


Urinary Retention as the Presenting Clinical Manifestation of Unstable Thoracic Spinal Fracture with Diffuse Idiopathic Skeletal Hyperostosis

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ABSTRACT: Patients with diffuse idiopathic skeletal hyperostosis (DISH) are at high risk for unstable vertebral fracture, which can be frequently missed. An 80-year-old man with pre-existing muscle lower limb weakness due to frailty was referred from another hospital, presenting with progressive urinary retention and its related symptoms, which had been treated as a urinary tract infection at previous hospital. One week prior to our visit, he had fallen. On arrival, he appeared lethargic and unable to follow commands. He denied any back pain. Computed tomography identified a T10 fracture and dislocation associated with DISH. Although immediate surgical fixation was performed, the patient did not recover from the neurological deficits. Diagnostic delay of DISH-associated vertebral fracture can occur due to both patients' and clinicians' delayed action. We believe this case report can help clinicians recognize this potentially devastating condition.

KEYWORDS: Hyperostosis, diffuse idiopathic skeletal, spinal injuries, traumatology, urinary retention

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Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) is a systemic disorder characterized by progressive abnormal ossification of ligaments.¹ The condition commonly affects the anterior longitudinal ligaments of the vertebrae in elderly patients.² Patients with DISH are highly susceptible to unstable vertebral fractures, even after low-energy trauma, resulting in secondary neurological deficits.³ Diagnosis of vertebral fractures associated with DISH is frequently delayed due to the condition's initially asymptomatic nature or unspecified pain in some patients and missed diagnosis or misinterpretation by physicians.⁴ Early recognition of this injury is crucial to avoid further complications. Here, we describe a unique case in which urinary retention and its related symptoms were the only clinical manifestation of an underlying thoracic vertebral fracture coexisting with DISH following a minor traumatic event. Our report should help raise clinician awareness of this potentially devastating injury.

Case Presentation

An 80-year-old Japanese man was referred from another hospital presenting with altered mental status, loss of appetite, and urinary retention. He had been in a long-term care facility for 10 years due to mild sequelae after measles, which he had

contracted when he was young. He was able to ambulate with the aid of a walker and independently perform all activities of daily living. Two and a half months prior to his visit to our hospital, surgical treatment was performed for a left femoral neck fracture resulting from a ground-level fall. Since then, the patient had needed a wheelchair due to his inability to cooperate and difficulty with rehabilitation. One week before his visit, he had fallen when he got out of his wheelchair independently. Three days after the fall, he was taken to a local clinic because of fever and urinary retention and was diagnosed with a urinary tract infection. On the day of his visit to our hospital, he presented with progressive altered mental status and loss of appetite over 3 days, which brought him to the psychiatric hospital, where no abnormal findings were revealed by head computed tomography (CT). Thereafter, he was referred to our hospital for further evaluation.

On arrival, he appeared lethargic, poorly responsive to questions, and unable to follow commands, which did not allow us to acquire an informative history and perform a thorough physical examination. The patient's vital signs were as follows: Glasgow Coma Scale score of 13 (E4V4M5), 24 breaths per minute, heart rate of 70 beats per minute, blood pressure of 107/70 mmHg, oxygen saturation of 98%, and temperature of 36.7°C. Lower abdominal distention was evident without any



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Figure 1. Sagittal CT image of thoraco-lumbar spine demonstrating transverse fracture and dislocation of the T10 vertebra (triangle arrow). Thick ossification of the anterior longitudinal ligament extending more than 4 contiguous vertebral bodies can be seen (arrow).



Figure 2. Bilateral hydronephrosis is visible (triangle arrow).

