

## Spinal fractures in the setting of diffuse idiopathic skeletal hyperostosis conservatively treated via orthosis: illustrative cases

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**BACKGROUND** Fractures in patients with diffuse idiopathic skeletal hyperostosis (DISH) are considered highly unstable injuries with high risk for neurological injury. Surgical intervention is the standard of care for these patients to avoid secondary spinal cord injuries. Despite this, certain cases may necessitate a nonoperative approach. Herein within, the authors describe three cases of cervical, thoracic, and lumbar fractures in the setting of DISH that were successfully treated via orthosis.

**OBSERVATIONS** The authors present three cases of fractures in patients with DISH. A 74-year-old female diagnosed with an acute fracture of a flowing anterior osteophyte at C6–C7 treated with a cervical orthosis. A 78-year-old male with an anterior fracture of the ankylosed T7–T8 vertebrae managed with a Jewett hyperextension brace. Finally, a 57-year-old male with an L1–L2 disc space fracture treated with a thoraco-lumbo-sacral orthosis. All patients recovered successfully.

**LESSONS** In certain cases, conservative treatment may be more appropriate for fractures in the setting of DISH as an alternative to the surgical standard of care. Most fractures in the setting of DISH are unstable, therefore it is necessary to manage these patients on a case-by-case basis.

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**KEYWORDS** DISH; spinal arthropathies; spine trauma; fractures

Diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier disease, is a type of noninflammatory arthritis principally characterized by excessive calcification of soft tissue in the spine resulting in gradual ossification of the anterior longitudinal ligaments and entheses.<sup>1–3</sup> Hyperostosis of the spine was initially described by Forestier and Rotes-Querol in 1950, the disease was then further characterized and defined as DISH by Resnick et al.<sup>1,2</sup> Diagnostic criteria for DISH includes the presence of calcification and ossification in the anterolateral aspect of at least four adjacent vertebrae, preservation of disc height without degenerative disc disease, along with the absence of apophyseal joint ankylosis, sacroiliac joint erosion, sclerosis, or bony fusion.<sup>1,2</sup> The pathogenesis of DISH is currently unknown, although it is thought to be associated with obesity, hypertension, diabetes mellitus, along with

deficiencies in mineralization inhibitors such as Matrix Gla protein.<sup>2,4,5</sup> Disease prevalence widely varies in the literature (2.9%–42%) likely due to differences in cohorts and diagnostic criteria.<sup>2,4,6</sup> DISH is rarely diagnosed before the age of 40 and most findings demonstrate increased prevalence with age.<sup>2,7</sup> The sex distribution is considerably different between races and ethnic groups.<sup>2,4,6,7</sup>

DISH patients are at risk for neurological complications due to spinal stenosis.<sup>8</sup> Moreover, segmental fusion along the dorsal vertebrae and across the intervertebral spaces results in an ankylosed spine with a high risk for three-column injuries even in low energy traumas such as falls.<sup>9–11</sup> Ankylosis most frequently occurs in the thoracic spine, particularly T7–T11.<sup>12</sup> Lower cervical and upper lumbar segments of the spine are also commonly involved in DISH.<sup>1,13,14</sup> Ossification of the thoracic

**ABBREVIATIONS** CT = computed tomography; DISH = diffuse idiopathic skeletal hyperostosis; MRI = magnetic resonance imaging; TLSO = thoraco-lumbo-sacral orthosis.

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spine is typically seen in the anterior aspect of the vertebral bodies, whereas posterior spinal osteophytes are common in the cervical segments. In the lumbar region, disc space narrowing and ossification of the interspinous ligaments may occur.<sup>1,13,14</sup> Fractures in the setting of DISH are associated with high rates of mortality and complications. Due to the inherent three-column injury following these fractures, nonoperative (conservative) treatment is typically not recommended, and the standard of care is surgical management.<sup>15–17</sup> In this report, we describe three cases of spinal fractures in the setting of DISH that were treated successfully through a conservative approach.

## Illustrative Cases

### Case 1

A 74-year-old female presented to an outside hospital with a chief complaint of neck pain after falling backward while roller skating. Computed tomography (CT) images of the cervical segment showed normal spinal alignment. Extensive ossification was indicated for both the posterior and anterior longitudinal ligaments extending from the C2–C3 to the C6–C7 level. Further findings demonstrated discontinuity involving the anterior C6–7 osteophyte with widening of the disc space (Fig. 1A). On radiograph, flowing anterior osteophytes consistent with the presence of DISH were most apparent at the C2–C3 and C3–C4 levels as well as contiguous ossification of the posterior longitudinal ligament spanning C3–C5 (Fig. 1B). The C6–C7 disc space level was partially obscured by superimposed osseous and soft tissue structures. Magnetic resonance imaging (MRI) also identified discontinuity of the anterior osteophyte at the C6–C7 level. As a result of these image findings, the patient was diagnosed with an acute fracture of the flowing anterior osteophyte at C6–C7 in the setting of DISH. Relevant findings included severe canal stenosis with moderate compression of the cord with no abnormal cord signal extending from C2–C7 predominantly due to ossification of the posterior longitudinal ligament. The patient remained neurologically intact throughout the admission and was discharged in a PMT cervical orthosis collar to be worn at all times. A 6-week follow-up upright cervical radiograph showed no abnormal movement at the C6–C7 level (Fig. 1C). Neck pain was no longer indicated upon neck rotation,

