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CASE REPORT

A Rare Case of Diffuse Idiopathic Skeletal Hyperostosis with Stable Burst Fracture of D12 Vertebra Presenting with Low Back Pain: A Case Report

[1]Amit A Sequeira, [2]Gautam Das, [3]Karan Patel

ABSTRACT

Diffuse idiopathic skeletal hyperostosis (DISH) is a relatively poorly studied disease in the Indian population. The characteristic radiologic findings of this disorder are the presence of "flowing ossification" especially along the anterolateral borders of the vertebral bodies. Trivial trauma, especially involving hyperextension of the spine, can lead to vertebral fractures, sometimes with serious neurological deficits. We present a case of DISH involving fracture of D12 and L1 vertebrae, following a minor fall at home, which was managed conservatively due to multiple risk factors for surgery. The patient presented to our pain clinic with signs and symptoms suggestive of the lower lumbar facet and bilateral sacroiliac joint arthropathy, without any features of D12, L1 fracture or any neurological deficits. We postulate that the lower lumbar and SI pathology could be a sequel of the orthotic support used in the conservative management of DISH in this patient or a natural progression of the disease.

Keyword: Burst fracture, DISH, Flowing ossification

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INTRODUCTION

Diffuse idiopathic skeletal hyperostosis (DISH) is a skeletal disease of unclear etiology most commonly presenting in the elderly male population. The hallmark of this disorder is the ossification of the ligaments along the anterolateral part of the vertebral column. Extra-axial entheseal ossification and bony spurs can also be presentations of this disease. The diagnosis is primarily radiological. Osteoporosis is usually not associated with DISH but could be controversial depending on the methods used to assess bone mineral density.[1] These patients can present with vertebral fractures following trivial injury especially involving hyperextension of the spine.

[1,3]Fellow, [2]Director

[1-3]Daradia Pain Clinic, Kolkata, West Bengal, India

Corresponding Author: Amit A Sequeira, Fellow, Daradia Pain Clinic, Kolkata, West Bengal, India e-mail: amitseq@gmail.com

CASE REPORT

A gentleman aged 75-year presented to the clinic with a history of low backache. The pain was non-radiating, midline and exacerbated on extension of the back, rising from sitting position and turning sideways in bed. The patient noticed the above symptoms for the past 4 to 5 months. His pain detect score (PDS) was 4. Previous medical history included Type 2 diabetes mellitus, Ischemic heart disease, Parkinsonism, and hypertension, all of which were reasonably controlled with medications.

A detailed history-taking revealed that the patient had suffered a fall on his back at home one year ago. Immediately after the fall, he experienced pain in his upper back along the midline. Both flexion and extension were painful at that time. Referral to a neurosurgeon and subsequent magnetic resonance imaging (MRI) revealed that he had suffered a fracture of D12, L1 vertebrae. Since he had no detectable neurological deficits and because of his advanced age with multiple comorbidities, a conservative line of management involving the use of Taylor's brace was advised together with regular follow-up with his doctor. The pain in the upper back lasted for 2 to 3 months following which he was reasonably comfortable for a few months. About 6 months after the initial injury he developed the low back ache for which he presented to our clinic.

On examination, no neurological deficits were noted. Percussion of the spine in the region of the fracture failed to elicit any tenderness. Lateral rotation and extension of the spine produced pain in the low back and gluteal region.

On palpation, bilateral paramedian tenderness was present; corresponding to L3-4, L45 facets, both sacroiliac joints were tender. Extension of the spine mimicked the pain the patient had been experiencing the past few months.

The spine X-ray, computed tomography (CT) scan, and MRI findings were typical of the DISH. A transverse fracture of the D12 vertebral body, with wedge compression of L1, was noted. The detailed radiological findings can be found later in this article.

From the above findings, a provisional diagnosis of DISH with facet arthropathy (lower lumbar region), bilateral SI joint arthropathy and a stable vertebral body fracture at D12, the L1 level was made.

It was presumed that the relative immobilization of the dorsal and upper lumbar spine due to Taylor's brace altered the biomechanics and weight distribution along the lower lumbar spine, causing hypertrophy of the lower facet joints and sacroiliac joints with resulting arthropathic symptoms.

