International Journal of Otorhinolaryngology and Head and Neck Surgery
Bustos FA et al. Int J Otorhinolaryngol Head Neck Surg. 2021 Mar;7(3):533-535
http://www.ijorl.com
pISSN 2454-5929 | eISSN 2454-5937

DOI: https://dx.doi.org/10.18203/issn.2454-5929.ijohns20210691

Case Report

Diffuse idiopathic skeletal hyperostosis: an uncommon complication in head and neck surgery

Felipe A. Bustos[1], Felipe A. Capdeville[2]*, Daniel A. Rappoport[2,3], Luis F. Zanolli[4], Fabio Valdes[2], Hugo E. Rojas[2], Jose M. Contreras[4], Giancarlo Schiappacasse[5], Arturo J. Madrid[2]

[1]Head and Neck Unit, Instituto Nacional del Cancer, Santiago, Chile
[2]Department of Surgery, Clinica Alemana de Santiago, Santiago, Chile
[3]Department of Surgery, Hospital Clínico de la Universidad de Chile, Santiago, Chile
[4]Department of Otorhinolaryngology, Clinica Alemana de Santiago, Santiago, Chile
[5]Department of Radiology, Clinica Alemana de Santiago, Santiago, Chile

Received: 06 January 2021
Revised: 11 February 2021
Accepted: 12 February 2021

***Correspondence:**
Dr. Felipe A. Capdeville,
E-mail: fcapdeville@alemana.cl

ABSTRACT

Diffuse idiopathic skeletal hyperostosis (DISH) is a degenerative disorder of unknown etiology that most often occurs in male patients over 50. Dysphagia is its main symptom, but they can also have dyspnea, otalgia, cough, sore throat, foreign body sensation in the pharynx, sleep apnea and glottic alterations. We present a case report and review the literature about this entity. We report a case of an oral squamous cell carcinoma that received a commando surgery and tracheostomy tube. Decanulations attempts were unsuccessful initially due to DISH. Conservative management was successful and complete rehabilitation performed, achieving decannulation 18 months after surgery. DISH can be a source of many different symptoms that may appear or be exacerbated after any surgery, and produce a postoperative complication. Conservative management is usually the best treatment, leaving surgical interventions for severe symptomatic patients. The knowledge of this entity and a high level of suspicion are very important for a proper diagnosis and management.

Keywords: Forestier syndrome, Hyperostosis, DISH

INTRODUCTION

Diffuse idiopathic skeletal hyperostosis (DISH) is a degenerative disorder of unknown etiology that most often occurs in male patients over 50. Is associated with obesity, glucose intolerance and diabetes mellitus.[1]

Dysphagia is its main symptom, but they can also have dyspnea, otalgia, cough, sore throat, foreign body sensation in the pharynx, sleep apnea and glottic alterations.[2] We want to present a case that turned symptomatic, just after a commando surgery and a review of the literature.

The article was approved by the scientific and ethical committee of "facultad de medicina clínica alemana universidad del desarrollo".

CASE REPORT

70 years old man with a history of tobacco use, arterial hypertension and dyslipidemia. He presented a two

months history of right mandibular tumor in the alveolar ridge without accompanying symptoms.

Physical examination revealed a 2 centimeter verrucous tumor of the right mandibular alveolar ridge with no suspicious cervical lymph nodes. Incisional biopsy confirmed a well-differentiated squamous cell carcinoma. Imaging study with PET-CT revealed no cervical or distant metastasis.

The case was reviewed at tumor board, and right-hemimandibulectomy with bilateral neck dissection, tracheostomy and reconstruction with a left fibular free flap was performed. The patient had an uneventful postoperative course in the ICU.

On postop day 12 tracheostomy decannulation was attempted, but patient required re-cannulation 6 hours after due to laryngeal stridor. Laryngoscopy evidentiated adequate swallowing and no airway aspiration signs.

A new decannulation attempt was performed, but the patient presented severe dyspnea and recannulation was needed. A new laryngoscopy showed glottic edema, salivary retention and bilateral vocal fold palsy in the median position. Laryngeal electromyography observed normal phasic activity with no signs of acute denervation or neural lesions.

