

Dysphonia, Stridor, and Dysphagia Caused By Diffuse Idiopathic Skeletal Hyperostosis: Case Report and Review of Literature

Abstract

Diffuse idiopathic skeletal hyperostosis (DISH) also known as Forestier disease is a noninflammatory, systemic skeletal disease of unknown etiology. DISH is usually asymptomatic but may compress the posterior wall of the aero digestive tract and lead to dysphagia, globus, hoarseness, stridor, dyspnea, and neurological problems. Although dysphagia is not uncommon among the presenting symptoms of DISH but dysphonia and stridor are rarely reported. We report a 68-year-old man who presented with a history of progressive dysphagia over 1 year and recent dysphonia and stridor secondary to cervical osteophytes. We discuss the symptoms, radiological features, and management of this uncommon case of DISH in conjunction with review of literature.

Keywords: *Dysphagia, diffuse idiopathic skeletal hyperostosis, forestier disease, hyperostosis, stridor*

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Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) also known as Forestier disease, senile ankylosing hyperostosis, and ankylosing vertebral hyperostosis, is a noninflammatory, systemic skeletal disease of unknown etiology. DISH is a common but underdiagnosed disease that usually observed in elderly people and has been reported to affect up to 10% of patients over 65 years of age.^[1] DISH leads to ossification of ligaments and entheses of the spine and peripheral skeleton. The diagnosis is primarily radiological characterized by the ossification of the anterior longitudinal ligament, with osteophytes formation along the spinal column at least four contiguous vertebral bodies, a minimal degree of degenerative disc disease; and absence of apophyseal joint ankylosis and sacroiliac joint fusion, erosions, or sclerosis.^[2] Osteophytes accompanying degenerative disease of the cervical spine, and ankylosing spondylitis are the most common pathological entities that can be confused with DISH.

DISH is usually asymptomatic but may compress the posterior wall of the aero digestive tract and lead to dysphagia, globus, hoarseness, stridor, dyspnea,

and neurological problems. Cervical hyperostosis in patients with DISH is responsible for 17%–28% incidence of dysphagia, and about 8% of patients failed to respond to conservative treatment and surgical excision through an anterior cervical approach was required.^[2]

Although dysphagia is not uncommon among the presenting symptoms of DISH, dysphonia and stridor are rarely reported. We herein report a case of DISH causing dysphagia, dysphonia, and stridor and also discuss the clinical and imaging features, surgical management, and outcomes in conjunction with the literature review.

Case Report

Presentation

A 68-year-old man who presented with a history of progressive dysphagia over 1 year and recent dysphonia and stridor. In examination, cervical spine range of motion was decreased in extension and flexion but he had no pain during movement and no tenderness over the spinous processes. Neurologic examinations were normal. Deflection of barium and a narrowed esophagus at C3-6 was observed in Video-fluoroscopy. X-ray, computed tomography (CT) and magnetic resonance

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imaging (MRI) [Figure 1] of the C-spine demonstrated multi-level anterior cervical osteophytes at C3-T1 without disc degeneration, the largest measuring 23 mm at the level of C3 and C4 [Figure 2], consistent with a diagnosis of DISH. Compression effect of osteophytes with no other abnormalities was found on endoscopic examination. Sacroiliac joints and apophyseal joints were intact laboratory tests were normal and MRI did not reveal soft-tissue mass or abnormal signal change. Therefore, the diagnosis of ankylosing spondylitis, infection, and malignancy were rule out. Imaging and other studies suggested that osteophytes were the cause of these symptoms and given the progression of the symptoms, excision of osteophytes performed through an anterior lateral approach.