The patient was prescribed a 5-day course of thiocolchicoside, paracetamol, and ondansetron together with duloxetine 20 mg at bedtime for 2 months. Follow up was advised after 3 weeks. A diagnostic facet and SI joint injection were planned on the subsequent visit only if there were no reduction of symptoms.

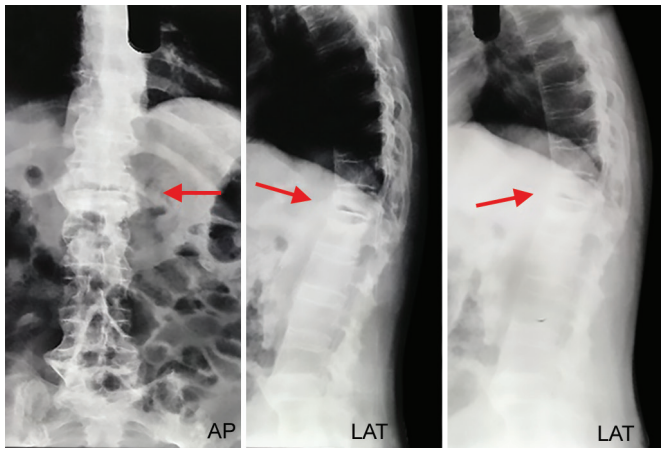


Fig. 1: Plain X-ray

RADIOLOGICAL FINDINGS

Plain X-ray

Flowing ossifications were seen along the anterolateral aspects of the visualized spine–more than four contiguous vertebrae.

Lucency seen between the ossification and the anterior aspect of the vertebral bodies on the lateral view. Transverse fracture noted through the lower half of the body of D12 with loss of height (arrow in Fig. 1).

Computed Tomography

Anterior wedging of L1 seen. Bridging osteophyte seen at L4,5. Rest of vertebral bodies appear normal. Facet hypertrophy is seen. Intervertebral disc spaces are normal. Sl joints normal.

A transverse fracture is seen through the lower half of the body of D12 (arrow in Fig. 2A). Vacuum was seen at the fracture site. Sclerosis of the upper half of the vertebral body seen is suggestive of compression. Extrusion of discal material seen anterior to the vertebral body with calcification within. Mild retropulsion was seen. No significant canal narrowing. Extensive ossification of the posterior elements seen at this level.

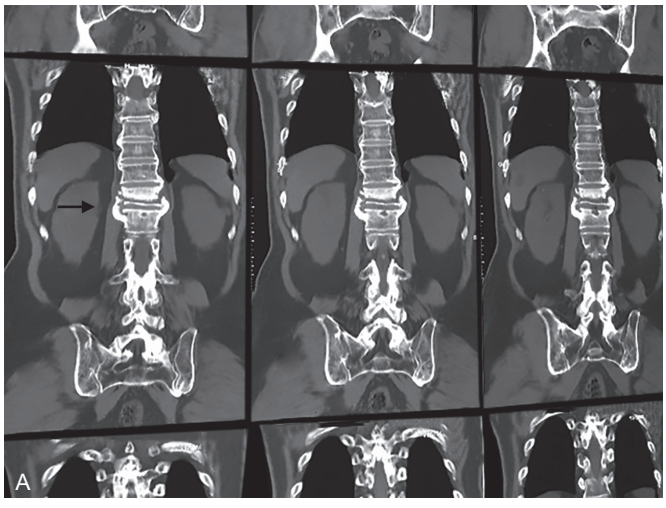
Lower dorsolumbar spine: Flowing ossification seen along the anterolateral aspect of the entire visualized spine (arrow in Fig. 2B). Thin lucency was seen between the ossification and the anterior aspect of the vertebral bodies.

