Diffuse Idiopathic Hyperostosis Manifesting as Dysphagia and Bilateral Cord Paralysis: A Case Report and Literature Review

Amer Sebaaly[1,2], Ghassan Boubez[1], Tarek Sunna[3], Zhi Wang[1], Elie Alam[4], Apostolos Christopoulos[5], Daniel Shedid[3]

Key words
- Diffuse idiopathic hyperostosis
- Dysphagia
- Vocal cord paralysis

Abbreviations and Acronyms
CT: Computed tomography
DISHphagia: Diffuse idiopathic hyperostosis dysphagia

From the [1]Department of Orthopedic Surgery, Centre Hopitalier de l'Université de Montréal, Montréal, Quebec, Canada; [2]Faculty of Medicine, Saint Joseph University, Beirut, Lebanon; [3]Department of Neurosurgery, Spine Unit, Centre Hopitalier de l'Université de Montréal, Montréal, Quebec, Canada; [4]Otolaryngology Department, University of Miami, Miller School of Medicine, Miami, Florida, USA; and [5]Department of Otorhinolaryngology, Centre Hopitalier de l'Université de Montréal, Montréal, Quebec, Canada

To whom correspondence should be addressed: Amer Sebaaly, M.D., M.Sc. [E-mail: amersebaaly@hotmail.com]

Citation: World Neurosurg. (2018) 111:79-85. https://doi.org/10.1016/j.wneu.2017.12.063

Journal homepage: www.WORLDNEUROSURGERY.org

Available online: www.sciencedirect.com

BACKGROUND: **Diffuse idiopathic hyperostosis (DISH) is characterized by calcifications affecting mainly the spinal anterior longitudinal ligament. This disease is mainly asymptomatic but cervical osteophytes can sometimes cause dysphagia (DISHphagia), hoarseness, and even dyspnea.**

CASE DESCRIPTION: **We report, for the first time in the medical literature, a case of a 76-year-old patient with DISH causing an important dysphagia as well as bilateral vocal cord paralysis causing critical dyspnea. The patient was surgically treated by anterior resection of the osteophytes and application of bone wax, with significant clinical improvement and no radiologic recurrence after 2 years of follow-up.**

DISCUSSION AND CONCLUSION: **A thorough literature review didn't yield any article reporting on bilateral vocal cord paralysis caused by DISH. Management of this condition is typically multidisciplinary, and treatment of cervical osteophyte-associated dysphagia or respiratory compromise is primarily medical, after performing necessary tests to rule out other causes of dysphagia. Surgical intervention is warranted when medical treatment fails, when there is weight loss, a significant airway compromise or sleeping alterations. A treatment algorithm is proposed in the end of this review for symptomatic anterior osteophytes caused by DISH in the mobile cervical spine.**

INTRODUCTION

Forestier and Rotes-Querol[1] first described diffuse idiopathic hyperostosis (DISH) in 1950, with calcifications affecting mainly the anterior longitudinal ligament and less frequently the posterior longitudinal ligament.[2] DISH can manifest in a variety of ways, but in the majority of cases it is asymptomatic to the point at which some authors do not consider it a disease but an incidental findings on spine radiographs.[3] Its prevalence is variable, mainly because of different diagnostic criteria used, and ranges between 2.9% and 30% in the general population (3% of men at age 40, 18.8% of men at age 60, and 32.1% of men at age 80).[3-6] DISH affects frequently the mobile cervical spine (10% to 30%), mostly in men older than 60 years old (M:F = 2:1).[6] Nonetheless, the diagnosis is made when images obtained done for other reasons, in the knowledge that this disease is frequently asymptomatic.[4,7] Cervical osteophytes can nonetheless become symptomatic, causing most commonly dysphagia but also hoarseness, dysphonia, and even dyspnea.[2,3] Dysphagia, caused by anterior osteophytes in the cervical spine and referred by some authors as DISHphagia,[8] affects only 0.1% to 6% of patients with DISH.[5,6] Fewer patients have upper extremity compromise,[5] and the association with acute respiratory distress is even more uncommon.[4] There is no report in the medical literature, to our knowledge, of bilateral vocal cord paralysis cause by cervical DISH.

We herein report a case of patient with DISH causing important dysphagia and bilateral vocal cord paralysis causing critical dyspnea. We also review the literature for similar cases and propose a treatment algorithm for symptomatic anterior osteophytes caused by DISH in the mobile cervical spine.

CASE REPORT

A white 76-year-old man was referred in September 2013 to the otolaryngology and head and neck surgery clinic for laryngeal obstruction. The man was already known for dysphagia that had progressed since 2008. Clinical findings and investigations had led to the diagnosis of idiopathic left vocal cord paralysis in 2010. On the other hand, between 2010 and 2013, there was an increase of dysphonia, dyspnea on exertion, and dysphagia, which had led to a weight loss of 40 pounds (20% of his weight).

