

Dysphagia Due to Diffuse Idiopathic Skeletal Hyperostosis of the Cervical Spine

Servikal Omurganın Diffüz İdiyopatik Hiperostozuna Bağlı Gelişen Disfaji

ABSTRACT

Diffuse idiopathic skeletal hyperostosis (DISH) or Forestier's disease is a common disorder of unknown etiology that is characterized by ossification of the anterior longitudinal ligament of the spine and various extra-spinal ligaments. We present the case of a 54-year-old woman with progressive dysphagia due to DISH of the cervical spine, which is a relatively rare pathology in neurosurgical practice. The cervical osteophytes extending from C2 to C4 and external compression of the pharyngoesophageal segment by the large osteophytes were demonstrated by X-ray, magnetic resonance imaging, and computed tomography. Surgical removal of the large osteophytes and a short-term nonsteroidal anti-inflammatory drug regimen led to the resolution of dysphagia. The clinical manifestations, diagnosis, and treatment of this unusual condition are discussed.

KEY WORDS: DISH, Dysphagia, Hyperostosis, Osteophyte, Spine, Treatment

ÖZ

Yaygın idiyopatik iskelet hiperostozu (DISH) ya da Forestier Hastalığı, etyolojisi bilinmeyen ve omurganın anterior longitudinal ligamenti ve çeşitli ekstra-spinal ligamentlerin ossifikasyonu ile karakterize bir hastalıktır. Bu raporda; nöroşirürji pratiğinde nadir bir patoloji olan, servikal omurganın DISH'inin ilerleyici disfajiye neden olduğu 54 yaşında bir kadın olgu sunulmuştur. C2-4 arasında uzanan servikal osteofitlerin varlığı ve faringo-özefagial segmentin büyük osteofitler tarafından eksternal basısı X-ray, manyetik rezonans görüntüleme ve bilgisayarlı tomografi ile gösterilmiştir. Büyük osteofitlerin cerrahi olarak çıkarılması ve kısa süreli nonsteroid anti-inflamatuar ilaç tedavisi disfajinin giderilmesi ile sonuçlanmıştır. Bu nadir durumun klinik bulguları, tanı ve tedavisi tartışılmıştır.

ANAHTAR SÖZCÜKLER: DISH, Disfaji, Hiperostoz, Omurga, Osteofit

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INTRODUCTION

Various local structural lesions such as oropharyngeal tumors, vascular pathologies, retropharyngeal abscesses, and anterior cervical osteophytes may lead to mechanical esophageal dysphagia (1-4, 7, 8). Anterior cervical osteophytes occur in 20-30% of the elderly population and generally remain asymptomatic (3). Although rare, patients with anterior cervical osteophytes may complain of dysphagia particularly when the osteophytes are extraordinarily large (4). Large anterior osteophytes of the cervical spine may also be seen in diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier's disease (1). 17% of patients with DISH have been reported to be characterized by dysphagia (4).

In this case report, we describe a surgically treated 54-year-old woman with dysphagia caused by DISH.

CASE REPORT

A 54-year-old woman referred to our clinic because of progressive dysphagia only for solids and cervical pain for 5 years. There was no previous history of trauma to the neck. She had no history of systemic disease. She had been seen by the gastroenterology, otolaryngology and psychiatry clinics and given many medications including antidepressants and anxiolytics.

A physical examination was unremarkable. Examination of the neck revealed no goitre. There was decreased range of spinal motion in the cervical region. The neurological examination was normal. Routine blood tests were normal. Previous investigations including fibre-optic endoscopy of the pharynx and esophagus, and neck magnetic resonance imaging (MRI) were normal. A lateral cervical spinal X-ray and cervical MRI revealed giant cervical osteophytes at the ventral portion of the C 2/3/4 vertebral bodies and contiguous calcification of the anterolateral cervical vertebra bodies (Figure 1A,B). Reconstruction of the cervical spine computed tomography (CT) showed external compression of the pharyngoesophageal segment by the large osteophytes (Figure 2).

Surgery was performed with an anterolateral approach. The prevertebral fascia was separated and the trachea and esophagus were retracted on the left side and the carotid sheath on the right. The C2-4 level was exposed and identified by lateral scope

examination. Large anterior osteophytes were removed with Kerrison rongeurs and high speed air drill until the anterior spinal surface from C2 to C4 was flat on palpation and on lateral scope control (Figure 3). A nonsteroidal anti-inflammatory drug (NSAID) regimen of naproxen sodium 275 mg twice daily was given for 2 weeks. The patient was discharged on NSAID and without any complication.