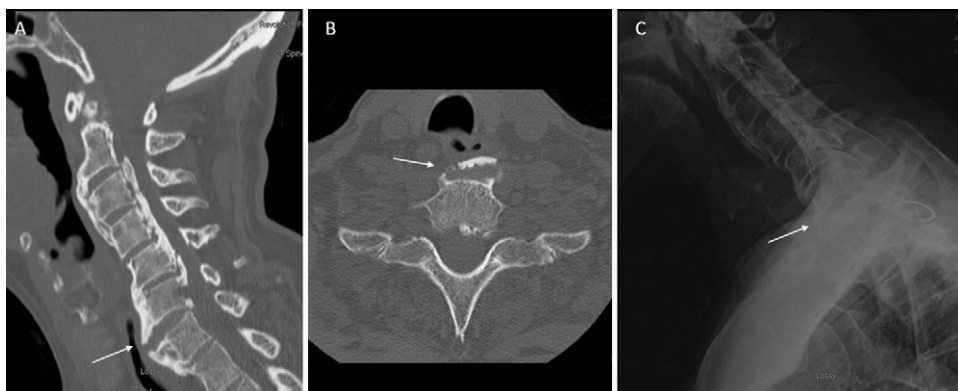
flexion, and extension. At this point, the brace was slowly weaned without complication.

### Case 2

A 78-year-old male with a history of DISH was brought to the emergency department by ambulance after he slipped and fell at home. The patient complained of new back pain since the fall. CT of the thoracic spine indicated an acute appearing anterior fracture of the DISH ankylosis at T7–T8 with widening of the anterior disc space and sharp appearing fracture margins (Fig. 2A). The fracture propagates to the anterior superior corner of the T8 vertebral body posteriorly and inferiorly through the posterior inferior endplate, but not clearly into the posterior elements. There was a small amount of paravertebral hematoma seen anteriorly and laterally adjacent to the fracture site on the axial images (Fig. 2B). The patient presented and remained neurologically intact throughout his admission. After a discussion with the patient and his family, the patient was discharged with a Jewett brace to be worn anytime head of bed is greater than 30 degrees or when out of bed. A 7-week follow-up radiograph indicated acceptable alignment of the thoracolumbar spine with no widening of the T7–T8 space, due to interval healing (Fig. 2C). The patient demonstrated excellent recovery and was permitted to begin weaning the brace at this point.

### Case 3

A 57-year-old male with a history of epilepsy and cognitive delay was brought to the emergency department by ambulance; he complained of back pain after having a seizure and falling to the ground, hitting the bathroom sink while doing so. Impressions from CT of the lumbar spine demonstrated a distraction-type fracture at L2 with associated widening of the anterior disc space (Fig. 3A). This was concurrent with fractures of the left L2 transverse process and spinous process. Multilevel intervertebral disc space height loss and vertebral body osteophyte formation were indicated, most advanced at L2–L3 (Fig. 3B). These findings led to a diagnosis of L1–L2 disc space fracture with posterior element involvement in the setting of DISH. Given these findings, and the patient's baseline comorbidities, the patient and his family elected to attempt conservative therapy as opposed to surgical intervention. This was conservatively treated with a thoraco-lumbo-sacral



**FIG. 1. A:** CT scan of the cervical spine without IV contrast, sagittal view. An anterior osteophyte fracture is present through C6–C7 with sharp fracture margins and associated widening of the C6–C7 disc space. **B:** CT scan of the cervical spine without IV contrast, axial view at the C6–C7 disc space. **C:** Follow-up lateral projection radiograph of the cervical spine with good alignment.



**FIG. 2. A:** CT scan of the thoracic spine without IV contrast, sagittal view. *Arrow* indicating T7–T8 sharp fracture margins. **B:** CT scan of the thoracic spine without IV contrast, axial view. *Arrow* indicating paravertebral hematoma. **C:** Postbrace lateral projection radiograph of the thoracolumbar spine. *Arrow* demonstrated site of injury aligned without widening due to interval healing.

orthosis (TLSO) brace to be worn at all times. The patient was at their neurological baseline throughout their hospitalization. The patient reported improved back pain during the 6-week follow-up. At the 12-week follow-up radiograph, the fracture fragment appeared reduced and was now anatomically positioned without a visible fracture line (Fig. 3C). The patient was permitted to wean the TLSO brace at this point.

