Forestier's Disease Presenting as a Retropharyngeal Mass in an Elderly with Dysphagia

Dear Editor,

Diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier's disease, is a non-inflammatory enthesopathy of unknown etiology. Affecting predominantly men, it results in flowing, robust ossification of the anterior longitudinal ligament of the spine. In contrast to ankylosing spondylitis, the disc space itself is usually spared and by definition, it affects 4 or more intervertebral levels.¹ We report a case of diffuse idiopathic skeletal hyperostosis presenting as a retropharyngeal mass in an elderly male patient with dysphagia for years.

Case Report

An 81-year-old male presented to the emergency department because of dysphagia with recent aggravation for years. Physical examination showed thyroid enlargement. Laryngoscopy revealed a bulging posterior hypopharyngeal wall that resembled a retrophayngeal mass (Fig. 1A). His neck radiograph in lateral view (Fig. 1B) showed large ventral marginal osteophytes (VMO) and ossification of the anterior longitudinal ligament (OALL) from C3 through C7 of the spine, which was consistent with diffuse idiopathic skeletal hyperostosis (DISH). Although esophagogram showed no esophageal obstruction, multidetector computed tomography (MDCT) depicted severe posterior compressions on the hypopharynx and esophagus by large VMO and OALL of spine at C2/3 through T2. (Figs. 1 C and D) Dysphagia caused by DISH gradually improved under the conservative treatment with peroral acetaminophen in powder form and soft diet.

Discussion

Degenerative change of the cervical and thoracic spine may result in Forestier's disease, which is characterised

Fig. 1. An 81-year-old male patient complained of progressive dysphagia for years. (A) His laryngoscopic image reveals a bulging posterior hypopharyngeal wall (large arrows) with obliteration of bilateral piriform sinuses like a retrophayngeal mass. (B) The neck radiograph in lateral view shows a soft tissue density enlarged thyroid gland (small arrowheads) and large ventral marginal osteophytes and ossification of the anterior longitudinal ligament (small arrows) from C3 through C7 of the spine and narrowing of the hypopharynx. (C & D) Multidetector computed tomography (MDCT) of the neck with reconstructed axial image shows thyroid goiter and large ventral marginal osteophytes and ossification of the anterior longitudinal ligament (small arrows) of spine at C2/3 through C6 and the visible C7 through T2 levels with severe posterior compression on the hypopharynx and the esophagus with invagination (large arrowhead) of the posterior tracheal wall in the upper thorax.

by extensive spinal osteophytosis and endo-chondral ossification of paravertebral ligaments and muscles.^{1,2} Diagnosis of DISH is based on the radiographic criteria of Resnick et al,¹ including (i) presence of flowing osteophytes on the anterolateral margin of at least four consecutive vertebral bodies, (ii) preservation of disk heights, and (iii) lack of degenerative arthritis of the apophyseal joints and sacroiliac joints. The etiology of DISH is unknown. Some metabolic conditions, such as dyslipidaemia, hyperinsulinemia, hyperuricaemia, and hypertension, are considered as risk factors because of their frequent associations with DISH.³ Degenerative changes in the cervical spine are a common problem in the elderly but DISH can also occur with predilection of OALL in older men.^{3,4}

Dysphagia from DISH (DISHphagia) affects only 0.1% to 6% of adults in their lifetime.² The mechanisms of DISHphagia include

(i) direct impact of flowing osteophytes causing luminal impingement,

(ii) peri-esophageal inflammation and edema induced by

chronic irritation of laryngoesophageal soft tissues,

(iii) any osteophyte strategically compressing the

esophagus at the cricoid cartilage level,

- (iv) pain and muscle spasm owing to irritation, or
- (v) combination of any of these mechanisms.²

DISH of the cervical or thoracic spine may also cause acute dyspnea, dysphonia, foreign body sensation and aspiration of liquid.^{4,5} Generally, DISHphagia, at first, is caused by mechanical obstruction followed by inflammation and fibrosis.⁵ DISHphagia may be mild in elderly patients because the sensory discrimination threshold of laryngopharynx increases with increasing age.⁶

We suggest clinicians to consider treating elderly with mild dysphagia and suspected retropharyngeal mass as DISHphagia patients. Swallowing is a complex, multi-stage process consisting of oral, pharyngeal, and esophageal phases.³ The etiology of dysphagia is multifactorial (i.e. anatomical, mechanical or neurologic) and it may occur in any elderly patients complaining of symptoms of dysphagia. Its treatment also needs a multidisciplinary approach.³Imaging modalities are important in the diagnosis of DISHphagia. Plain radiography, esophagography, videofluoroscopy of swallowing, MDCT and magnetic resonance imaging (MRI) can detect its presence.^{1,5,7} Most of the elderly patients with DISHphagia initially have good responses to medical treatments with liquid forms of non-steroid anti-inflammatory drugs and soft diet. After cautiously ruling out other intrinsic or extrinsic causes of difficulty in swallowing, surgical resection of the VMO may be undertaken for resolution of symptoms.^{5,7}

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