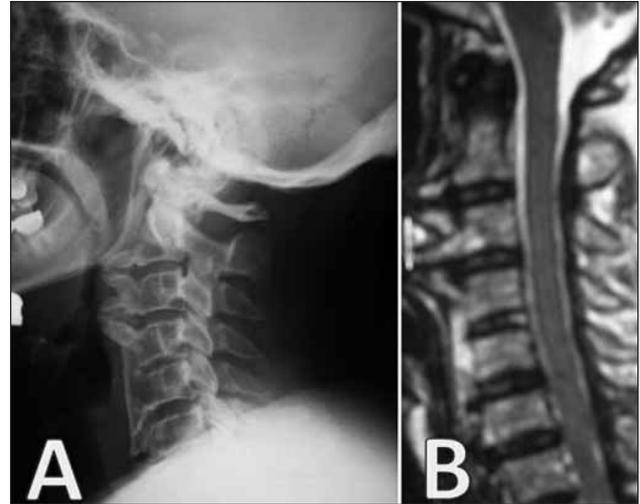


Figure 1: Lateral cervical spinal X-ray (A) and a T2-sagittal cervical MRI (B) revealed giant cervical osteophytes at the ventral portion of C 2-4 and contiguous calcification of the anterolateral cervical vertebral bodies.

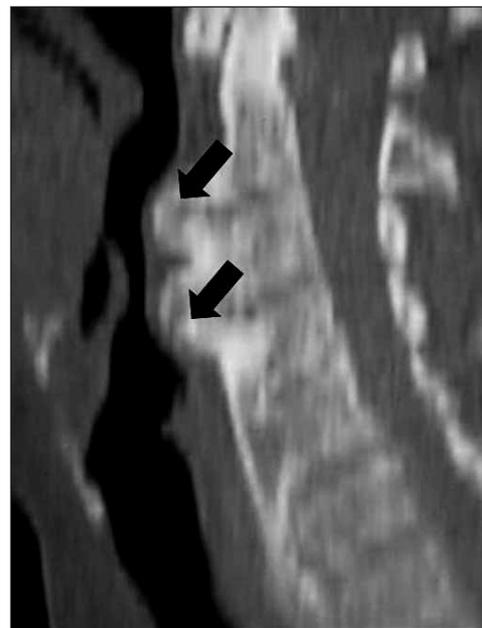


Figure 2: Pre-operative reconstruction of cervical spine computed tomography shows external esophageal compression by the large osteophytes (Arrows).



Figure 3: Post-operative lateral cervical spinal X-ray shows successful removal of the large osteophytes.

DISCUSSION

Large anterior cervical osteophytes and/or DISH may lead to progressive dysphagia. The possible mechanisms of dysphagia include mechanical compression to esophagus, pharyngo-esophageal irritation which induces peri-esophageal edema, inflammation, and a local inflammatory reaction resulting in cricopharyngeal spasm and esophageal denervation (5-7).

Lateral cervical radiography is important for diagnosis in patients with DISH and/or cervical osteophyte-induced dysphagia. Cervical MRI may show additional pathologies including cervical spondylosis. Although esophagoscopy is important to rule out pathologies causing intrinsic compression, it must be performed very carefully because of the risk of esophageal perforation during the esophagoscopy procedure in patients with cervical osteophytes (9). A lateral neck radiograph during barium swallow (barium esophagograms) may demonstrate narrowing of the esophagus due to the vertebral osteophytes (4). We could not perform esophagogram because the patient refused to swallow barium. Hence, we performed reconstruction of the cervical spine computed tomography to show external esophageal compression by the large cervical osteophytes (Figure 2).

Dysphagia caused by cervical osteophytes may be treated conservatively or surgically (4, 7, 8, 10). Patients may need surgery when the osteophytes are remarkably large. Although in many DISH cases treatment may not be necessary, in the presence of dysphagia surgical removal of the large osteophytes should be considered. We treated our patient surgically due to the large size of the cervical osteophytes. Removal of only large cervical osteophytes, rather than multilevel anterior resection of DISH, resulted in rapid and progressive resolution of symptoms within 2 months. NSAID may be helpful in resolving peri-esophageal inflammation following surgical treatment as seen in our case.

In summary, DISH should be suspected in older patients presenting with progressive dysphagia. Reconstruction of cervical spine CT may be helpful in demonstrating the external esophageal compression. In such cases, surgical removal of only large osteophytes combined with NSAID therapy may relieve symptoms.

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