C).

Discussion

DISH patients are prone to fracture after minor trauma due to its changed biomechanical properties.⁵⁾ Several studies have demonstrated that patients with fractures of the ankylosing spine have a two- to five-fold higher risk of neurological deficits on hospital admission than those without ankylosing spine, often due to a failure to recognize the presence of a fracture.⁶⁾ Secondary worsening of neurological status may also occur due to inadequate immobilization, unwarranted transfers, or imprudent spinal

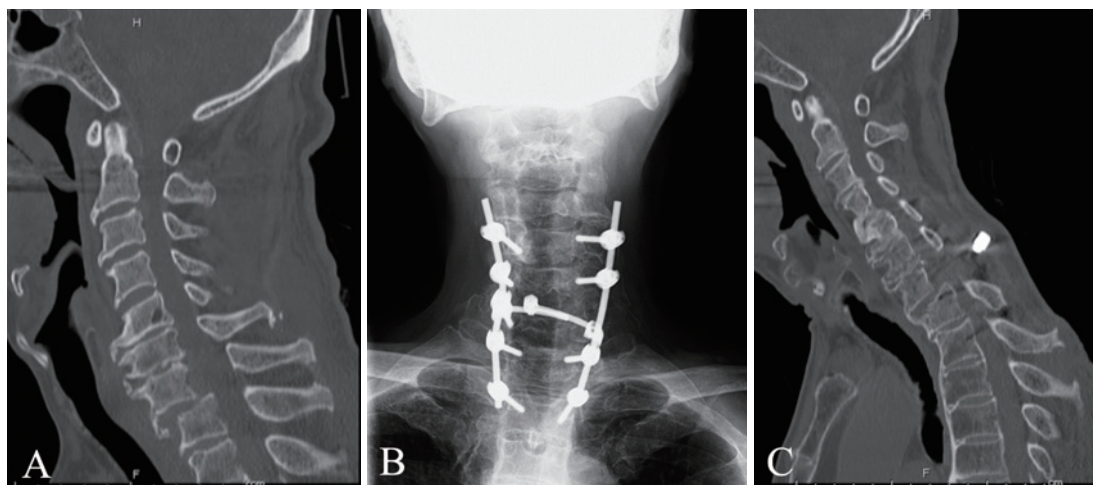


Fig. 3 Pre- and postoperative radiological studies of case 3.

Preoperative computed tomography reveals unstable C7 transverse fracture with listhesis (A). Postoperative cervical A-P X-ray reveals the fusion levels (B). The follow-up CT at 1 year postoperatively reveals bony fusion between the fractured ends along the anterior longitudinal ligament (C).

manipulation.⁷⁾ The fracture site in DISH is subjected to a large bending moment by the lever arm, and it may be more unstable than dynamic radiological studies suggest.⁸⁾ The presence of OPLL and delayed diagnosis are important factors that contribute to the development of neurological deterioration.⁹⁾ Based on these past reports, surgical treatment is generally recommended for unstable spinal fracture in cases of DISH.

The most popular surgical method for vertebral fractures of DISH cases is posterior surgery with instrumentation. Posterior surgeries are expected to recover the pre-existing spinal alignment, stabilize the injured spinal segments, and complete direct neural decompression if necessary.^{4,10)} Many reports recommended posterior fixation using instrumentation between at least the three-above and the three-below vertebral levels.^{3,11)} Alternatively, an adjunctive anterior procedure may be necessary when there is significant comminution of the vertebral column, as with compression burst fractures or distraction extension injuries where there is a characteristic “fish-mouth” defect—and also in situations where the majority of the compressive pathology is located anteriorly.⁴⁾ Although Dhall¹²⁾ pointed out that in patients with ankylosing spine with a Thoracolumbar Injury Classification System score of four or more, a posterior percutaneous screw-rod fixation and fracture reduction may be considered, the study did not include cases with significant bony gap like in our present study.

In the three cases that we reported here, the dislocations of the fracture sites were such that a total circumferential approach would generally be considered necessary. Nevertheless, bony fusion was achieved after 6 months or 1 year with posterior fixation alone in all the three cases.

In terms of the biology of DISH, there is some evidence

that the capacity for osteosynthesis is vigorous in patients with DISH as follows. It has been reported that the newly formed immature bone at the sites of bridging ossification is more prevalent in human cadaveric spines with DISH than those without DISH.¹³⁾ Recently, Kanematsu et al. revealed that new bones formed at the vertebral fracture site of a patient with DISH during anterior procedure 1 month after the initial injury. They pathologically confirmed significant remodeling in a case of thoracolumbar fracture that was performed with additional anterior bone grafting after posterior fixation.¹⁴⁾ Considering these findings and the results of our cases, anterior column injury of the DISH patients, including those with remarkable dislocation, might be treatable with posterior fixation alone.

