

Odontoid Fracture in a Patient With Diffuse Idiopathic Skeletal Hyperostosis

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Abstract

Nonoperative management of fractures in the setting of diffuse idiopathic skeletal hyperostosis (DISH) or ankylosing spondylitis is often unsuccessful. The subaxial spine is a common site of hyperextension fractures in the setting of DISH. Fractures of the upper cervical spine are uncommon in DISH. We report, to our knowledge, the first case describing successful nonoperative management of a type 2 odontoid fracture in a patient with DISH. We discuss the patient's initial presentation, physical examination, imaging findings, and management. A 73-year-old male presented with neck pain to the emergency department after sustaining a ground-level fall. Computed tomography of the cervical spine demonstrated a minimally displaced type 2 odontoid fracture in the setting of extensive DISH. He was immobilized with a hard cervical collar as the definitive management of his fracture. The collar was discontinued after 3 months. At his 2-year follow-up, he had a stable fibrous nonunion at the fracture site with tolerable neck pain. Flexion–extension radiographs demonstrated a stable alignment, and nonoperative management was continued. In selected patients with odontoid fractures in the setting of DISH, there is a role for nonoperative management alongside close monitoring.

Keywords

odontoid fracture, DISH, hyperostotic spine, diffuse idiopathic skeletal, hyperostosis, nonoperative management

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Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) also known as Forestier disease is a noninflammatory condition predominantly affecting the spine. The condition was first described in 1950, but as of date, the exact cause remains unknown.¹ Diffuse idiopathic skeletal hyperostosis causes calcification and ossification of the anterolateral spinal column and spinal ligaments (anterior longitudinal ligament).² The prevalence of this condition varies by population but appears to increase with age.³⁻⁵ Studies have also indicated that the disease is common and more severe in men than in women.⁶ The prevalence of DISH ranges from a low of 3.9% among African blacks⁵ to nearly 50% in Pima Indian males older than 44 years.⁴ The diagnosis of DISH is obtained via axial radiographic findings according to the Resnick criteria, whereas the newer criteria of Utsinger include the peripheral manifestations of the condition.^{7,8} Although the relatively newer criteria has lowered the threshold needed to diagnose the condition, it is still frequently diagnosed at a fully developed or late stage.^{9,10}

The ankylosis of a vertebral segment causes spinal stiffness and can contribute to a baseline level of pain. This chronic pain can often mask the onset of spinal fracture/instability, causing a delay in diagnosis.^{10,11} Diffuse idiopathic skeletal hyperostosis can predispose to spinal fractures from low-energy

mechanisms.^{2,10} Low-energy trauma is a common method of injury to the fastest growing population (geriatric) in the United States, and this population also have a greater tendency for upper cervical spine fractures.¹² Odontoid fractures comprise 9% to 15% of all cervical spine fractures and are the most common spinal fracture in patients older than 70 years of age.¹²⁻¹⁴ Odontoid fractures are classified as type 1, 2, or 3, depending on the location of the fracture. Type 2, fracture at the base of the odontoid, is the most common type of odontoid fractures. We report a case of a type 2 odontoid fracture in a patient with DISH successfully managed nonoperatively with a long-term follow-up.

Case Report

A 73-year-old male presented to the emergency department after sustaining a ground-level fall due to a syncopal episode.

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Figure 1. Sagittal computed tomography (CT) of the cervical demonstrating the odontoid fracture in the setting of diffuse idiopathic skeletal hyperostosis (DISH).

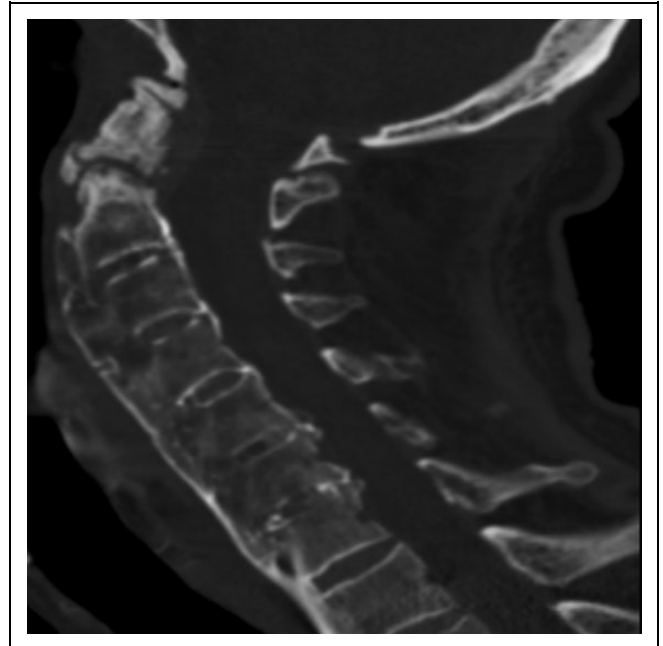


Figure 3. Sagittal computed tomography (CT) of the cervical spine performed at 2-year follow-up demonstrating the nonunion at the fracture site with minimal displacement.

