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Case Report

Progressive dysphagia and dysphonia secondary to DISH-related anterior cervical osteophytes: A case report

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ABSTRACT

Background: Dysphagia due to diffuse idiopathic skeletal hyperostosis (DISH)-related anterior cervical osteophytes is not uncommon. However, this rarely leads to dysphonia and/or dysphagia along with lifethreatening airway obstruction requiring emergency tracheotomy.

Case Description: A 56-year-old male presented with progressive dysphagia and dysphonia secondary to DISH-related anterior osteophytes at the C3-C4 and C4-C5 levels. The barium swallow, X-ray, magnetic resonance imaging, and computed tomography scans confirmed the presence of DISH. Utilizing an anterior cervical approach, a large beak-like osteophyte was successfully removed, while preserving the anterior annulus. After clinic-radiological improvement, the patient was discharged with a soft cervical collar and nonsteroidal anti-inflammatory drug (NSAID).

Conclusion: Large anterior osteophytes in Forestier disease/DISH may cause dysphagia and dysphonia. Direct anterior resection of these lesions yields excellent results as long as other etiologies for such symptoms have been ruled out.

Keywords: Anterior cervical osteophyte, Diffuse idiopathic skeletal hyperostosis, Dysphagia, Dysphonia

INTRODUCTION

Diffuse idiopathic skeletal hyperostosis also known as Forestier's disease is a common degenerative enthesopathic disorder of unknown etiology. It is characterized by ossification of the longitudinal ligaments, especially the anterior longitudinal ligaments (ALL) in the spine. [1] DISH occurs in 12-30% of patients and is more common in males over 65 years of age. Some patients may present with large symptomatic anterior cervical osteophytes that cause dysphagia and dysphonia. Rarely, DISH leads to aspiration pneumonia, stridor, dyspnea, or airway obstruction requiring emergency tracheotomy. [4] Obstruction, when it occurs, most often involves the C5-C6, followed by C4–C5, C2–C3, and C3–C4 levels; it is least common at C1–C2.[7]

In this case, a 56-year-old male presented with a large symptomatic anterior osteophyte/DISH lesion at the C4–C5 level causing progressive dysphagia and dysphonia.

CASE REPORT

A 56-year-old male presented with the 1-year onset of progressive dysphagia and 8 months of dysphonia. Notably, his neurological examination was intact. The barium swallow showed

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a narrowed esophageal lumen at the C4-C5 level [Figure 1]. X-ray and computed tomography studies revealed the classical "Ostrich beak" changes of the ossified ALL at the C4-C5 level; the disc spaces were preserved [Figures 2 and 3]. Utilizing an anterior cervical approach, the large beak-like osteophyte was removed, while preserving the anterior annulus; the extent and adequacy of resection was confirmed intraoperatively utilizing a lateral radiograph [Figures 4 and 5]. Postoperatively, he was discharged on antiinflammatory medications for 10 days.

DISCUSSION

Dysphagia due to anterior cervical osteophytes is rare.^[5] However, even more unusual is the onset of both dysphagia and dysphonia due to anterior cervical DISH. Isolated dysphagia is present in nearly 75% of the cases, along with less frequent; dyspnea (14%), symptoms/signs of aspiration



Figure 1: Barium swallow test showing narrowing of esophageal lumen at C4-C5 levels.



Figure 2: Diffuse idiopathic skeletal hyperostosis-related large anterior osteophytes at C4-C5 and C3-C4 compressing trachea and esophagus from behind.

(9%), stridor (3%), cervical pain (3%), cough (3%), and dysphonia (2%).[4]



Figure 3: Computed tomography axial cut showing large anterior osteophyte at C4-C5 level impinging on the esophagus and trachea.



Figure 4: Large osteophyte that was removed in piece meal.



Figure 5: Postoperative lateral view of cervical spine X-ray.

Imaging of DISH to rule out other causes of dysphagia/ dysphonia

Besides preoperative images (e.g., X-ray, computed tomography, and magnetic resonance imaging), esophageal and laryngoscopic examinations combined with fluoroscopic barium swallowing tests are essential to both establish the diagnosis of DISH contributing to dysphagia/dysphonia and to provide a clear guide for treatment.

Surgery

Patients who fail conservative measures typically become surgical candidates. Surgery should consist of direct anterior removal of osteophytes/DISH, with preservation of the anterior annulus.[3] Such surgical resection of osteophytes usually results in excellent symptomatic relief. Fusion may occasionally be warranted if there are accompanying signs of instability.[6]

Recurrence of DISH

Recurrence of cervical DISH is rare, but may develop slowly and progressively at the average rate of 1 mm/year. In addition, it may be associated with segmental instability at the index operated level.[2]

Role of nonsteroidal anti-inflammatory drugs (NSAIDs)

After total hip replacement, NSAIDs are typically utilized postoperatively to prevent the recurrence of DISH osteophytes. However, the utility of NSAIDs following cervical spine surgery for DISH is not as well understood. Ruetten et al. recommend the postoperative use of indomethacin 50 mg twice daily for 10 days with gastric protection, plus fractionated radiation with five doses of 2 Gray (4 days interval) if not contraindicated with patients over 55 years of age.

Postoperative bracing

The postoperative use of a soft cervical collar for 6 weeks combined with speech therapy was found to improve clinical outcomes in some patients undergoing anterior cervical DISH surgery. The role of bisphosphonate etidronate disodium in the prevention of ligament ossification/DISH in animal tests has not yet become standard in humans.[8]

CONCLUSION

Large cervical anterior osteophytes in Forestier disease (DISH) rarely cause dysphagia and dysphonia. Here, direct surgical resection of the anterior osteophytes/DISH produced an excellent result.

Declaration of patient consent

Patient's consent not required as patients identity is not disclosed or compromised.

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Conflicts of interest

There are no conflicts of interest.

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