Due to the poor bone quality inherent in DISH, we did not perform intraoperative corrective manipulation using pedicle screws, but only *in situ* fixation in the prone posture on a surgical frame, because excessive correction for dislocation might lead to instrumentation failure. If posterior fixation alone can be used to treat fractures with significant dislocations as aforementioned, newer surgical techniques such as transdiscal screwing or penetrating endplate screws,^{15,16)} which have been recently reported, are promising for the treatment of these cases. Although Sedney et al. reported that the tendency of autofusion in ankylosing patients lends a theoretical advantage to minimally invasive surgeries with osteosynthesis (rather than arthrodesis), which are essentially internal fixation techniques,¹⁷⁾ further studies are necessary to determine whether satisfactory bony fusion can be achieved with percutaneous instrumentation alone.

Although the need for additional anterior fixation for spinal fractures in DISH patients with significant dislocation is controversial, this report suggests that posterior

fixation alone is sufficient for such cases owing to its tendency for osteosynthesis.

Conflicts of Interest Disclosure

Authors have no conflict of interest in this research.

References

- 1) Belanger TA, Rowe DE: Diffuse idiopathic skeletal hyperostosis: musculoskeletal manifestations. *J Am Acad Orthop Surg* 9: 258-267, 2001
- 2) Mader R, Verlaan JJ, Buskila D: Diffuse idiopathic skeletal hyperostosis: clinical features and pathogenic mechanisms. *Nat Rev Rheumatol* 9: 741-750, 2013
- 3) Caron T, Bransford R, Nguyen Q, Agel J, Chapman J, Bellabarba C: Spine fractures in patients with ankylosing spinal disorders. *Spine (Phila Pa 1976)* 35: E458-E464, 2010
- 4) Whang PG, Goldberg G, Lawrence JP, et al.: The management of spinal injuries in patients with ankylosing spondylitis or diffuse idiopathic skeletal hyperostosis: a comparison of treatment methods and clinical outcomes. *J Spinal Disord Tech* 22: 77-85, 2009
- 5) Callahan EP, Aguilera H: Complications following minor trauma in a patient with diffuse idiopathic skeletal hyperostosis. *Ann Emerg Med* 22: 1067-1070, 1993
- 6) Westerveld LA, Verlaan JJ, Oner FC: Spinal fractures in patients with ankylosing spinal disorders: A systematic review of the literature on treatment, neurological status and complications. *Eur Spine J* 18: 145-156, 2009
- 7) Westerveld LA, van Bommel JC, Dhert WJ, Oner FC, Verlaan JJ: Clinical outcome after traumatic spinal fractures in patients with ankylosing spinal disorders compared with control patients. *Spine J* 14: 729-740, 2014
- 8) Yunoki M, Suzuki K, Uneda A, Okubu S, Hirashita K, Yoshino K: The importance of recognizing diffuse idiopathic skeletal hyperostosis for neurosurgeons: A review. *Neurol Med Chir (Tokyo)* 56: 510-515, 2016
- 9) Okada E, Yoshii T, Yamada T, et al.: Spinal fractures in patients with diffuse idiopathic skeletal hyperostosis: A nationwide multi-institution survey. *J Orthop Sci* 24: 601-606, 2019
- 10) Rustagi T, Drazin D, Oner C, et al.: Fractures in spinal ankylosing disorders: a narrative review of disease and injury types, treatment techniques, and outcomes. *J Orthop Trauma* 31(Suppl 4): S57-S74, 2017
- 11) Werner BC, Samartzis D, Shen FH: Spinal fractures in patients with ankylosing spondylitis: etiology, diagnosis, and management. *J Am Acad Orthop Surg* 24: 241-249, 2016
- 12) Dhall SS, Wadhwa R, Wang MY, Tien-Smith A, Mummaneni PV: Traumatic thoracolumbar spinal injury: an algorithm for minimally invasive surgical management. *Neurosurg Focus* 37: E9, 2014
- 13) Kuperus JS, Westerveld LA, Rutges JP, et al.: Histological characteristics of diffuse idiopathic skeletal hyperostosis. *J Orthop Res* 35: 140-146, 2017
- 14) Kanematsu R, Hanakita J, Takahashi T, Tomita Y, Minami MJ: Thoracic hyperextension injury with opening wedge distraction fracture in DISH-consideration of surgical strategy based on intraoperative pathological findings. *J Clin Neurosci* 75: 231-234, 2020
- 15) Ikuma H, Takao S, Inoue Y, Hirose T, Matsukawa K, Kawasaki K: Treatment of thoracolumbar spinal fracture accompanied by diffuse idiopathic skeletal hyperostosis using transdiscal screws for diffuse idiopathic skeletal hyperostosis: Preliminary results. *Asian Spine J* 15: 340-348, 2021
- 16) Hishiya T, Ishikawa T, Ota M: Posterior spinal fixation using penetrating endplate screws in patients with diffuse idiopathic skeletal hyperostosis-related thoracolumbar fractures. *J Neurosurg Spine* 9: 1-6, 2021
- 17) Sedney CL, Daffner SD, Obafemi-Afolabi A, et al.: A comparison of open and percutaneous techniques in the operative fixation of spinal fractures associated with ankylosing spinal disorders. *Int J Spine Surg* 7: 23, 2016

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