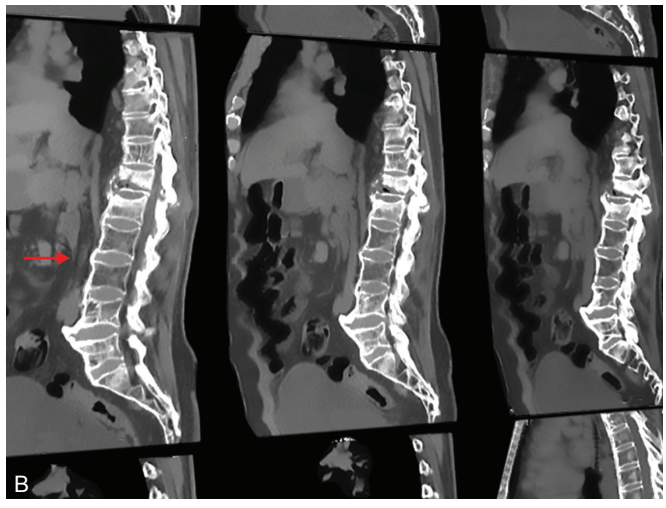
There is anterior wedging of L1. Presence of bony excrescences/bridging osteophyte from L1. Ligamentum flavum ossification seen in the lower dorsal level causing mild spinal canal narrowing. Opacification of the posterior longitudinal ligament seen at L2/3 (Figs 2C and D). Facet hypertrophy seen at multiple levels. SI joints are normal.

