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CASE REPORT

FORESTIER SYNDROME CAUSING DYSPHAGIA: A CASE REPORT

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ABSTRACT

Diffuse idiopathic skeletal hyperostosis, or Forestier's disease, is characterised by increased bone mass. Its etiology is currently unknown. The disease predominantly manifests as cervical and back pain. Some patients also present with additional symptoms like dysphagia, neck pain, and dyspnea. Several cases of surgical treatment are reported in the medical literature. Surgical treatment is attached to the specific symptoms or complications and generally involves osteophytectomy through the usual anterior approach. The existence of diffuse idiopathic skeletal hyperostosis has been related to older age, male sex, obesity, hypertension, atherosclerosis, and diabetes mellitus. This case report elucidates our patient-centric approach to managing diffuse idiopathic skeletal hyperostosis and aims to share insights from our experience.

Keywords: Dysphagia, ligaments, ossification, spinal osteophytosis, surgery

INTRODUCTION

Diffuse idiopathic skeletal hyperostosis (DISH), or Forestier's disease, is a perplexing condition characterised by the ossification of paravertebral ligaments and muscles. Notable clinical manifestations include dysphagia, neck pain, and dyspnoea (1-4). According to Childs (5), DISH is assumed to be a different kind of osteoarthritis (OA) without the degenerative intervertebral disc and joint degenerative qualities seen in classic OA. Conversely, Kuperus et al. (4) mentioned in their article that DISH's pathogenesis is occult. The existence of DISH has been related to older age, male sex, obesity, hypertension, atherosclerosis, and diabetes mellitus (4). This report delineates the case of a 66-year-old male who presented with persistent dysphagia over a year, culminating in a significant weight loss of 25 kilograms within six months. An exhaustive literature review was undertaken concurrently with this case study to enhance our comprehension of DISH.

CASE REPORT

A 66-year-old man, a retired counterterrorism officer, was admitted to the neurosurgery department. He had experienced stinging in his hands and progressive regression in fine motor skills in the upper extremities, such as the inability to button up or hold a spoon. He also complained of dysphagia for the last year, leading to a weight loss of 25 kilograms over six months. His medical and family histories were unremarkable. He had received physical therapy for 15 days, and although the stinging in his hands slightly improved, there was no improvement in motor skills and swallowing difficulties. Upon admission to our facility, he underwent a two-phase surgical intervention: excision of the anterior calcified disc and osteophytes compressing the oesophagus, posterior decompression by laminectomy and posterior stabilisation.

In the preoperative computed tomography (CT), widespread, bridging, osteophytic new bone formations were observed in the anterior left paracentral part of the C2-C7 vertebral body in the anterior longitudinal ligament trace. In addition, the C4-C5-C6-C7 vertebral bodies appeared to be fused anteriorly and posteriorly. The findings were initially compatible with Forestier's disease. At the C4-C6 level, ossifications in the posterior longitudinal ligament trace were causing narrowing in the central canal (Figure 1). A preoperative magnetic resonance imaging was performed and in addition to the above findings,

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there were degenerative changes at the C3-4 C4-5 disc levels and a significant narrowing of the bilateral neural foramen secondary to dorsal osteophytes. At the C3-4 disc level, the spinal canal anterior-posterior diameter was measured as 6.8 mm and was narrowed (Figure 2).

A three-level (C3, 4, 5) anterior decompression involving osteophyte drilling was executed, supplemented by posterior decompression by laminectomy and stabilisation using lateral mass screws. In the postoperative CT, heterogeneity and occasional air densities were observed at the level of posterior elements in C2, 3, and 4 vertebrae. Bilateral screws and fixators were observed at these levels. It was observed that the anterior osteophytes between the C2 lower endplate and the C5 upper endplate were excised (Figures 3, 4). The perioperative phase was uneventful, with no significant bleeding or neurological complications. However, postoperative feeding led to aspiration, necessitating intubation and subsequent transfer to the intensive care unit. Despite a normal neurological examination, the patient's significant weight loss had resulted in muscular atrophy, prompting a tracheostomy. Due to persistent respiratory and swallowing challenges, he was on a nasogastric tube and eventually discharged in the 2nd postoperative month with appropriate nutrition. At the 6th month follow-up, respiratory and swallowing difficulties were persistent, and the patient still had a tracheostomy and a nasogastric tube.

DISCUSSION

Forestier's disease, or DISH, is a rare ossifying pathology characterised by spinal and peripheral enthesopathy (6). The condition shows a male predilection and an increasing prevalence with age. Its incidence is estimated to be between 8-10% among individuals aged 65 years and over. It is rarely diagnosed in those under 45 years of age (7). Contemporary studies suggest potential risk factors for DISH include high body mass index, insulin-independent diabetes mellitus, acromegaly, obesity, hypervitaminosis A, human leukocyte antigen (HLA)-B27, HLA-B5, HLA-A11, and ankylosing spondylitis (6, 8).