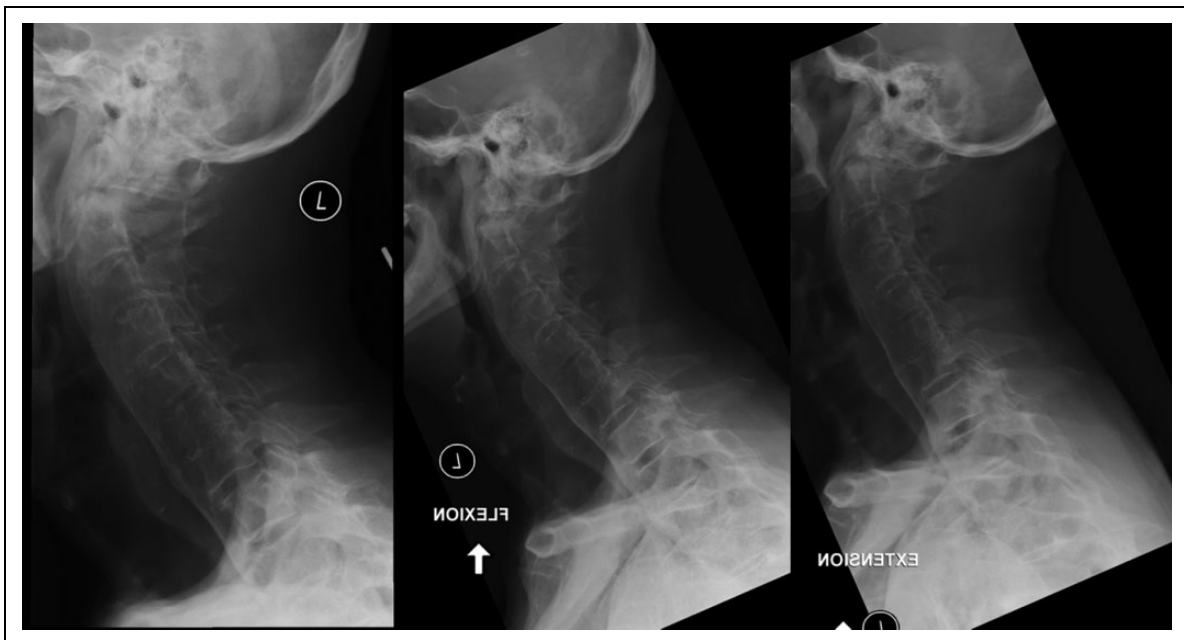


Figure 2. Lateral, flexion, extension radiograph at 2-year follow-up demonstrating a stable nonunion. Radiographs and CT of the cervical spine demonstrated a stable fracture nonunion of the odontoid fracture (Figures 2 and 3).

He was experiencing some neck pain. He had no weakness in the upper or lower extremities. He had no numbness and was ambulating with no difficulty. Per the emergency department protocol, a computed tomography (CT) scan of the head and cervical spine was performed. Cervical spine CT demonstrated DISH from C2 to T2 and a minimally displaced type 2 odontoid fracture (Figure 1). There was initial concern whether the

fracture is acute or chronic. However, since he was having pain and was neurologically intact, he was offered nonoperative management consisting of a hard cervical collar. At 2-year follow-up, the patient was experiencing mild neck pain. Radiographs and CT of the cervical spine demonstrated a stable fracture nonunion of the odontoid fracture (Figure 2). After discussing nonoperative versus operative options, the patient

and treatment team elected to continue with nonoperative management.

Discussion

Odontoid fractures are common injuries in the geriatric population.¹²⁻¹⁹ Odontoid fractures with minimal displacement can be treated nonoperatively with immobilization. In the setting of severe displacement or neurological changes, there's a role for operative management. Fractures of the cervical spine in the setting of hyperostosis (DISH, ankylosing spondylitis) are often encountered in the subaxial spine, usually at C6–C7.2 Hyperostotic spine fractures often result in 3-column injuries that render the spine unstable and often benefit from surgical intervention.

Odontoid fractures in the setting of DISH are rare. Diffuse idiopathic skeletal hyperostosis and ankylosing spondylitis of the cervical spine lead to a long lever arm similar to a long bone. This predisposes to unstable fractures often with displacement in extension. In our patient, C2 was the proximal extent of his DISH, and he did not sustain a neurological injury due to the minimal displacement of the fracture. He also did not have a 3-column injury or instability that required acute surgical intervention.

Nonunions can develop following nonoperative management of odontoid fractures in up to 88% of geriatric odontoid fracture.¹⁵⁻¹⁸ However, the effect of radiographic nonunion on clinical outcome is debatable and does not appear to correlate with patient-reported outcomes.¹⁷⁻¹⁹ In patients who fail nonoperative management, there is a role for surgical intervention, with C1–C2 posterior arthrodesis being the most effective.¹⁹ In a series of geriatric odontoid nonunions, 16 patients undergoing C1–C2 arthrodesis were compared to 28 patients undergoing nonoperative management at 5-year follow-up, and there was no difference in clinical outcomes.¹⁹

Several series have reported on the management of hyperostotic spine fractures (DISH and ankylosing spondylitis).^{2,10,20-23} To our knowledge, we are reporting the first successful nonoperative management of an odontoid fracture in the setting of DISH. Paley et al, in their series of 6 DISH patients with spine fractures, report on 1 patient with an odontoid fracture who was initially managed nonoperatively and 2 years following injury developed myelopathy at the fracture site.¹⁰ He was successfully managed with C1–C2 arthrodesis and halo immobilization. Whang et al, in their series of 18 DISH patients with spine fractures, had 1 patient with a C2 fracture (unclear whether it was an odontoid fracture). The patient had a complete spinal cord injury and was managed with halo immobilization and subsequently died.²² Lastly, Tsuji et al reported on a 73-year-old male with DISH and undiagnosed odontoid fracture that resulted in torticollis. The patient underwent surgical correction of the deformity and occipitocervical fusion.²⁴

In conclusion, we report the first, to our knowledge, successful nonoperative management of an odontoid fracture in the setting of DISH. Inherently, fractures in the setting of DISH

or ankylosing spondylitis are highly unstable, and it is seldom that nonoperative management succeeds. However, in the setting of nondisplaced odontoid fracture without neurological deficits, a trial of nonoperative can be attempted and possibly succeeded as demonstrated in our case.

Declaration of Conflicting Interests

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