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Case Reports & Case Series

Dysphagia, dysphonia & dyspnoe caused by ostrich beak-like anterior C1-C2 cervical osteophyte

Hardik Sardana (M.Ch.)*, Hitesh Inder Singh Rai (MBBS), Amandeep Kumar (M.Ch.), Deepak Agrawal (M.Ch.), S.S. Kale (M.Ch.)

Department of Neurosurgery and Gamma Knife, All India Institute of Medical Sciences, Ansari Nagar, New Delhi, India

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ABSTRACT

Diffuse idiopathic skeletal hyperostosis (DISH) or Forestier's disease is associated with cervical osteophytes, most commonly at the C5–6 level. We report a large beak-like anterior osteophyte at C1/C2 level presenting with severe dysphagia, dysphonia and dyspnoea.

1. Introduction

Cervical spinal osteophytes are seen in 10 to 30% of the general population; however, they are largely asymptomatic [1,5–7]. Extrinsic compression by large anterior osteophytes may occur in cervical spondylosis, most commonly in diffuse idiopathic skeletal hyperostosis (DISH) or Forestier's disease [2–4]. The obstruction occurs most commonly at the C5/6 vertebral level and less commonly at C4/5, C2/3 and C3/4, and is rarest at the C1/2 level [2]. We present a 48 year old male with giant anterior osteophyte at the C1/C2 level suffering from progressive dysphagia to both solids and liquids, along with dysphonia and dyspnoea.

2. Case report

A 55-year-old man was being evaluated for progressive dysphagia to both solids and liquids for the past 2 years that involved episodes of choking and aspiration that progressively increased in frequency. It was associated with dyspnoea, dysphonia and persistent foreign body sensation in the oropharynx. There was no history of hearing loss, tongue deviation or fasciculations or symptoms suggestive of bulbar palsy. Neurological examination was normal. His oral examination revealed a large protuberance in the retropharyngeal wall (Fig. 1A, B). The lateral skull topograph, sagittal and axial computerized tomography (CT) scan, and sagittal T2w magnetic resonance imaging (MRI) film (Fig. 1C, D, E, F) revealed marked anterior ostrich beak-like osteophyte at C1-C2 level, with marked ossification of anterior longitudinal ligament and relative preservation of intervertebral disc spaces.

After detailed evaluation, the patient was taken up for definitive surgery. A trans-oral approach was utilized to drill the large beak-like osteophytes (Fig. 2). The patient was kept on Ryle's tube feeding for 1 week after which he was allowed orally. He had significant reduction in dysphagia and dyspnoea. A comparison of the preoperative and postoperative cervical spine X rays (lateral view) is represented in Fig. 3.

3. Discussion

Zahn recognized cervical osteophytes causing dysphagia as early as 1904 [8,9]. However, the first two cases of spondylitic dysphagia were reported by Mosher in 1926 [10]. Iglauer reported the first surgical excision of an osteophyte 12 years later, in 1938 [11].

Radiographic criteria for diagnosis of Forestier's Disease (Resnick, et al) [12], as seen in our patient, are as follows: 1) Flowing calcification and ossifications within the anterior longitudinal ligament of at least four contiguous vertebral bodies, and most commonly involving the lower thoracic spine. Four is an arbitrary figure designed to differentiate DISH from spondylosis deformans, a distinction that may prove to be artificial. 2) A paucity of radiographic signs of degenerative disc disease, although these may coexist. 3) Absence of apophyseal joint ankylosis and sacroiliac joint erosion, sclerosis, or intra-articular osseous fusion. This condition occurs more frequently in men than in women, typically in their 50's and 60′ [3].

According to the published literature, anterior cervical osteophyte is responsible for dysphagia (the most frequent symptom) in 6—28% of patients with DISH [3,13,14], although most patients with such osteophytes remain totally asymptomatic. Isolated dysphagia is present in nearly 75% of the cases and associated with dyspnoea in only 14% of

* Corresponding author.
E-mail address: hardiksardana@yahoo.co.in (H. Sardana).