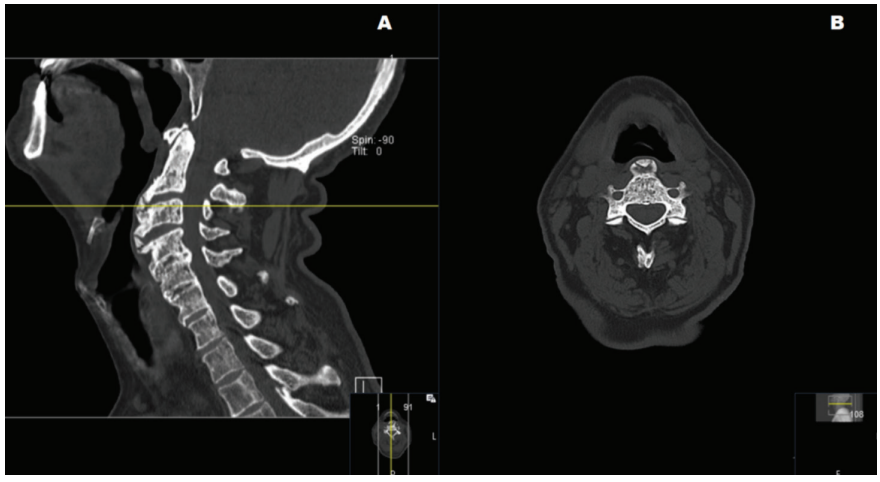

Figure 1: Preoperative CT images, (A) sagittal reconstruction (B) axial scan.
CT: Computed tomography

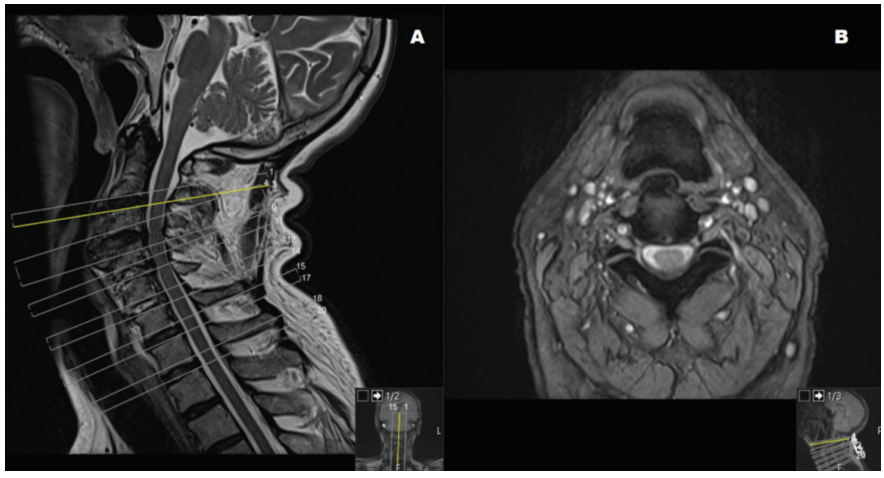

Figure 2: Preoperative MR images (A) sagittal reconstruction (B) axial scan.
MR: Magnetic resonance

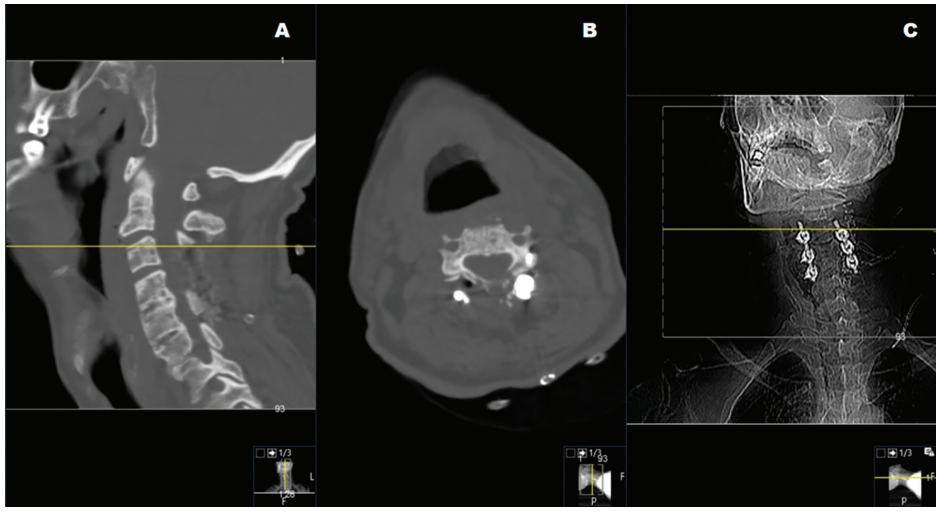

Figure 3: Preoperative CT images, (A) sagittal reconstruction, (B) axial scan, (C) anteroposterior scout view.
CT: Computed tomography