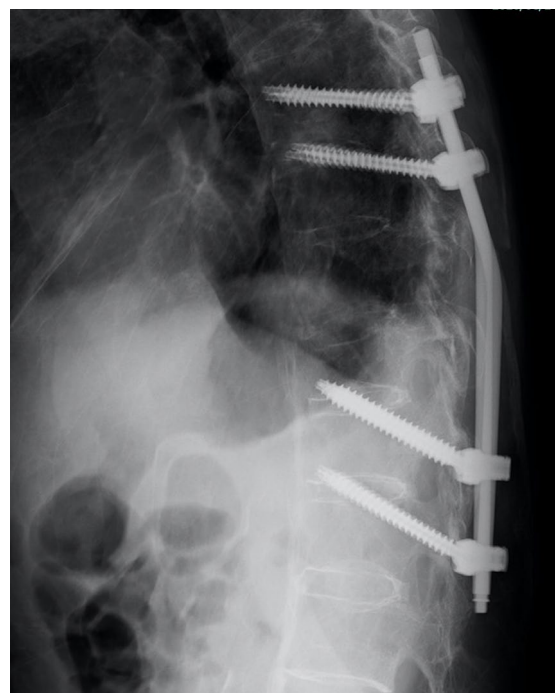


Figure 3. Postoperative spine X-rays showing T8-L1 posterior fixation.

remarkable findings on chest exam. Apparent weakness in his bilateral lower extremities was noted. The patient denied back pain. Blood test results showed elevated blood urea nitrogen of 63 mg/dL and creatinine level of 2.93 mg/dL. CT scan of the torso identified transverse fracture and dislocation of the T10 vertebra with extensively ossified anterior longitudinal ligament in the thoracic and lumbar spine region (Figure 1). Bilateral hydronephrosis was also detected (Figure 2). A limited physical examination revealed the absence of bilateral lower extremity reflexes and a decrease in anal sphincter tone. Active lower extremity motion was observed bilaterally; however, we were unable to administer a manual muscle test or assess skin sensation to touch in his torso or lower limbs.

Based on his history of falling from the wheelchair and examination, the patient was diagnosed with a T10 fracture associated with DISH, which due to its unstable nature had developed into a secondary neurological impairment, leading to urinary retention and subsequent kidney injury. The patient was immediately transferred to a tertiary care hospital where posterior fixation of the spinal fracture was successfully performed on the same day (Figure 3). The patient developed delirium and aspiration pneumonia, which was treated with antimicrobial therapy. He was discharged to a rehabilitation hospital on the 16th post-operative day, when it was observed his neurological deficits had not improved. Persistent delirium prevented the patient from performing rehabilitation. Postoperative treatment including lumbar brace or teriparatide

were therefore not applied. When the patient was transferred to a long-term care facility 2.5 months following the surgery, persistent neurological impairment was observed.

Discussion

Our case report attempts to illustrate the importance of early recognition and treatment of vertebral fractures associated with DISH, in which neurological deficits can develop following a recent episode of low-energy trauma. Physicians should keep in mind that progressive urinary retention and its related symptoms could be the only clinical signs of underlying unstable vertebral fractures in vulnerable patients.

Spinal DISH is defined as ossification of at least 4 adjacent vertebral bodies confirmed by radiologic study.¹ Although cervical DISH can induce dysphagia, the condition is usually

asymptomatic or causes minimal restriction of spinal mobility.^{5,6} Little is known about the etiology of DISH. An increase in the prevalence of DISH has been suggested due to its association with older age, obesity, diabetes mellitus, and hypertension.⁶ An important clinical feature is that the patients with DISH are prone to vertebral fracture following even a minor trauma such as a ground-level fall.^{4,6,7} This is explained by a mechanism in which the contiguous osseous fusion of the vertebrae acts as a “long-lever arm,” which can precipitate a displaced vertebral fracture.⁴ Owing to the condition’s instability, there is an increased risk of delayed spinal cord injury (SCI) and neurological impairment.

Diagnosis of DISH-associated vertebral fractures tends to be delayed.^{3,4,7,8} Some patients with this condition complain of only mild back pain or may not even notice back pain, which renders them reluctant to seek medical attention immediately after injury.^{8,9} From the doctor’s standpoint, insufficient sharing of information about this condition among emergency physicians or primary care physicians may be responsible for a delayed diagnosis.^{4,10} Non-specific tenderness of the back after a minor trauma would not warrant a thorough radiologic investigation. Hence, CT or magnetic resonance imaging is recommended to avoid a missed or delayed diagnosis.⁸ Otherwise, DISH-associated unstable vertebral fractures would not be recognized until neurological deficits appear. A recent nationwide survey in Japan reported that diagnosis was delayed in 40% of cases.¹¹ In the present case, the delay in coming to the hospital is attributed to the fact that the patient did not complain of any back pain following a trivial fall. Additionally, his pre-frail condition, in which he had already had weakness of lower limbs and had been dependent on the wheelchair after a hip fracture, resulted in manifestation of only urinary retention and its related complications caused by unstable thoracic spinal fracture.