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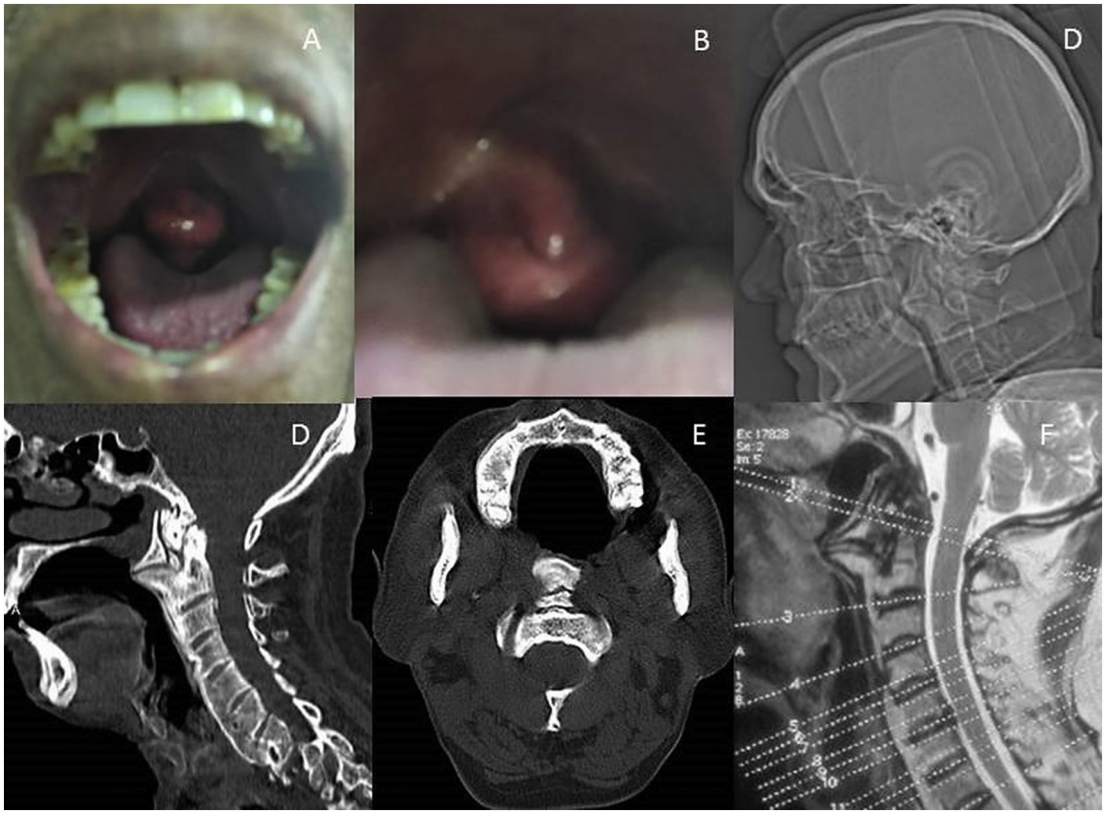

Fig. 1. A) Clinical photograph of the patient of the patient showing a large protuberance in the retropharyngeal wall. B) Magnified view of the same. C) Lateral skull topograph, D) Sagittal CT, E) Axial CT, F) Sagittal MRI images of the patient showing marked anterior beak-like osteophyte at C1-C2 level, with marked ossification of anterior longitudinal ligament and relative preservation of intervertebral disc spaces.

