

CASE REPORT

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## Diffuse Idiopathic Skeletal Hyperostosis (DISH)—A Rare Etiology of Dysphagia

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**Abstract:** A 72-year-old gentleman presented to the hospital with progressively worsening dysphagia to soft foods and liquids. He was diagnosed with severe pharyngeal dysphagia by modified barium swallow. A CT scan of the neck with IV contrast showed anterior flowing of bridging osteophytes from C3-C6, indicative of DISH, resulting in esophageal impingement. He underwent resection of the DISH segments. Following the surgery, a PEG tube for nutrition supplementation was placed. However, the PEG tube was removed after five months when the speech and swallow evaluation showed no residual dysphagia. DISH is a rare non-inflammatory condition that results in pathological ossification and calcification of the anterolateral spinal ligaments.

**Keywords:** DISH, dysphagia, diffuse idiopathic skeletal hyperostosis

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## Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) with cervical involvement causing dysphagia is a rare entity. It was described as an abnormal ossification along the anterior and lateral aspects of the vertebral column in 1950 by Forestier and Rotes-Querol.<sup>1</sup> We report a case of a patient with progressive dysphagia secondary to DISH.

## Case Presentation

A 72-year-old gentleman with coronary artery disease, atrial fibrillation, hypertension, gout, and obstructive sleep apnea presented to the hospital with progressively worsening dysphagia to soft foods and liquids for a few days. He had begun to experience dysphagia five years ago and had restricted his diet to mainly soft foods and liquids. He denied fatigue, neck pain, drooping of eye lids, difficulty in breathing, hoarseness, dysarthria, numbness or limb weakness. Surgical history was significant for bilateral cataract surgeries and childhood exploratory laparotomy after a traumatic incident. He denied a family history of cancer. His medications included warfarin, digoxin, atenolol, lovastatin, colchicine, hydrochlorothiazide, omeprazole, albuterol and ipratropium inhalers. He used to smoke 1 pack per day for 40 years, but quit 20 years ago. Esophagogastroduodenoscopy and colonoscopy were done three years ago and were unremarkable. Vital signs were stable and the physical exam was unremarkable. Laboratory tests, including an autoimmune panel were normal. CT scan of the head was normal. A speech and swallow evaluation revealed dysphagia to thin liquids. A modified barium swallow (MBS) revealed severe pharyngeal dysphagia. CT of the neck with IV contrast showed anterior flowing of bridging osteophytes from C3-C6, indicative of DISH, causing esophageal impingement (Fig. 1). He also had a MRI of the neck with gadolinium that showed anterior flowing of bridging osteophytes from C3-C6 consistent with diffuse idiopathic skeletal hyperostosis, C3-C6 spinal stenosis and degenerative changes at C3-C4 (Fig. 2). He underwent corpectomy of C4 with C3-C5 spinal fusion and resection of the C3-C6 DISH segments. Although post-operative radiological studies showed an anatomical improvement, an esophagram revealed residual dysphagia (Fig. 3).

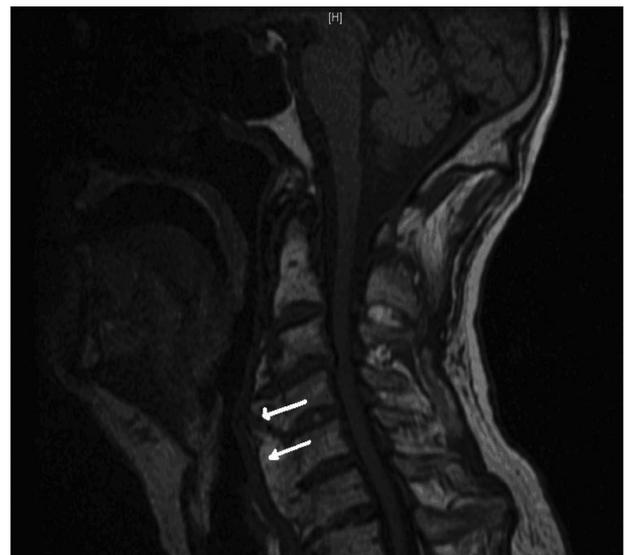


**Figure 1.** CT of neck with IV contrast (bone window) showing anterior flowing of bridging osteophytes (arrows) from C3-C6, indicative of DISH.

He was discharged with a percutaneous endoscopic gastrostomy (PEG) tube. Five months later, he underwent speech and swallow evaluation, which showed marked functional improvement and the PEG tube was removed.

## Discussion

Diffuse idiopathic skeletal hyperostosis, also known as Forestier's disease,<sup>2</sup> is a non-inflammatory condition



**Figure 2.** MRI of neck with gadolinium showing anterior flowing of bridging osteophytes (arrows) from C3-C6 consistent with DISH, C3-C6 spinal stenosis and degenerative changes at C3-C4.



