

## Image Report

## Clinical and radiological features of Forestier's disease presenting with dysphagia

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### Abstract

**Background:** Diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier's disease, is a rheumatologic condition characterized by ossification of the spinal ligaments and tendons. Large anterior osteophytes are typically present in the lower cervical levels, while upper cervical ossification resulting in dysphagia is very rare.

**Methods:** Here, we presented a patient with Forestier's disease involving massive ossification of the anterior longitudinal ligament extending from C3 to C4 downward contributing to severe dysphagia.

**Results:** A 65-year-old male presented with cervical pain and dysphagia. The computed tomography of the cervical spine demonstrated massive anterior longitudinal ligament ossification (DISH) extending from C3 to C7. There was an additional large osteophyte at the C3-C4 level, and also a high-grade intracanalicular C6-C7 cervical stenosis due to ossification of the posterior longitudinal ligament. The patient was offered surgical intervention (e.g., resection of the C3-C7 anterior DISH and anterior cervical discectomy/fusion at the C6-C7 level), but he declined.

**Conclusions:** When conservative management fails to resolve severe dysphagia for cervical DISH/Forestier's disease, anterior surgical resection is typically performed. In this case, the patient refused surgery and opted for conservative management strategies.

**Key Word:** Cervical spine, diffuse idiopathic skeletal hyperostosis, Forestier's disease, non-surgical options

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## INTRODUCTION

Diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier's disease, is a rheumatologic condition characterized by ossification of the spinal ligaments and tendons. It occurs in up to 30% of the population, but is mostly seen in patients between the ages of 60 and 80.<sup>[1,2]</sup>

Cervical DISH usually appears as large anterior longitudinal ligament osteophytes involving the lower cervical spinal canal; upper cervical DISH resulting in dysphagia is very rare. Here we presented a patient who exhibited C3-C7 anterior cervical DISH with dysphagia, accompanied by C6-C7 intracanalicular cord compression due to ossification of the posterior longitudinal ligament.

## METHODS

### Clinical history

A 65-year-old male presented with cervical pain and dysphagia (e.g., inability to eat solid food for the past year). The neurological examination revealed no motor or sensory deficits; the patient only exhibited diffuse hyperreflexia to the upper and lower extremities, without Babinski responses.

### Radiographic findings

The computed tomography of the cervical spine demonstrated massive anterior longitudinal ligament ossification extending from C3 to C7 (DISH); there was an additional large osteophyte at C3-C4, and high-grade C6-C7 intracanalicular cervical stenosis due to ossification of the posterior longitudinal ligament [Figures 1–4]. The magnetic resonance imaging study confirmed these lesions (e.g., showing displacement of the larynx and trachea anteriorly, and a kinked esophagus) [Figure 5]. Based on these findings, the patient was diagnosed cervical DISH, also known as Forestier's disease.

### Refusal of surgery

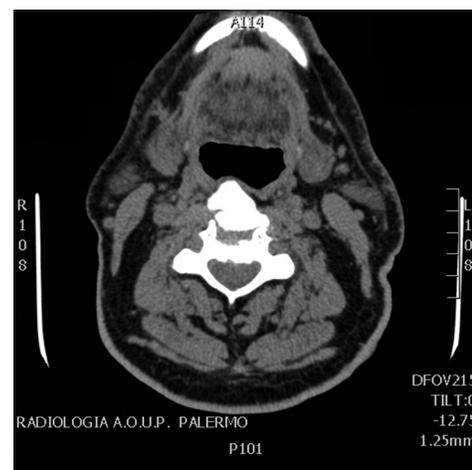
Despite the dysphagia attributed to C3-C7 DISH and C3-C4 osteophyte, along with C6-C7 ossification of the posterior longitudinal ligament (OPLL), the patient refused surgery. Rather, the patient opted for conservative management, and was discharged on steroids and postural therapy.

## DISCUSSION

The diagnosis of Forestier's disease or DISH is primarily radiological; its etiology is still unknown.<sup>[3]</sup> Three radiologic criteria are utilized to diagnose DISH: continuous ossification of the anterior longitudinal ligament involving at least four contiguous vertebral



**Figure 1:** Sagittal multiple planar reconstruction (MPR) computed tomography scan showing C3-C4 anterior ossification with large osteophytes. White arrow: trachea kinking on the superior margin of the large anterior osteophyte



**Figure 2:** Axial computed tomography scan showing upper C3-C4 anterior ossification with posterior impression on the dorsal surface of the trachea and its initial displacement



**Figure 3:** Axial computed tomography scan showing C6-C7 high-grade stenosis with calcification of the intervertebral disc and posterior osteophyte



**Figure 4:** Three-dimensional volume rendering showing the cervical spine with anterior ossification

bodies, lack of intervertebral ankyloses or fusion, and preservation of intervertebral disk height.<sup>[8]</sup> DISH typically involves the lower cervical levels, and only rarely is seen in the upper cervical spine. Often DISH is asymptomatic, and is just incidentally diagnosed on cervical computed tomography and X-ray examinations.<sup>[9]</sup> However, some cases of massive DISH cause dysphagia (up to 6%), dysphonia, and dyspnea.<sup>[1,2,5,9,10]</sup> Dysphagia is usually attributed to DISH involving ossification of the anterior longitudinal ligament with ventral osteophytes; this is variously attributed to an inflammatory process, neuropathy, and limitation of pharyngolaryngeal motion. Here, the massive C3-C7 DISH combined with the large cervical osteophyte at C3-C4, near the anatomical anchorage of the esophagus, likely explained this patient's severe dysphagia.

### Treatment of diffuse idiopathic skeletal hyperostosis

The initial treatment of cervical DISH is typically conservative therapy, for example, the use of anti-inflammatory medication, steroids, muscle relaxants, and postural education to optimize swallowing.<sup>[10]</sup> Surgery should be considered when conservative management fails and/or in case of severe dysphagia. Common surgical management includes anterior cervical osteophylectomy alone.<sup>[11]</sup> However, subsequent anterior cervical fusion may be necessary if instability results.<sup>[1,4,6,7,9]</sup> In the case presented, the patient refused surgery, and was discharged on steroids to pursue postural therapy to facilitate swallowing.



**Figure 5:** Magnetic resonance imaging showing the displacement of larynx and trachea and the kinking of the esophagus caused by C3-C4 anterior osteophyte. White arrow: trachea kinking on the superior margin of the large anterior osteophyte

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### Conflicts of interest

There are no conflicts of interest.

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