Surgical technique

The patient was placed in the supine position with neck extension. We used Smith Robinson approach with longitudinal skin incision at the left side and satisfactory access from C2 to C7-T1 was achieved. The dissection was medial to the carotid sheath and lateral to the tracheoesophageal tract. By retracting the strap muscles and pharynx, the osteophyte was easily palpable. The prevertebral fascia was divided and the lateral borders of the osteophytes exposed by elevating the longus colli. Large osteophytes were clearly visible and resected from C3 to T1 using an osteotome to make a smooth surface. To avoid postoperative hematoma, bleeding from the cancellous bone was stopped with bone wax. Intraoperative lateral fluoroscopic view was taken to ensure all relevant levels had been successfully decompressed. The wound was closed with 2.0 vicryl to platysma and 3.0 monocryl to skin drain was removed after 48 h. The operation time was 83 min, and the blood loss was about 150 ml.

Follow-up

The patient was mobilized the day after the surgery. Postoperative lateral cervical radiograph [Figure 3] showed a significant resolution of osteophytes and CT scan showed significant reduction in laryngeal inlet obstruction. The patient reported significant improvement in the subjective symptoms of stridor, neck motion, and dysphonia soon after surgery but dysphagia did not recover immediately and after 3-month follow-up, he was able to tolerate a regular diet. During follow-up (26 months), the recurrence of symptoms or osteophytes on imaging was not observed.

Discussion

DISH is the second-most common cause of ventral cervical osteophytes after degenerative conditions, with a prevalence of 2%–4% in patients older than 40 years and increases up to 28% in those older than 80 years. The pathogenesis of disease is poorly understood. Several studies investigated association between DISH and trauma, endocrine, and metabolic disease but the strongest correlation observed with sex and age.^[3,4]

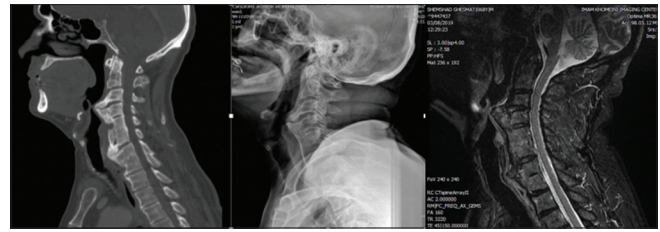


Figure 1: X-ray, computed tomography and magnetic resonance imaging of the patients' cervical spine

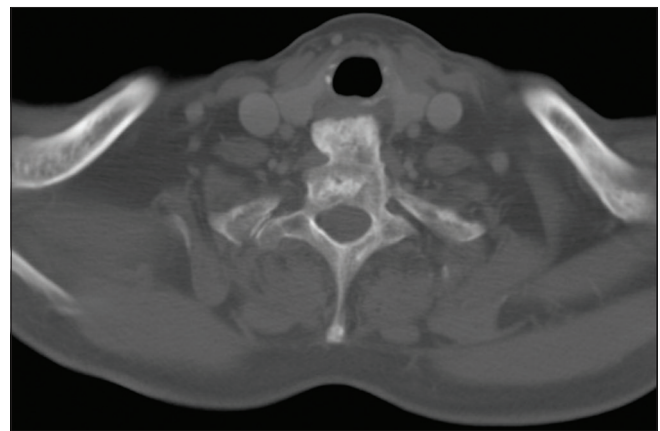


Figure 2: Axial view of cervical computed tomography scan demonstrated anterior osteophyte height



Figure 3: Postoperative lateral cervical spine X-ray

Although DISH is generally asymptomatic, symptoms due to cervical ossification can be even life-threatening. Pain and stiffness are the most frequent symptoms. Six percent to twenty-eight percent of patients complain of dysphagia due to external compression to the aerodigestive tract and associated nerves, inflammatory edema as a result of repetitive mechanical trauma, fibrosis, adhesion formation, and muscle spasm with 8%–10% requiring surgical treatment. Globus, choking, bronchial aspiration, dysphonia, hoarseness, stridor, dyspnea, and more exceptionally respiratory compromise due to compression of the upper respiratory airway are other symptoms related to DISH.^[5]