The patient was discharged with a tracheostomy tube placed. After a month a new laryngoscopy revealed salivary retention in the hypopharynx, moderate symmetrical arytenoid edema. Right vocal fold in the paramedian position and decreased mobility. Left vocal cord presents a notorious decreased mobility. Laryngeal opening was only achieved in forced inspiration by left vocal cord mobility.

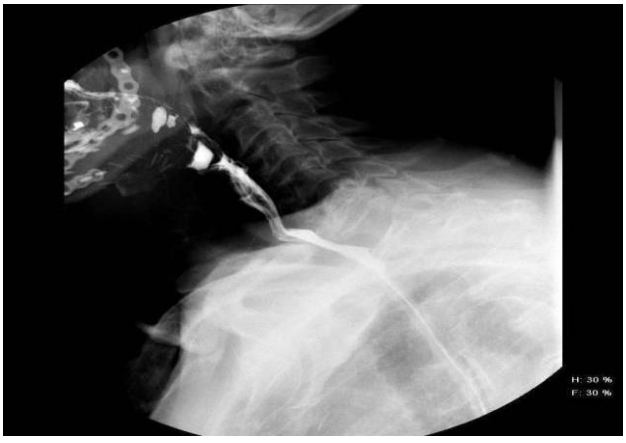

Figure 1: Contrast esophagogram: an anterior displacement of the cervical oesophagus is observed due to the presence of ossification of the anterior longitudinal ligament between C4 and C6 vertebral bodies.

Esophagogram (figure 1) and neck CT (figure 2) revealed anterior displacement of the cervical esophagus and bilateral crico-arytenoid junction due to a protruding mass caused by the presence of ossification of the anterior longitudinal ligament in C4 to C6 vertebral bodies.

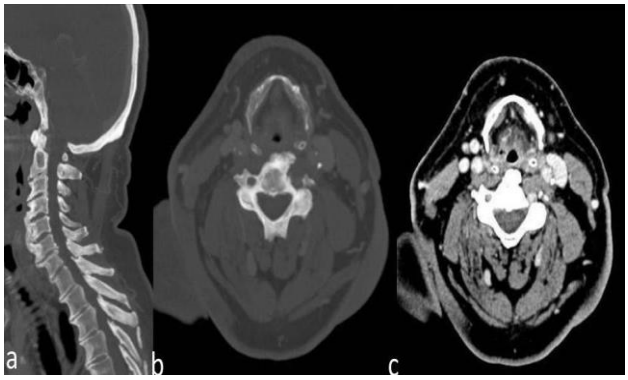

Figure 2: Neck CT (a) sagittal reconstruction, (b and c) axial projections: ossification of the anterior longitudinal ligament most evident between C4 and C6. Which contacts the posterior wall of the upper airway and mildly displaced crico-aryteoind junction.

The patient had a neurosurgical and spine surgery consultation. He underwent a conservative management with NSAIDs and phonoaudiologic therapy with complete rehabilitation and tracheostomy tube decannulation 18 months after his initial surgery.

DISCUSSION

This disease was first reported in 1950 by Forestier and Rotes-Querol, who described it as "senile ankylosing hypersotosis of the spine".[3]

There are many terms to describe this condition including Forestier's disease, cervical spondylosis, ankylosing spondylosis, degenerative disc disease, spondylosis deformans and ankylosing hyperostosis but in 1975 DISH was introduced.[4]

DISH is a frequent condition but rarely diagnosed. In the United States, the estimated prevalence is 25% in men and 15% in women age 50 and older, and 35% in men and 26% in women among those 80 years and older.

Its main characteristic is the ossification of the anterior longitudinal spinal ligament (ALL), with large osteophytes that flow down the spine, producing a typical candle wax appearance. The most typical involved sites are the cervical and thoracic vertebral segments.

These osteophytes are known as "hypertrophic anterior cervical osteophytes (HACO)" and are responsible for compressive pharyngoesophageal and laryngotracheal symptoms, mainly dysphagia.

In the cervical spine, the most commonly affected segments are C5-C6 (40%), C4-C5 (23%), C3-C4 (14%) and C2-C3 (14%). During its evolution, DISH can develop ossification at extraspinal locations.