Examination of the larynx showed bilateral vocal cord paralysis, which required a tracheostomy in October 2013. Computed tomography (CT) of the neck

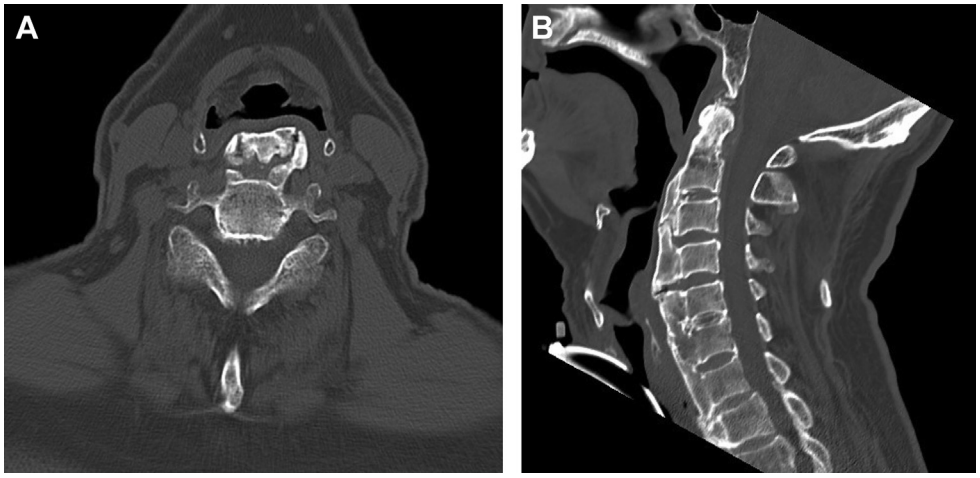

Figure 1. Preoperative computed tomographic scan of patient showing large osteophyte on (**A**) axial and (**B**) sagittal views with compression of the esophagus.

performed at the same time showed no lesion that could explain this paralysis (Figure 1). The only scan abnormality was ossification of the anterior longitudinal ligament, which protruded into prevertebral soft tissues over all the cervical spine, even pushing the inlet of the esophagus. Bilateral vocal cord paralysis caused severe dysphagia in this patient. He had to be fed by gavage only. The paralysis was still present in March 2014, and a neurosurgical consultation was requested to address the only abnormality that was identified in the scan.

The patient was operated on in July 2014 by anterior resection of the osteophytes with a high-speed burr and application of bone wax for hemostasis. His postoperative course was unremarkable, and the suction drain was removed on the second postoperative day. A postoperative CT scan is shown in Figure 2.

Follow-up evaluations in speech therapy 1 month and 2 months after surgery showed a significant improvement in the patient's voice quality, breathing, and swallowing ability. Examination of the larynx in September 2014 showed an outline of movement of the right vocal cord. These findings enabled progressive resumption of oral feeding. Three months later, the clinical evaluation demonstrated normal swallowing and normal breathing. The patient's tracheostomy tube was thus removed, as was his gastrostomy tube. Two years later, the patient had been regularly evaluated at the ear, nose, and throat clinic and demonstrated normal

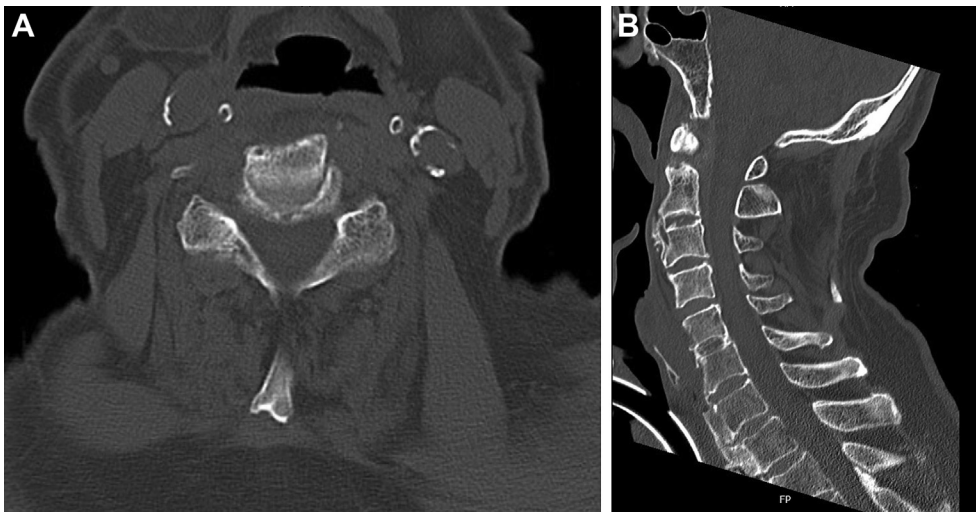

Figure 2. Immediate postoperative computed tomographic scan showing resection of the osteophytes.