Magnetic Resonance Imaging

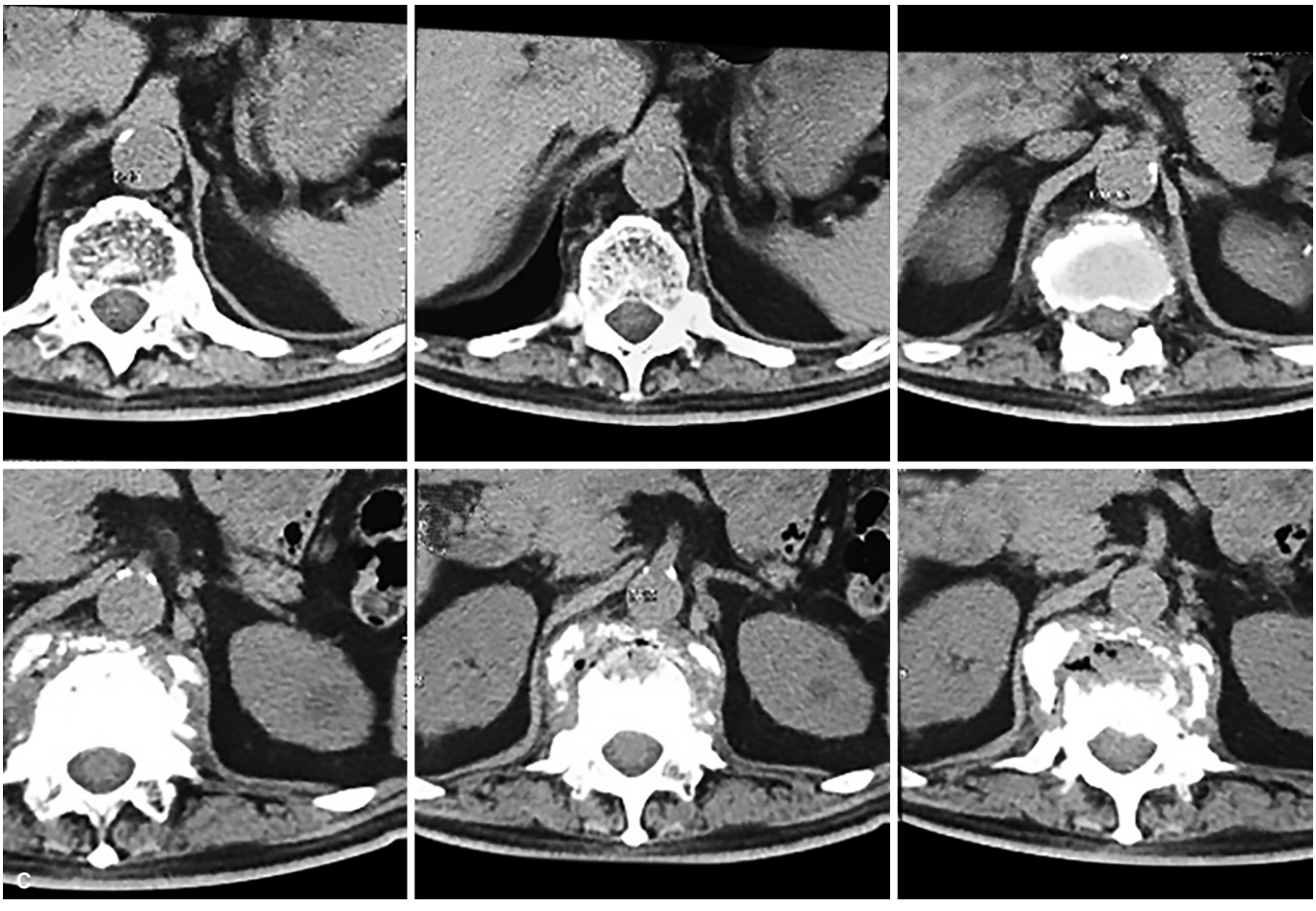
Lower Dorsolumbar Spine

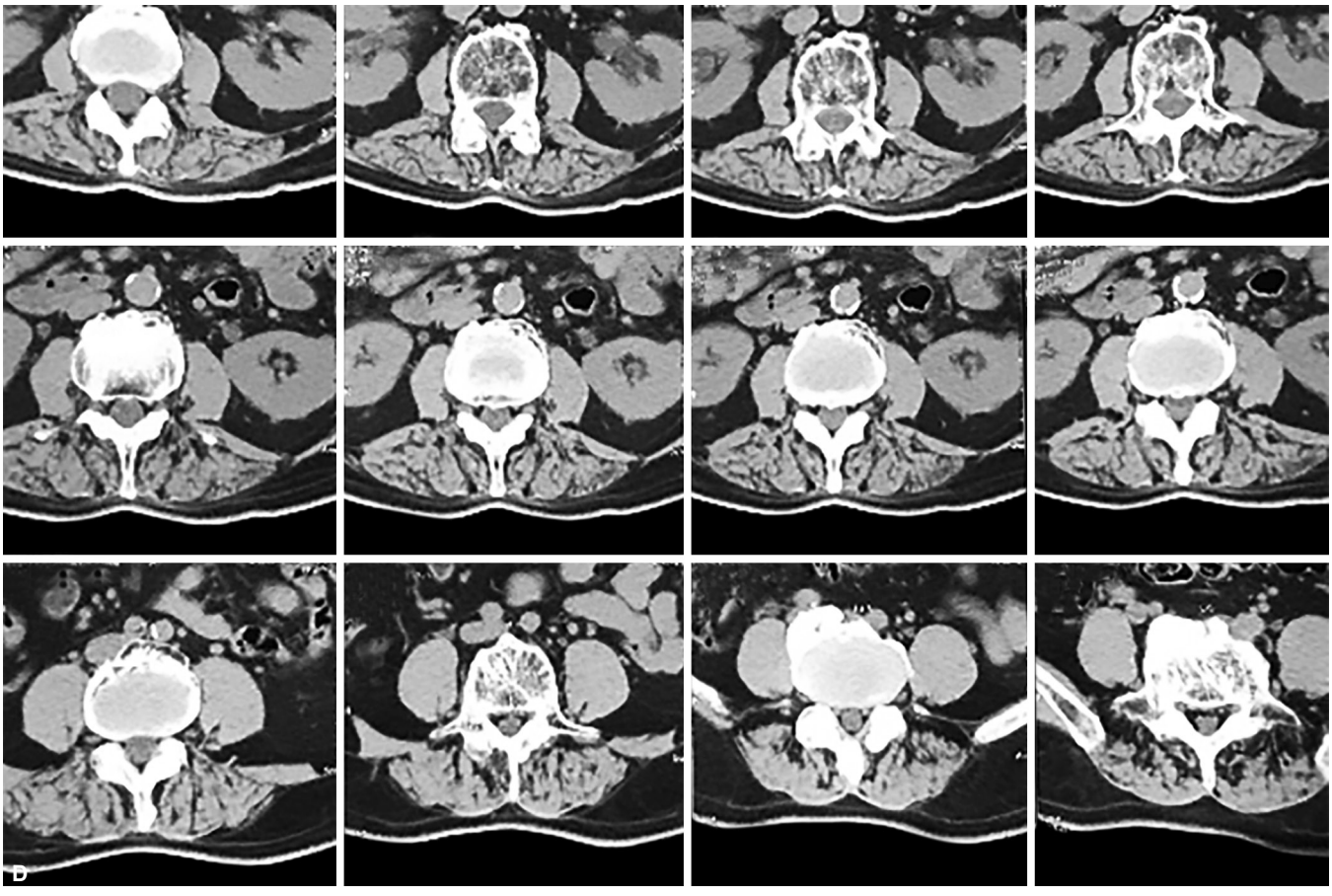
Axial T2 (Fig. 3A) and T1 (Fig. 3B) sequences showing the low signal area seen between the vertebral body and the anterior longitudinal ligament (ALL)–most probably protruded discal material with calcification within. Mild retro-pulsion of the vertebral body seen with focal



