Case Report

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The Integrated Management of Cervical hyperostosis in Patients with Dyspnea and Dysphagia to Reduce Risk of Airway Compromise: A Case Series

J Preece; James Metkus; V Shah*; M Mortensen

1Department of Otolaryngology and Head and Neck Surgery, Albany Medical Center, Albany NY, USA.
2Division of Otolaryngology-Head and Neck Surgery, Department of Surgery, Stony Brook Medicine, Stony Brook NY, USA.
3Albany Medical College, Albany NY USA.

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*Corresponding Author: V Shah, Albany Medical College, Albany NY, 704-770-6896, 344 Quail Street, Albany, New York 12208 USA.
Email: shahv1@amc.edu

Abstract

Objectives: Diffuse idiopathic skeletal hyperostosis (DISH) is a non-inflammatory condition diagnosed by the presence of flowing vertebral ossifications along the anterior longitudinal ligament. Limited literature on cervical spine DISH, or cervical hyperostosis (CH), demonstrates that this sub-type of DISH may present with severe oropharyngeal symptoms. Two common manifestations of CH are dysphagia and dyspnea. The purpose of this case series was to provide more insight into detection and management of CH to potentially reduce the risk of impending airway compromise.

Methods: Three patients with CH were selected from two institutions. Each patient presented with similar symptoms of dysphagia and dyspnea. The presentation, diagnosis, and management plan for each patient is described.

Results: Patients ranged from 53 to 76 years of age. All patients presented with dyspnea and dysphagia at the initial appointment, and all patients had co-existing GERD. Patient 1 and Patient 3 patients had vocal cord paralysis and pharyngeal wall fullness due to CH. Patient 2 had vocal cord paralysis and posterior glottic edema due to CH. Patient 1 and Patient 2 underwent anterior resection of their cervical osteophytes and tracheostomy. Dysphagia in both patients resolved after surgery, but their dyspnea persisted. Patient 1 required a transverse cordectomy for bilateral vocal cord paralysis, which treated his dyspnea. Patient 2 had normal vocal fold mobility post-operatively. Patient 3 had a similar pathophysiology and presentation to Patient 1, but she was lost to follow-up.

Conclusion: Existing literature demonstrates that several factors contribute to dyspnea in CH, including vocal fold paralysis and glottic edema. This series demonstrated that not only was surgery successful for partial resolution of symptoms, but also treatment with cordectomy, speech/swallow therapy, and acid-reducers was also beneficial.

Keywords: Skeletal hyperostosis; tracheostomy; dyspnea; dysphagia; cervical osteophytes.

Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) is a non-inflammatory condition characterized by osteophytic growth along the anterior longitudinal ligament. The disease process was first described after Forestier discovered radiographic changes in elderly males with spinal stiffness [1]. The prevalence of DISH increases with age, affecting 25-27% of men over 50-years-old and 12-15% of women over 50-years-old [2-4]. DISH is diagnosed by the presence of flowing vertebral ossifications at four contiguous vertebrae, preserved intervertebral disc height, absent ankylosis at the facet-joints, and absent sacroiliac joint changes [5]. DISH is considered mainly asymptomatic and is debated on whether it manifests with clinical symptoms [6]. Cervical spine DISH, or cervical hyperostosis (CH), is a less common manifestation of DISH and is associated with more severe symptoms, including dysphagia and dyspnea [5-6]. Dysphagia and respiratory compromise are the most common complaints that lead patients to seek surgical treatment. The objective of this case series was to present three patients complaining of dyspnea due to CH at two institutions and their subsequent treatment. Additionally, a comprehensive review of the literature for patients with airway obstruction and CH was performed.
Case Series Presentation

The following are summaries of three patients with CH and their subsequent treatment. **(Table 1)** shows a summary of patient findings.

**Table 1: Patient Findings.**

<table>
<thead>
<tr>
<th>Patient</th>
<th>Age</th>
<th>Presenting Symptoms</th>
<th>Nasolaryngoscopy</th>
<th>Etiology</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>72</td>
<td>Hoarseness, Dyspnea, Dysphagia</td>
<td>Bilateral Vocal Fold Paralysis</td>
<td>C4-C6 Cervical Osteophytes</td>
</tr>
<tr>
<td>2</td>
<td>79</td>
<td>Dysphagia, Dyspnea</td>
<td>Supraglottic Edema</td>
<td>C2-C6 Cervical Osteophytes</td>
</tr>
<tr>
<td>3</td>
<td>53</td>
<td>Hoarseness, Dysphagia</td>
<td>Left Vocal Fold Paralysis</td>
<td>C4-C6 Cervical Osteophytes</td>
</tr>
</tbody>
</table>