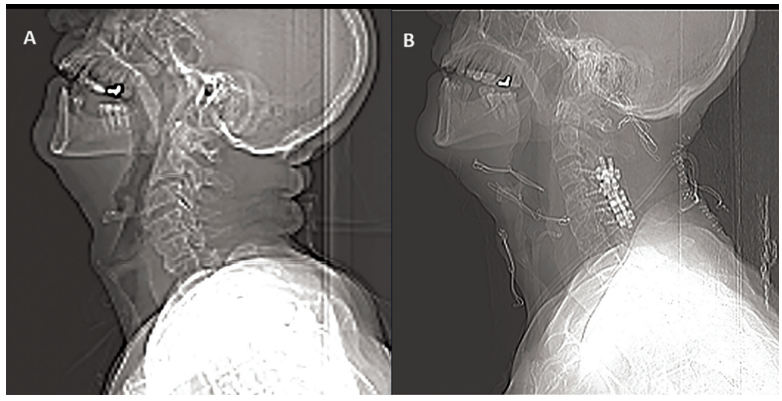

Figure 4: Preoperative (A) and postoperative (B) CT lateral scout views.
CT: Computed tomography

Common sites of DISH involvement are the spine, pelvis, patella, and calcaneus, with the sacroiliac joint being an exception (6). The thoracic region is almost universally affected (96%), while the lumbar (90%) and cervical regions (78%) are involved less frequently (7).

Clinical studies report that 17-28% of individuals with DISH display symptoms of dysphagia attributable to cervical osteophytes (2). Other common symptoms include neurovascular compression, movement restriction, cervicalgia, stridor, and hoarseness (6).

Diagnosis of DISH is typically confirmed through radiological assessments. Three radiological classification criteria exist: (I) Calcification and bone bridge formation in at least two contiguous vertebrae, (II) Preservation of intervertebral disc height, and (III) Absence of ankylosis in interapophyseal joints (4, 6). CT is the diagnostic tool of choice, with sagittal, coronal, and three-dimensional reconstructions providing an elaborate perspective of the osteophytes' relationship with adjacent anatomical structures (6).

Conservative treatment options are typically recommended for symptomatic DISH cases with non-severe clinical findings. These encompass non-steroidal anti-inflammatory drugs, steroids, muscle relaxants, dietary modifications, and anti-reflux treatments. Patients' refractory to conservative treatment may necessitate surgical intervention. Surgical resection of osteophytes has been recognised as an effective treatment modality in severe cases, particularly those presenting with airway obstruction. Anterior cervical, posterolateral, and transpharyngeal approaches are potential surgical methods, depending on individual patient characteristics and disease severity (6, 7).

No early postoperative complication was present in the presented case. Nevertheless, these procedures are prone to some complications, including surgical site hematomas, development of Horner's syndrome, paralysis of recurrent or superior laryngeal nerves, dysphonia, oesophagal rupture, or spinal instability (8). Unfortunately, late complications, such as aspiration, lead to an extended hospital stay along with a late and limited recovery.

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REFERENCES

1. Aydin E, Akdogan V, Akkuzu B et al. Six cases of Forestier syndrome, A rare cause of dysphagia. Acta Otolaryngol 2006;126(7):775-8. [Crossref]
2. Legaye J. Forestier's syndrome: a rare cause of dysphagia. a case report and review of the literature. Acta Orthop Belg 2020;86(2):216-9. https://pubmed.ncbi.nlm.nih.gov/33418609/ [Crossref]
3. Zárate-Kalfópulos B, Jerez-Saldaña O, Romero-Vargas S et al. Enfermedad de Forestier. Reporte de un caso y revisión bibliográfica [Forestier disease. Case report and literature review]. Cir Cir 2012;80(5):451-4.. Cir Cir 2012;80(5):451-4. [Crossref]
4. Kuperus JS, Mohamed Hoesein FAA, de Jong PA et al. Diffuse idiopathic skeletal hyperostosis: etiology and clinical relevance. Best Pract Res Clin Rheumatol 2020;34(3):101527. [Crossref]
5. Childs SG. Diffuse idiopathic skeletal hyperostosis: Forestier's disease. Orthop Nurs 2004;23(6):375-82. [Crossref]
6. Mader R. Diffuse idiopathic skeletal hyperostosis: time for a change. J Rheumatol 2008;35(3):377-9. [Crossref]
7. Karaarslan N, Gürbüz MS, Çalışkan T et al. Forestier syndrome presenting with dysphagia: case report of a rare presentation. J Spine Surg 2017;3(4):723-6. [Crossref]
8. Soares D, Bernardes F, Silva M et al. Diffuse idiopathic skeletal hyperostosis (DISH)-phagia: a case report and review of literature of a rare disease manifestation. Cureus 2023;15(10):e47221. [Crossref]