**Figure 3.** Post-operative esophagram showing residual dysphagia (arrow) [fluid entering esophagus and trachea] and anatomical correction of osteophytes.

with pathological ossification and calcification of the anterolateral spinal ligaments and attachment sites of the tendons and ligaments. Ossification along the posterior vertebral border has also been noted.<sup>3</sup> While etiology of DISH remains unknown, various hypotheses were proposed relating to mechanical factors, diet, metabolic conditions and environmental exposures.<sup>4</sup> Specifically, mechanical factors leading to ligament stretching, high fluoride levels in water, excess of vitamin A, long term isotretinoin therapy, and metabolic conditions that produce increased insulin-like growth factor-1 to stimulate osteoblasts have been documented as potential causative factors.<sup>5</sup> While some confirmation came from clinical studies, there were conflicting results reported as well. The pathogenesis of DISH has been explained as an enthesopathy due to pathological osteoblast differentiation and ossification.<sup>6</sup> Prevalence is higher in men (typically in their 60s) than women.<sup>1</sup> The incidence increases with age and varies by population.

Symptoms of DISH depend on the localization and involvement of adjacent structures. Patients may experience neck, thoracic spine, lumbar spine and/or extremity pain and disability. Spinal morning

stiffness is experienced by the majority of patients. Dysphagia and symptoms pertaining to the knee, shoulder and elbow are less common. Dysphagia, odynophagia, foreign body sensation, stridor, obstructive sleep apnea, and aspiration of solids and liquids may occur when large anterior osteophytes of the cervical spine impinge or displace the adjacent structures of the gastrointestinal and respiratory tracts.<sup>7</sup> Dysphagia is usually progressive and greater for solids than liquids. The osteophytes arising from the C5-C6 level frequently cause dysphagia, followed by C4-C5.<sup>8</sup> Stridor may be a rare manifestation of the large C2-C3 anterior osteophytes, while point pressure between osteophytes and the posterior cricoid cartilage results in hypopharyngeal ulceration causing odynophagia.<sup>9</sup> Spinal cord compression can happen in patients with posterior ligament ossification and this may lead to myelopathy and radiculopathy.<sup>10</sup> All these symptoms can manifest due to mechanical obstruction, inflammation in the immediate surrounding of the osteophytes or spasm due to pain. Presumably, a summation of all these factors plays a role in producing the symptoms. Physical findings include decrease range of spinal motion, particularly when the thoracic segment is involved, and palpable nodules when the appendicular skeleton is involved. Neurological findings may be elicited when posterior ligament calcification involves the spinal cord. Differential diagnoses for DISH include Ankylosing spondylitis (AS) and Spondylosis deformans (SD). There are spurs seen in the region of the cervical and lumbar spine similar to DISH but involvement of the anterior longitudinal ligament in the thoracic region is not seen in SD. AS is more common in males than females and there is an association with ligamentous ossification and syndesmophytes similar to DISH but the anterior longitudinal ligament is not involved and the bony bridges involve the outer margin of the annulus fibrosis. In addition, erosions and bony ankylosis of the sacroiliac and apophyseal joints are seen in AS. Radiological imaging of the spine will show squaring of the vertebrae also known as “bamboo spine.” Other conditions that may be associated with DISH include gout,<sup>11</sup> rheumatoid arthritis,<sup>12</sup> Paget’s disease<sup>13</sup> and chondrocalcinosis.<sup>14</sup> Evaluation of progressive dysphagia should include speech and swallow evaluation,



barium swallow studies, motility studies, otolaryngologic examination, and other radiological studies, which include CT and MRI of the head and neck. Endoscopy should be utilized with caution as this may cause perforation of esophagus.<sup>15</sup> An emphasis is placed on diagnostic imaging to diagnose DISH. The radiological findings of DISH may occur in the absence of clinical symptoms. Resnick and Utsinger proposed radiographic criteria for the diagnosis of DISH.<sup>5</sup> The Resnick radiographic criteria for the diagnosis of DISH include: (a) the presence of flowing ossification and calcification along the anterolateral aspect of at least four contiguous vertebral bodies, (b) the preservation of intervertebral disk heights in the involved vertebrae, (c) the absence of apophyseal joint bony ankylosis and sacroiliac joint erosion.<sup>16</sup> Extra-spinal ossification in DISH may occur at ligamentous attachments and para-articular soft tissues.<sup>17</sup> There are no specific laboratory abnormalities that have been associated with DISH.

Treatment of DISH depends on the presence of symptoms. Physical activity and physical therapy with heat can give symptomatic relief in patients with DISH. The pain associated with DISH is treated similar to patients with osteoarthritis. Dysphagia, radiculopathy, myelopathy, recurrent laryngeal nerve palsy, and stridor need surgical intervention.<sup>11</sup>

## Conclusion

DISH is an uncommon etiology of dysphagia, which is a common symptom. There have been other case reports that have described dysphagia as a manifestation of DISH.<sup>18,19</sup> Although a rare cause of dysphagia, DISH can cause considerable morbidity in the geriatric population. Keeping this clinical entity in the differential diagnosis is important in arriving at the diagnosis.

## Disclosures

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If this article contains identifiable human subject(s) author(s) were required to supply signed patient consent prior to publication. Author(s) have confirmed that the published article is unique and not under consideration nor published by any other publication and that they have consent to reproduce any copyrighted material. The peer reviewers declared no conflicts of interest.

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