Dysphonia as the presenting symptom of DISH has rarely been reported. Mechanical obstruction of the larynx, reduction in glottal mobility due to inflammation and recurrent laryngeal nerve paralysis because of compression of the osteophyte are suggested cause of dysphonia.^[6] Stridor caused by cervical osteophytes are an uncommon entity. Myelopathy and quadriplegia have been reported in patients with DISH who had also ossification of posterior longitudinal ligament.^[7] Nelson *et al.*, in a retrospective review in 134 patients with DISH reported 3.8% of the patients presented with dysphagia and three patients required tracheotomy for acute airway obstruction.^[8]

Symptomatic cases can be managed with diet modifications, nonsteroid anti-inflammatory drugs, steroids, anti-reflux medications, postural changes, speech and swallow therapy, and muscular relaxants noninvasive management of respiratory compromise often fails and may require emergency tracheotomy until surgical excision perform. Surgical intervention is reserved for Symptomatic cases that do not response to nonoperative treatment, progressive dysphagia with unintentional weight loss and those with respiratory complaints.

Although anterior osteophytes is commonly found in the older population and is usually asymptomatic, but can be a source of considerable morbidity and potential life-threatening airway obstruction. Recognizing this clinical entity is imperative in establishing the diagnosis and initiating appropriate treatment.

C5 to C6 are the most common vertebrae involved in 40% of cases followed by C4 to C5. The level of osteophytes has been suggested to correlate with presenting symptoms. Lower cervical vertebrae osteophytes at the levels of C4 to C6 can impinge on the esophagus and the upper cervical spine osteophytes can impinge more on the oropharynx, resulting respiratory compromise, and stridor. Osteophytes larger than 12 mm can become symptomatic. Strasser *et al.* showed a correlation between the size of the osteophytes (>10 mm) and the possibility of aspiration by video fluoroscopy,^[9] although some studies revealed that the size of the osteophytes did not related to the severity of the symptoms.

A significant improvement in symptoms was more frequently seen with surgical treatment than with conservative treatment.^[4] Osteophyte resection appears to be effective in surgical treatment of DISH, although complete resolution of symptoms can take many months, particularly in those with airway obstruction.^[1] It has been reported that about 10% of patients with DISH-related dysphagia require surgical decompression. Airway obstruction and dysphonia caused by a combination of impinging osteophytes, aspiration, and reflux lead to inflammatory response followed by tissue fibrosis and neurologic damage. The prognosis for the restoration of these symptoms remains unclear. Improvement of symptoms could be in the

immediate postoperative period^[10,11] or in the following 3–6 months.^[3,12] Urrutia *et al.* reported significant improvement in dysphagia 2 weeks after surgery.^[11] Patients usually demonstrate significant symptomatic improvement within 3 months' postoperatively but some studies reported that several months may be required for the symptoms to relieve.

Indication for vertebral fusion to avoid re-operating and the rate of recurrence are not fully explained. Miyamoto *et al.*^[12] found a 100% radiologic and 29% clinical recurrence rates after anterior decompression without fusion. Osteophytectomy alone might be a good option in elderly patients but regarding the recurrence of dysphagia for patients <70 years old, simple resection without fusion will be an issue. von der Hoeh *et al.*^[10] showed that when anterior arthrodesis added to osteophytectomy, no recurrence was occurred, whereas other studies reported no recurrence with osteophytectomy alone.^[13] Some studies reported that instrumented fusion is not warranted,^[14] whereas others suggested fusion when instability is apparent.^[15]

Conclusion

DISH should be considered in differential diagnosis of the patients with dysphagia, upper airway symptoms, and dysphonia. Endoscopic examination and imaging techniques include lateral cervical X-ray and CT scan, barium swallow, and MRI can help to reach a correct diagnosis. Airway obstruction and dysphonia caused by DISH rarely reported but in order to avoid delay in diagnosis and treatment, physicians especially gastroenterologists, ENT, and spine surgeons should be aware of DISH as a potential cause of these symptoms. Resection of anterior cervical osteophytes is considered to be highly effective in symptomatic patients.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form the patient(s) has/have given his/her/their consent for his/her/their images and other clinical information to be reported in the journal. The patients understand that their names and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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