**Case 1**

71-year-old male presented to clinic for evaluation of dysphagia, dyspnea, and hoarseness. Two years prior to presentation, the patient was treated for respiratory distress with intubation and was subsequently decannulated. Past medical history included long-term SOB and GERD treated with a PPI. On exam, stroboscopy revealed bilateral vocal fold paralysis and posterior pharyngeal wall fullness concerning for CH (Figure 1). CT imaging showed severe osteophytes at C4-C5 and C5-C6 (Figures 2 and 3). Neurosurgery and otolaryngology collaborated to resect cervical osteophytes via an anterior neck approach. Postoperatively, the patient was admitted and given a thick diet. On postoperative day two, he was discharged without complications. Vocal fold paralysis and dyspnea were persistent after surgery, and the patient was surgically treated with left vocal fold transverse cordectomy and left medial arytenoidectomy. He received speech/swallow therapy and was eventually cleared for a regular diet. Although he continued to have vocal fold immobility, his dyspnea resolved after the cordectomy.

**Case 2**

76-year-old male presented to clinic for evaluation of worsening dysphagia and dyspnea. Past medical history included spine surgery for severe cord compression, tracheostomy for dyspnea with failed decannulation due to laryngeal edema, and GERD treated with a PPI. Nasolaryngoscopy showed bilateral vocal fold paresis and posterior glottic edema. On diagnostic laryngoscopy, he had a biopsy which was negative for carcinoma and was diagnosed with posterior glottic stenosis. Edema and a large bony protuberance overlying the cricopharyngeal muscle were also noted intraoperatively. CT imaging showed severe osteophytes at C4-C5 and C5-C6 compressing the larynx and upper esophageal sphincter. He underwent excision of tracheal granulation tissue and tracheostomy decannulation by otolaryngology. Later, he was treated by neurosurgery with anterior resection of cervical osteophytes. On postoperative day one, he was discharged without issue. The patient was seen by speech/swallow therapy and cleared for regular diet. Postoperatively, he had continued dysphagia but no longer reported dyspnea. Nasolaryngoscopy showed resolution of the protuberance with normal vocal fold mobility bilaterally.

**Case 3**

53-year-old woman presented to clinic for evaluation of hoarseness and dyspnea six months after treatment in the ICU for a drug overdose. While in the ICU, the patient was intubated three separate times. She was hoarse immediately after intubation and slowly improved but was still not near her normal voice. Relevant history included a current ½ pack per day smoking history, cervical neuropathy, TBI, post-polio syndrome, and HIV for which she took several medications. Nasolaryngoscopy showed left vocal cord paralysis with moderate reflux change, and stroboscopy showed vocal cord edema with left posterior pharynx fullness. The patient was prescribed a PPI and advised to quit smoking. Three months later, her swallow study showed mild dysphagia without aspiration, but the patient refused speech therapy and continued to smoke. CT imaging showed prominent anterior osteophytosis at C4-C6 levels causing mass effect on the left hypopharynx. Since being informed of these findings, the patient has not returned to the office.

**Discussion**

CH can cause various clinical manifestations involving the pharynx, larynx, and esophagus. In our series, two of our pa-
tients had complaints of dyspnea from CH requiring tracheos- 
tomy and surgery. Our third patient had symptoms consistent 
with CH but was not amenable to conservative treatment and 
has been lost to follow-up. Previous studies have shown CH 
leading to respiratory compromise and requiring intubation 
or tracheostomy [7-8]. In fact, existing literature points that 
dysphagia and respiratory compromise were the most com-
mon complaints, affecting 93% and 31% of patients with CH, 
respectively [9]. Reported explanations for dyspnea due to CH 
include compression, glottic edema, and vocal fold immobi-
licity [7-8]. In our series, dyspnea likely resulted from vocal fold 
immobility due to CH in Patients 1 and 3 and from vocal fold 
immobility and edema in Patient 2.

While CH treatment involves conservative strategies to reduce 
aspiration and GERD with reflux control and speech/swallow 
therapy, surgery has also been proven successful [8]. Cur-
rent literature demonstrates that most patients with CH are 
treated operatively with an anterolateral approach, achieving 
a high success rate [10]. While two patients were treated with 
anterior resection of the cervical osteophytes in this series, 
they did not have immediate resolution of their symptoms. 
For Patient 1, the vocal fold paralysis, and thus dyspnea, per-
sisted after resection and only resolved after cordectomy. For 
Patient 2, resection resulted in treatment of the edema and 
vocal fold paralysis, although the patient continued to have 
dysphagia. In contrast to the existing literature, in which most 
studies found resolution of dysphagia post-operatively, our 
study observed resolution of dyspnea by treating the underly-
ing cause. Overall, in patients with cervical DISH and dyspha-
gia, early treatment may prevent disease progression to air-
way compromise.

Conclusion

While DISH is traditionally believed to be asymptomatic, CH 
should be considered for complaints of dyspnea among older 
patients that present with dysphagia. Early treatment of CH in 
elderly patients with dysphagia should be considered to po-
tentially prevent airway compromise.

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