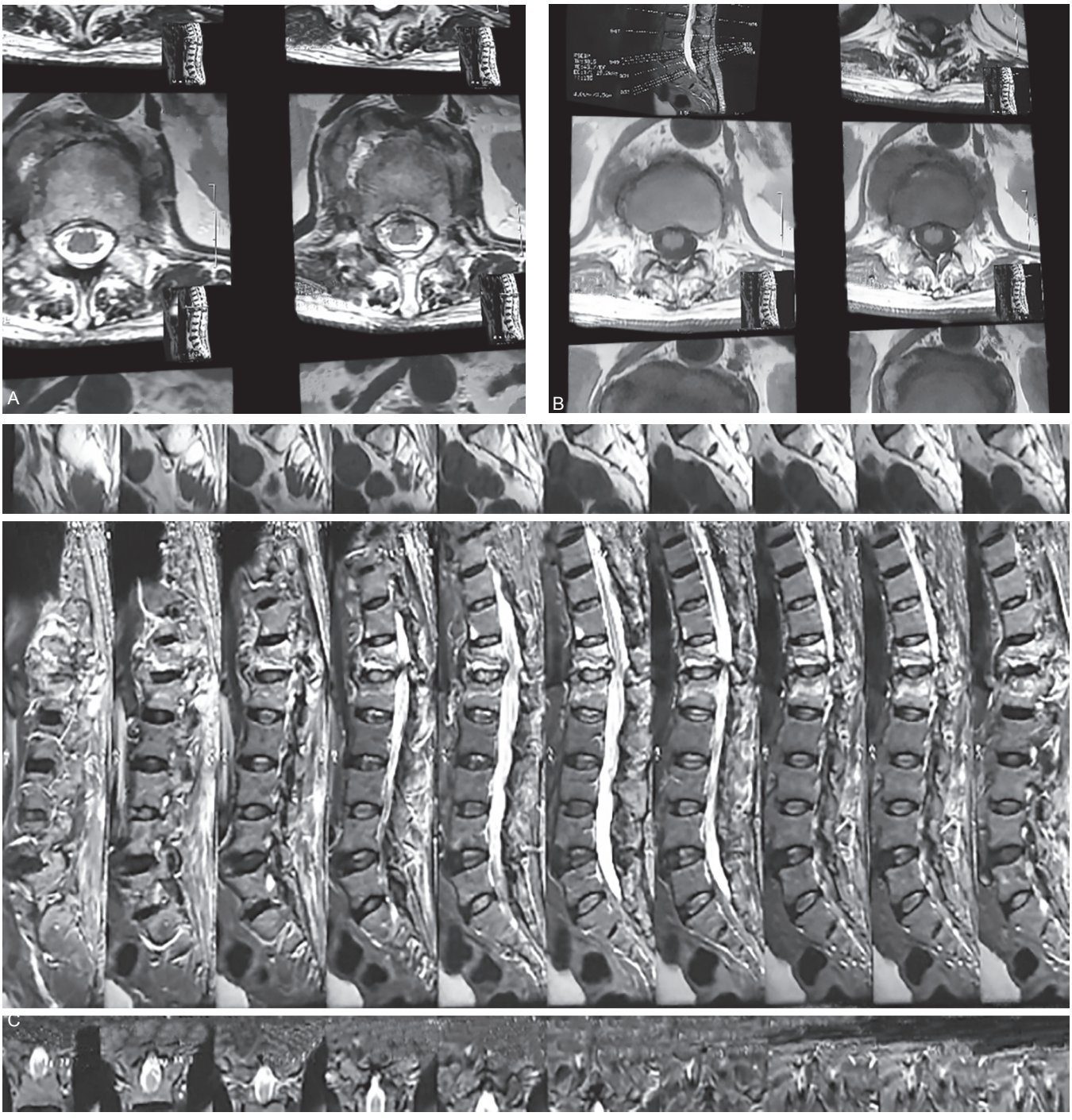


Figs 2A and B: Computed Tomography (CT)





Figs 2C and D: Computed Tomography (CT)



Figs 3A to C: Magnetic Resonance Imaging (MRI)

cord indentation no cord edema seen. Ossification and hypertrophy of the facets seen at this level. Straightening of the spine seen. Focal kyphosis was seen at D12 level (Fig. 3C). Transverse fracture line-showing high signal on T2 and low signal on T1 sequences–seen along the inferior aspect of the D12 vertebral body. Loss of height of D12 seen. There is a focal elevation of the anterior longitudinal ligament at this site (Fig. 3C). Anterior wedging of L1 seen. Bridging osteophyte was seen at L1. The focal altered signal is seen along the anterior aspect of D11. Disc dehydration seen at multiple levels without disc height narrowing. SI joints are normal.

DISCUSSION

The DISH, also known as Forestier's disease, spondylitis ligamentosa ossificans or senile. Ankylosing hyperostosis of the spine, is a skeletal disorder of unknown etiology described in the elderly and characterized by abundant bone formation, ossification, and calcification of connective tissue in spinal and extraspinal sites.[1] The principal manifestation of DISH is ligamentous ossification of the anterolateral aspect of the spinal column, sometimes leading to bony ankylosis.[2] The diagnosis of the DISH in most patients is made with radiography and, if needed,

further characterization may be provided by cross-sectional imaging, including CT and MRI. Diagnostic criteria suggested by Resnick are:[3]

- Presence of "flowing" calcification and ossification along the anterolateral aspect of at least four contiguous vertebral bodies with or without associated localized pointed excrescences at the intervening vertebral body–intervertebral disc junctions..
- Presence of relative preservation of intervertebral disc height in the involved vertebral segment and the absence of extensive radiographic changes of "degenerative" disc disease including vacuum phenomena and vertebral body marginal sclerosis.
- The absence of apophyseal joint bony ankylosis and sacroiliac joint erosion,

Sclerosis or Intra-Articular Osseous Fusion

All three radiographic criteria must be fulfilled to establish a definitive diagnosis of the DISH. The first criterion differentiates this disorder from typical lumbar spondylosis second criterion differentiates it from typical intervertebral osteochondrosis/disc disease, and the final criterion makes it distinct from Ankylosing Spondylitis.[3]

In DISH involving the thoracic spine, flowing ossifications are commonly seen along the right lateral aspect and not on the left lateral aspect, probably due to the inhibiting effect of pulsations of the descending aorta on the formation of osteophytes.[4]

Etiology and Incidence

DISH is primarily a disease affecting males. It usually manifests in the seventh decade of life.[1,3] Though it may be seen as early as the fourth or fifth decade of life.[5]

The incidence in the Caucasian population varies from 12.4[6] to 52%.[1] Statistical data in the Indian population was unavailable.

It has been associated with diabetes, lipidemia, and hyperuricemia.[5] Some studies have found associations with gout and rheumatoid arthritis.[2]

DISH patients were more likely to weigh more at a young age put on more weight, and their BMI was greater at the time of clinical evaluation.[2]

Clinical Features

Spinal symptoms are usually mild and include back stiffness and mild, intermittent and non-radiating dorsolumbar pain that becomes more prominent in middle age. Increasing pain and stiffness herald disease progression in the cervical and lumbar spine. Despite this, a large number of patients remain asymptomatic, and the disease is discovered incidentally,[5] about 15% of patients may present with serious neurological deficits that require neurosurgical intervention.[7]

Axial skeleton: Back stiffness, mild pain in the middle to lower back. Starts during middle age and persists for many years. Starts in the D-L spine. Appears early in the morning, dissipates with mild activity. May return late evening. Aggravated by sitting and cold and wet weather. After a few years, progresses to lumbar and cervical regions.

