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DIFFUSE IDIOPATHIC SKELETAL HYPEROSTOSIS-MEDIATED DYSPHAGIA

Medicine

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ABSTRACT

A 62-year-old man who presented with recurrent aspiration pneumonia and weight loss was diagnosed with oropharyngeal dysphagia due to unilateral vocal cord paralysis. Imaging of the neck by CT as well as ancillary tests have revealed diffuse idiopathic skeletal hyperostosis ('DISH') as the cause, mediated by pressure of an osteophyte on the recurrent laryngeal nerve. With the ageing of the populations clinicians may increasingly encounter diffuse idiopathic skeletal hyperostosis and should be aware of the syndrome and its unusual complications.

KEYWORDS

Dysphagia; Aspiration; Vocal cord paralysis; Diffuse idiopathic skeletal hyperostosis

INTRODUCTION

Dysphagia is a major problem to the patient, as well as sometimes an intriguing riddle to the clinician who has to correctly identify its pathogenesis to help his or her patient, as the following report demonstrates.

Case report

A 62-year-old patient, institutionalized for paranoid schizophrenia and treated with quetiapine 12.5 mg bid, was admitted with sudden fever (39.2ºC), desaturation (88%), sinus tachycardia (140/min) and hypotension (71/46 mmHg). He had a history of 2 recent hospitalizations for pneumonia and significant weight loss. C-reactive protein was 275 mg/dL. Chest X-ray demonstrated right lower lobe infiltrate. Cultures were negative. He recovered with intravenous fluids and tazobactam treatment.

Focused questioning yielded a history of recent dysphagia with immediate coughing upon swallowing. He had no known neurological disease. Neurological examination and head CT were normal. Flexible laryngoscopy demonstrated Rt. vocal cord paralysis and pooling of secretions and food in the right pyriform sinus and post-cricoid area. CT imaging of the chest/abdomen and endoscopy/colonoscopy were normal. On barium swallow test, contrast media remained in the glottis without passage to the esophagus. There was no movement of the epiglottis and contrast appeared in the trachea followed by enhancement of the bronchial tree. Neck CT demonstrated diffuse idiopathic skeletal hyperostosis (DISH) with calcification of the anterior longitudinal ligament and large anterior osteophytes (Fig. 1). Surgery was deemed unadvisable due to his psychiatric illness. Percutaneous endoscopic gastrostomy (PEG) for feeding and drug administration was placed and he was discharged back to his institution.

DISCUSSION

Our patient's recurrent aspiration pneumonia and cachexia were due to oropharyngeal dysphagia secondary to unilateral vocal cord paralysis. This entity is most commonly caused by malignancy (particularly lung and thyroid cancer) involving the vagus nerve or the recurrent laryngeal nerve in the skull- base, neck, or chest or it can be due due to iatrogenic nerve injury during neck surgery. Approximately one third of the cases remain idiopathic (1). Our patient is exceptional since he had no iatrogenic or neoplastic causation and pronounced cervical spinal column pathology was the only abnormality found (Fig. 1). Cervical osteophytes are very common but a rare cause of dysphagia (2).

Diffuse idiopathic skeletal hyperostosis (DISH, Forestier disease) is a non-inflammatory condition of unknown cause, manifesting increasing prevalence with age and male predominance (3). In a population-based study of DISH, 27% of men and 13% of women were positive, and over a third of the patients were older than 75 years of age. Diffuse idiopathic skeletal hyperostosis is characterized by enthesopathy (bony proliferation at sites of tendinous and ligamentous insertion of the spine) resulting in anterior cervical osteophytes and by ossification of the paravertebral ligaments. DISH is often an asymptomatic incidental finding on neck imaging - best demonstrated by CT which is considerably more sensitive than conventional radiography. The laboratory profile in diffuse idiopathic skeletal hyperostosis is unremarkable.

However, some of the patients may become symptomatic and develop pain and stiffness of the thoracic and cervical spine. Other notable but unusual symptoms include compression of the spinal cord (myelopathy), pressure on the airway (leading to dyspnea), or esophageal compression (causing dysphagia) (3). Thus, DISH-associated dysphagia may result from pressure of large anterior cervical osteophytes on the esophagus. However, another mechanism of dysphagia in diffuse idiopathic skeletal hyperostosis may be due to pressure in the neck mediating recurrent laryngeal nerve palsy, as in our patient. This phenomenon is exceedingly rare, although 3 cases involving bilateral vocal cord paralysis have been reported (4). In addition, degenerative osteophytes-associated vocal cord paralysis has also been described, albeit rarely (5).

CONCLUSIONS

The severity of the consequences of diffuse idiopathic skeletal hyperostosis (which may include recurrent aspiration, aspiration pneumonia, and cachexia) warrants surgery in selected cases. Wider recognition by clinicians is warranted, since they are likely to increasingly encounter DISH and its complications, due to the ageing of the population.

Legend to the figure

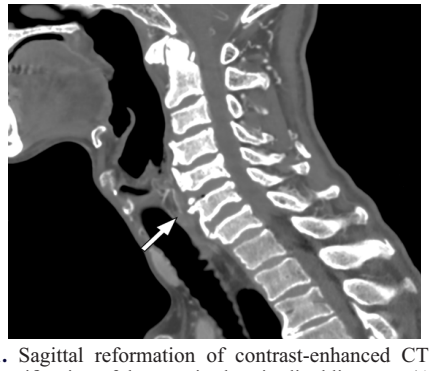

Figure 1. Sagittal reformation of contrast-enhanced CT of neck showing ossification of the anterior longitudinal ligament (ALL) with osteophytic protrusions consistent with diffuse idiopathic skeletal hyperostosis (DISH). One florid protrusion at the level of the T6 upper endplate (arrow) exerts pressure on the cricopharyngeal muscle (i.e. upper esophageal sphincter) and on the right recurrent laryngeal nerve causing unilateral vocal cord paralysis and recurrent aspirations.

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