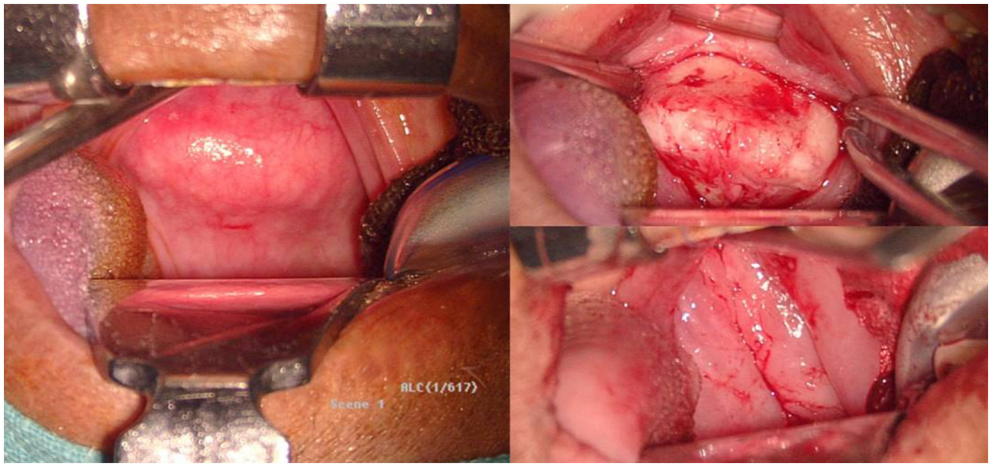

Fig. 2. Sequential steps involved in excision of the osteophyte.

patients. Aspiration (9%), stridor (3%), cervical pain (3%), cough (3%), dysphonia (2%) are other uncommonly reported presentations [13–17].

Pathophysiology of dysphagia includes mechanical compression causing esophageal obstruction, periesophagitis, peripharyngitis, cricopharyngeal spasm, impaired epiglottic motility, and distortion of the larynx and/or the laryngeal cartilages [11–17].

Surgical excision for commonly found osteophytes below C2 level is most easily accomplished through an anterior cervical discectomy approach, as described by Iglauer in 1938 [11], and then again by Patterson and Byerly [15], and Smith and Robinson [16], both in 1958. The much rarer osteophytes at C1–2 require a trans-oral decompression [17]. Transoral Endoscopic Resection of High Cervical Osteophytes has also been described [18].

Disclosures

None.

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N/A

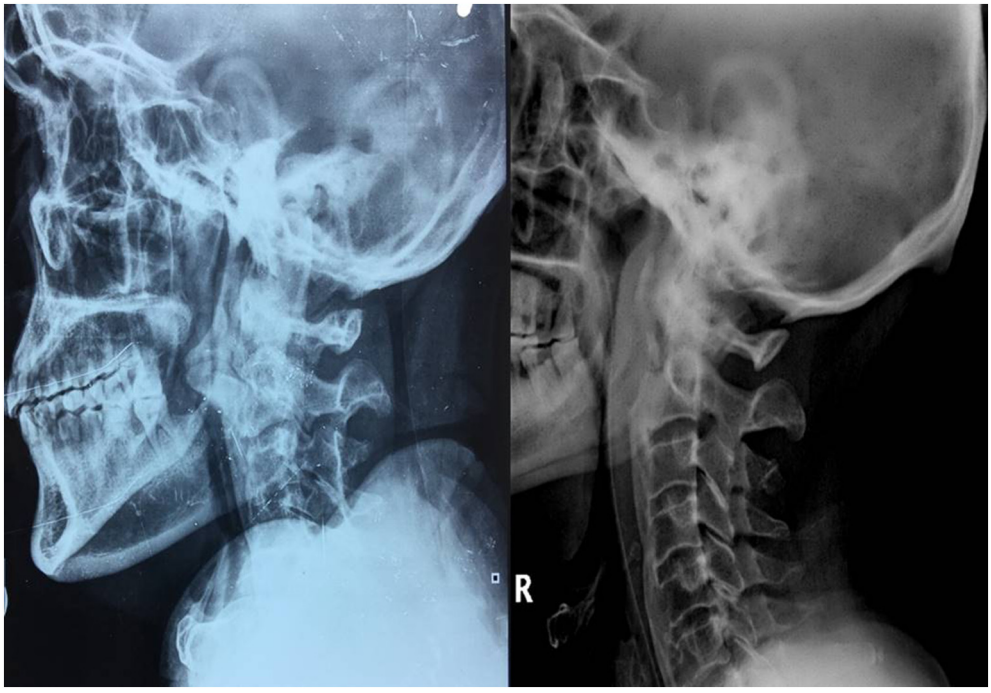

Fig. 3. Preoperative Xray Neck (left) & Postoperative Xray Neck (Right).

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