The lower thoracic region is more commonly affected than the cervical or lumbar regions.[5]

Cervical dysphagia may be due to cervical osteophytes.[8,9] Ossification of the posterior longitudinal ligament and ossification of the ligamentum flavum can give rise to neurologic symptoms.[3]

Extra-axial skeleton:[3]

- Recurrent achilles tendinitis.
- Recurrent tennis elbow.
- Progressive restriction of range of movements.
- Palpable calcaneal enthesophytes.
- Palpable olecranon enthesophytes.
- Nodular masses adherent to the quadriceps-patellar tendons

Biomechanics and Pathophysiology of fractures in DISH

Acute spinal fractures associated with DISH are not common but can lead to serious complications, including nonunion, deformity, neurologic injury, and death.[4]

Controversy exists about the implications of vertebral ossifications for the mechanical stability of the spine. It has been suggested that the fused segments are more prone to fracture even after minimal trauma.[10]

On the other hand, different studies have shown consistently higher bone mineral density (BMD) in patients with hyperostosis, imp3 ing a lower fracture risk.[1] Osteoporosis is not found in DISH[4,5] however, flowing ossifications may lead to overestimation of BMD values by dual X-ray absorptiometry.[1]

In patients with DISH, the bridging osteophytes and subsequent ankylosis of the spine makes it more prone to fracture, even after trivial trauma. The ossifications are most pronounced at the disk space level. In DISH, spine fractures can be of two types. The most common type is where the fracture line follows a path of least resistance, close to the middle of the vertebral body. The second type of fractures occur at the junction of an ankylosed segment with the healthy spine, at the level of the intervertebral disc.[4,5] The latter type of fracture is mostly seen in ankylosing spondylitis.[11]

Advanced cases of DISH are usually associated with spinal fractures. The thoracic and cervical segments are more commonly affected than the lumbar spine.[4]

Hyperextension is the most common mechanism of injury. The most common site of fracture is the center of the vertebral body, an area usually with the least bone mass (and thus least resistance to fracture) in the DISH.[12]

Management of Spine Fractures in DISH

Early fracture stabilization is a prerequisite to prevent complications like nonunion, osteolysis, late instability, and neurologic deficit, operative intervention is usually reserved for significantly displaced or unstable fractures. Conservative treatment with bed rest, orthotic devices, or halo vest can be used for more stable, minimally displaced fractures or as an adjunct to surgery.[11]

In our patient, we find that the transverse fracture at D12 occurs at the most predictable site of fracture-junction of a thoracolumbar spine as well as the midpoint of the vertebral body, the area of least bone mass in the DISH.

We postulate that the presence of diffuse hyperostosis especially in the posterior segment and flowing ossifications have probably prevented the unstable fracture from causing any neurological deficits.

So, the timely use of Taylor's brace for spine immobilization could have hastened fracture healing since the basic hyperostotic process in DISH is expected to speed union by exuberant bony consolidation of the fractured spine.[11]

CONCLUSION

DISH is a disorder whose incidence is poorly documented in the elderly male Indian subpopulation. We have presented this case which shows a typical radiological picture of the DISH. The relatively unstable vertebral fracture had to be managed conservatively due to the high surgical risk in this particular patient. The lack of progressive neurological dysfunction also played a major role in opting for this line of treatment. Probably, the presence of diffuse ossification prevented the unstable fracture from producing any neurological symptoms. So, the prompt use of Taylor's brace helped to immobilize the spine and facilitate the hyperostotic process that is typical of the DISH, thus hastening fracture healing.

Of particular interest is the development of painful secondary changes in the lower lumbar facet and SI joints, for the relief of which, this patient presented to our clinic. It is presumed that the abnormal load distribution secondary to the use of an orthotic device to stabilize the injured spine or a natural progression of the disease, could have led to the lower lumbar facet and Sl joint arthropathy.

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