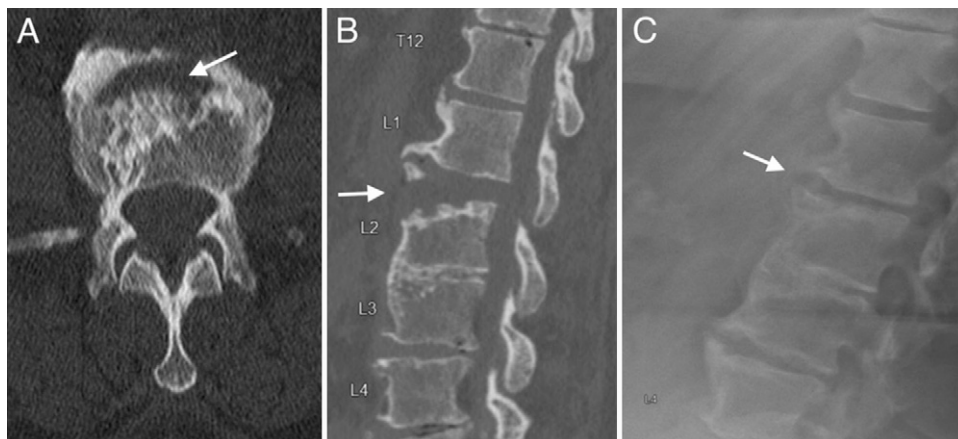
## Discussion

### Observations

DISH is a type of noninflammatory arthritic disease characterized by calcification and ossification of soft tissue within the spine causing ankylosis. In a healthy, nonankylosed spine, energy from impact trauma is distributed over mobile segments and their surroundings ligaments. In the setting of DISH, large lever arms of force are applied to the spine due to the presence of extensive multilevel fusion. Given these large lever arms, injuries that occur due to a relatively small force are compounded resulting in more severe fracture patterns and higher risk for neurological injury. Additionally as

these fracture typically occur over fused segments, the majority result in three column injuries by definition.<sup>18</sup> Given the inherent instability of these fractures, early recognition is necessary to avoid injury of the spinal column and neurological deterioration.<sup>19</sup> Recommendations for treating fractures in patients with DISH call for surgical intervention, while conservative treatment is generally avoided due to the high risk of secondary spinal cord injury.<sup>18,20</sup> Despite this, in certain cases surgical intervention may be less desirable due to underlying medical comorbidities or concurrent traumatic injuries requiring more urgent treatment. In our study we provide a brief case series describing how in these settings clinicians can opt for a nonoperative approach.<sup>17,21</sup> This is important to avoid the inherent risk of operating and may serve to benefit cases that necessitate a conservative approach.

As demonstrated in the three cases described above, certain fractures in patients with DISH may be stable enough to treat via bracing without surgical intervention. When considering these, the surgeon should carefully consider the biomechanics of the fracture in question as well as the biomechanics of the orthosis being selected to maximize the likelihood of successful healing. In the above cases all three



**FIG. 3. A:** CT scan of the thoracic spine without IV contrast, axial view. *Arrow* indicating L2 ligament avulsion. **B:** CT scan of the thoracic spine without IV contrast, sagittal view. Multilevel intervertebral disc space height loss is seen with *arrow* indicating distraction-type fracture. **C:** Postbrace lateral projection radiograph of the thoracolumbar spine. *Arrow* demonstrating reduced fracture fragment in standard anatomical position.

injuries were extension type injuries with normal facet orientations and without significant dislocation. Additionally, in these cases upright alignment, and the orthosis of choice in each case, allowed for significant resistance against extension such promoting fracture reapproximation and thus fusion. As such, in cases with good alignment and relative contraindications to surgical intervention, external orthosis may be a reasonable consideration in carefully selected cases. Despite this, it should be noted that the standard of care for these fractures remain surgical fixation and fusion.

## Lessons

By adding these cases to the literature, we aim to report scenarios in which conservative treatment may be more appropriate for fractures in the setting of DISH as an alternative to the surgical standard of care. Although most fractures in patients with DISH are unstable, it is necessary to manage these patients on a case-by-case basis.

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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: Page, Taher, Greeneway, Ammanuel, Meisner, Hanna, Josiah. Acquisition of data: Page, Greeneway, Hanna. Analysis and interpretation of data: Page, Bunch, Josiah. Drafting the article: Page, Taher, Ammanuel. Critically revising the article: Greeneway, Ammanuel, Bunch, Meisner, Josiah. Reviewed submitted version of manuscript: Greeneway, Ammanuel, Bunch, Meisner. Administrative/technical/material support: Hanna. Study supervision: Josiah.

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