DISH is usually asymptomatic and therefore latent; however, it can lead to significant morbimortality. The main symptom is dysphagia, which can occur in 28% of the patients correlating to segments C5-C6 and C4-C5.

Possible explanations are mechanical obstruction of food bolus in the posterior wall of the hypopharynx or esophagus; impaired epiglottic motility; incomplete glottal closure due to restricted vocal fold mobility; distortion of the larynx or the laryngeal cartilages; restriction of the anterior displacement and elevation movement of the larynx; peri esophagitis and peri pharyngitis, fibrosis and adhesions with fixation of the esophagus; cricopharyngeal spasm triggered by pressure on the esophagus. Is usually progressive, logic, improves with cervical flexion and worsens with hyperextension.

Other symptoms have been described such as dyspnea, otalgia, cough, sore throat, foreign body sensation in the pharynx, sleep apnea and glottic alterations.[2]

In 1976 Resnick and Niwayama established the radiological diagnostic criteria for this disease: calcification or ossification of the anterior longitudinal spinal ligament in at least 4 consecutive vertebral bodies; conservation of the height of the intervertebral disk in the affected areas; absence of apophyseal joint ankylosis and sacroiliac joint sclerosis.[5]

The most common radiographic features include linead new bone formation along the anterolateral aspect of the thoracic spine, a bumpy contour, subjacent radiolucency, and irregular and pointed bony excrescences at the superior and inferior vertebral margins in the cervical and lumbar regions.[2]

Treatment is usually directed to symptoms only. Strategies include hygiene and diet measures, swallowing and nutritional support, avoidance of aspiration pneumonia, physical therapy, and the use of NSAIDs, antalgiques and myorelaxants in case of mild dysphagia and cervical pain.

Surgical resection of anterior cervical osteophytes has been suggested as an effective, albeit last resort treatment in severely symptomatic patients, with good short and long-term results.

Usually it presents as a degenerative, slow evolving condition, but in our patient, it started the symptoms in the early postoperative period. Even though he hadn't signs of suspicion in the pre surgical images, we think that he must have had an initial hiperostosis that due the trauma of surgery, inflammation and fibrosis, it suddenly turned symptomatic after surgery, compromising decannulation and swallowing. Fortunately, he responded to conservative management and is asymptomatic nowadays.

CONCLUSION

DISH can be a source of many different symptoms that may appear or be exacerbated after any cervical surgery, and produce a postoperative complication. Conservative management is usually the best treatment, leaving surgical interventions for severe symptomatic patients. The awareness of this condition should increase diagnosis, and proper management of mild symptomatic patients, and anticipate its postoperative consequences in order to prevent complications and prompt early treatment. The knowledge of this entity and a high level of suspicion are very important for a proper diagnosis and management.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Vengust R, Mihalic R, Turel M. Two different causes of acute respiratory failure in a patient with diffuse idiopathic skeletal hyperostosis and ankylosed cervical spine. Eur Spine J Off Publ Eur Spine Soc Eur Spinal Deform Soc Eur Sect Cerv Spine Res Soc. 2010;19(2):130-4.
2. Pulcherio JOB, Velasco CMM de O, Machado RS, Souza WN de, Menezes DR de, Pulcherio JOB, et al. Forestier's disease and its implications in otolaryngology: literature review. Braz J Otorhinolaryngol. 2014;80(2):161-6.
3. Forestier J, Rotes-Querol J. Senile ankylosing hyperostosis of the spine. Ann Rheum Dis. 1950;9(4):321-30.
4. Resnick D, Shaul SR, Robins JM. Diffuse idiopathic skeletal hyperostosis (DISH): Forestier's disease with extraspinal manifestations. Radiology. 1975;115(3):513-24.
5. Caminos CB, Cenoz IZ, Louis CJ, Otano TB, Esáin BF, Pérez de Ciriza MTF. Forestier disease: an unusual cause of upper airway obstruction. Am J Emerg Med. 2008;26(9):1072-3.

Cite this article as: Bustos FA, Capdeville FA, Rappoport DA, Zanolli LF, Valdes F, Rojas HE, et al. Diffuse idiopathic skeletal hyperostosis: an uncommon complication in head and neck surgery. Int J Otorhinolaryngol Head Neck Surg 2021;7:533-5.