Bilateral Vocal Fold Paralysis With Airway Obstruction And Dysphagia Secondary To Diffuse Idiopathic Skeletal Hyperostosis: A Case Report

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Introduction

Diffuse idiopathic skeletal hyperostosis (DISH), or Forestier’s disease, is an idiopathic spinal disease common in patients beyond the sixth decade of life. The disease is characterized by flowing ossification of anterolateral vertebral ligaments and osteophyte formation along the spinal column. In most patients it is asymptomatic; symptoms that do arise are dependent on the location of disease. Although less common than thoracic level disease, cervical hyperostosis is capable of producing symptoms frequently encountered in otolaryngology practice, including dysphagia, stridor and airway obstruction. Reports of vocal cord paralysis caused by DISH are exceedingly rare. We present a case of progressive dysphagia, airway obstruction, and bilateral vocal cord paralysis caused by DISH and reversed with surgical osteophytectomy. Consideration of DISH in clinical practice and treatment options are discussed.

Case Report

A 61 year-old male with diabetes presented with 3 months progressive hoarseness, stridor and respiratory distress along with dysphagia and 30 lb weight loss. This resulted in tracheostomy tube placement 1 month prior followed by PEG tube placement. Perceptual vocal exam revealed mild dysphonia and range reduction. Flexible laryngoscopy showed bilateral vocal folds immobile in a paramedian position with a 3 mm posterior glottic opening and bulging of the posterior pharyngeal wall without edema.

Barium swallow demonstrated aspiration of thin fluids, and cervical radiograph revealed ossification of the C2-C5 anterior longitudinal ligament with preservation of intervertebral disc height. Cervical CT demonstrated massive midline anterior cervical osteophyte growth from the skull base to the C7 level. Cervical MRI confirmed anterior ossification. No sacroiliac erosion was noted on lumbosacral radiograph.

Swallowing exercises and a soft diet were implemented, however the patient remained dependent on tube feedings and exhibited persistent airway distress with tracheostomy capping. In the context of endogastric tube dependence and refractory dysphagia, osteophytectomy was performed. In addition, elective direct microlaryngoscopy was performed to investigate sources of vocal fold immobility. Palpation of the arytenoid cartilages revealed bilateral mobility with respect to the cricoarytenoid joint. Osteophytectomy via a left anterolateral approach was completed by the otolaryngology service followed by resection of osteophytes on levels C4-C7 by the neurosurgical team without complication or effect on spinal stability.

One day following surgery the patient demonstrated swallowing without difficulty and was discharged with the tracheostomy in place as vocal fold abduction remained limited. He consistently tolerated an oral diet and the endogastric tube was removed 3 weeks post-operation. Surprisingly, vocal fold motion gradually improved over the course of 5 weeks of follow-up. After 5 weeks tracheostomy occlusion resulted in no stridor, and the patient was decannulated without complication. After 9 months he returned to baseline weight and has since experienced no symptom recurrence.

Table 1. Previous reports vocal fold immobility secondary to DISH.

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Conclusion

Vocal fold paralysis secondary to DISH is an extremely rare cause of symptomatic airway obstruction. Although largely asymptomatic, DISH is common in elderly patients and capable of producing symptoms encountered in otolaryngology practice including airway obstruction and dysphagia.

In cases with progressive symptoms and workup revealing the features of hyperostosis, successful recognition of DISH can result in directed and effective therapy.

In the case presented, cervical osteophytectomy dramatically reversed vocal fold motion impairment and dysphagia secondary to hyperostosis, thus negating the need for prolonged tracheostomy and feeding tube dependence.

Discussion

Criteria for DISH include (1) bridging ossification along ≥ 4 contiguous vertebral bodies, (2) preservation intervertebral disc height, and (3) absence of sacroiliac erosion, joint fusion, and apophyseal ankylosis.1

Disease is common and prevalence increases with age, found in up to 25% of men and 15% of women above age 50.2 Cervical disease is less common than thoracic, and is most often asymptomatic. However, symptoms produced are common in otolaryngology practice- including dysphagia, odynophagia, dysphonia, and stridor.

Dysphagia is the 2nd most frequent symptom of cervical osteophytes after stiffness, reported in up to 28% of total cases.3

Vocal fold motion impairment in patients meeting DISH criteria is exceptionally rare, limited to 1 previous report, in which immobility was secondary to significant glottic edema and addressed with multiple laryngoscopic laser debulking procedures.1 Proposed mechanisms include paralysis with compression of the recurrent laryngeal nerve,4 direct arytenoid cartilage fixation by osteophytes, and vascular obstruction causing edema of the vocal fold.5 Of these mechanisms, only edema has been confirmed in previous reports.

No previous DISH report has demonstrated vocal fold paralysis in the absence of vocal fold edema, or reversal of motion impairment using osteophytectomy alone.

Mild symptoms of cervical hyperostosis are managed with conservative therapy, but severe dysphagia and airway obstruction refractory to conservative therapy are indications for surgery, most often osteophytectomy using a prevascular approach.1,3 This operation has a low complication rate, with laryngeal neuroparalysis reported in only 1.5% of cases. On long-term follow-up, 87% of patients followed over time experience significant symptom improvement.4

References