As for treatment, early surgical fixation is recommended in patients with DISH-associated fractures complicated by neurological deficits or if instability is obvious or confirmed by additional stress imaging to avoid secondary neurological deterioration.^{3,8,10} Regarding surgical procedure, 2-above and 2-below segmental fixation using the double endplates penetrating screw could be applied in patients with DISH to secure rigid fixation as in this case.¹² Although it has been reported that functional improvement should not be expected once neurological impairment occurs,⁸ our patient underwent immediate posterior fixation to prevent further SCI and enhance rehabilitation.

Conclusions

We report a unique case of DISH-associated unstable thoracic vertebral fracture in a patient with frailty presenting with only

urinary retention and its related complications. Neurologic impairment, including muscle weakness of the lower extremities, was masked by the patient’s pre-existing frailty. When clinicians encounter patients manifesting with acute urinary retention even after a recent trivial fall, further diagnostic study should be considered to avoid delayed recognition of this devastating fracture. Since an increase in the prevalence of DISH has been suggested, we believe that this case report can alert clinicians to be aware of this critical condition.

Author Contributions

HH, TY, and SM contributed to the conception, design, and drafting the manuscript. AT, MA, KY, and AN made a critical revision of the manuscript. All authors approved the final version of this manuscript for submission.

Informed Consent

Written informed consent was obtained from his legal guardian and his family.

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REFERENCES

1. Resnick D, Niwayama G. Radiographic and pathologic features of spinal involvement in diffuse idiopathic skeletal hyperostosis (DISH). *Radiology*. 1976;119:559-568.
2. Diederichs G, Engelken F, Marshall LM, et al. Diffuse idiopathic skeletal hyperostosis (DISH): relation to vertebral fractures and bone density. *Osteoporos Int*. 2011;22:1789-1797.
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)*. 2010;35:458-464.
4. 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*. 2009;18:145-156.
5. Mattioli F, Ghirelli M, Trebbi M, Silvestri M, Presutti L, Fermi M. Improvement of swallowing function after surgical treatment of diffuse idiopathic skeletal hyperostosis: our experience. *World Neurosurg*. 2020;134:e29-e36.
6. Mader R, Verlaan JJ, Buskila D. Diffuse idiopathic skeletal hyperostosis: clinical features and pathogenic mechanisms. *Nat Rev Rheumatol*. 2013;9:741-750.
7. Dan Lantsman C, Barkay G, Friedlander A, Barbi M, Stern M, Eshed I. Whole spine CT scan for the detection of acute spinal fractures in diffuse idiopathic skeletal hyperostosis patients who sustained low-energy trauma. *Spine (Phila Pa 1976)*. 2020;45:1348-1353.
8. Murakami Y, Mashima N, Morino T, et al. Association between vertebral fracture and diffuse idiopathic skeletal hyperostosis. *Spine (Phila Pa 1976)*. 2019;44:E1068-E1074.
9. Siasios I, Pollina J, Dimopoulos VG. Hemoptysis as the presenting clinical sign of a T8-T9 spine fracture with diffuse idiopathic skeletal hyperostosis changes. *Case Rep Emerg Med*. 2016;2016:1-6.
10. Katoh H, Okada E, Yoshii T, et al. A comparison of cervical and thoracolumbar fractures associated with diffuse idiopathic skeletal hyperostosis—a nationwide multicenter study. *J Clin Med*. 2020;9:208.
11. 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*. 2019;24:601-606.
12. Takeuchi T, Hosogane N, Yamagishi K, Satomi K, Matsukawa K, Ichimura S. Results of using a novel percutaneous pedicle screw technique for patients with diffuse idiopathic skeletal hyperostosis—the single or double endplates penetrating screw (SEPS/DEPS) technique. *Spine Surg Relat Res*. 2020;4:261-268.