Ege Tıp Dergisi / *Ege Journal of Medicine* 2012;51(4):259-261

Diffuse idiopathic skeletal hyperostosis (DISH): A rare cause of dysphagia

Disfajinin nadir bir sebebi: Diffüz idiopatik skeletal hiperosteozis (DISH)

Kandogan T[2] Sezgin Ö[3] Dalgiç A[1]

[1]Reyhanlı Devlet Hastanesi, Kulak Burun Boğaz Kliniği, Hatay, Türkiye
[2]İzmir Eğitim ve Araştırma Hastanesi, Kulak Burun Boğaz Kliniği, İzmir, Türkiye
[3]İzmir Eğitim ve Araştırma Hastanesi, Radyoloji Kliniği, İzmir, Türkiye

Summary

Diffuse idiopathic skeletal hyperostosis is a relatively common but often unrecognized systemic disorder observed mainly in the elderly. It can lead to back pain, dysphagia, myelopathy, musculoskeletal impairment and grossly unstable spine fractures after minor trauma, but most patients are free of symptoms, so that it is usually discovered fortuitously upon plain radiographs of the spine obtained for another reason. Here we reported a case in which the diagnosis of DISH was made upon evaluation for dysphagia.

Key Words: Diffuse idiopathic skeletal hyperostosis, dysphagia, elderly, Forestier disease.

Özet

Diffüz idiopatik iskelet hiperosoteozisi rölatif olarak nadir olan ve daha çok yaşlılarda görülen tanımlanmamış sistemik bir hastalıktır. Hastalık sırt ağrısı, disfaji, miyelopati, muskuloskeletal sistem zayıflığı gibi semptomlara ve küçük travmalar sonrasında omurgada büyük fraktürlere neden olabilir. Hastalık çoğunlukla asemptomatik seyreder ve bu yüzden genellikle başka nedenle yapılan radyolojik tetkiklerde ortaya çıkar. Biz bu makalede disfaji nedeni ile değerlendirdiğimiz ve diffüz idiopatik iskelet hiperosteozis tanısını koyduğumuz bir olgu sunduk.

Anahtar Sözcükler: *Diffüz idiopatik skeletal hiperosteozis, disfaji, yaşlılık, Forestier hastalığı.*

Introduction

Diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier disease was first described in 1950 by Forestier (1). It is a relatively common but often unrecognized systemic disorder observed mainly in the elderly (2). DISH is of unknown etiology, characterized by exuberant hyperostosis of the antero-lateral aspect of the spinal column, that sometimes leads to bone ankylosis, and by ossification of extra-spinal entheses (3). Flowing calcification along the anterior and lateral sides of the vertebral bodies produces the appearance of candle wax dripping down the spine (1).

DISH can lead to back pain, dysphagia, myelopathy, musculoskeletal impairment and grossly unstable spine fractures after minor trauma, but most patients are free of symptoms, so that DISH is usually discovered fortuitously upon plain radiographs of the spine obtained for another reason (1,2).

Yazışma Adresi: Tolga KANDOĞAN
İzmir Eğitim ve Araştırma Hastanesi, Kulak Burun Boğaz Kliniği, İzmir, Türkiye
Makalenin Geliş Tarihi: 02.05.2011 Kabul Tarihi: 23.06.2011

Here we reported a case in which the diagnosis of DISH was made upon evaluation for dysphagia.

Case Report

A 63 year-old man was referred to an otorhinolaryngology outpatient clinic for dysphagia. Past medical history was unremarkable. Dysphagia symptoms had worsened gradually over the last 4 months. Endoscopy of the pharynx and larynx showed a protuberance in the posterior pharyngeal wall at the level of hypopharynx.

Computed tomography (CT) scans confirmed the anterior ossification extending from C3 to C6 and impinging on the hypopharynx (Figure-1a,b). Nonsteroidal anti-inflammatory agents in combination with a muscle relaxant was used to relieve the dysphagic symptoms of the patient and he reported being free from dysphagia after 3 months.

Discussion

The exact etiology of DISH is unclear. However it is more common in type 2 diabetes. Among other possible factors, elevated levels of insulin growth factor or elevated levels of growth hormone may play a part in the

new bone formation (2-5). Primary hypertension, its cardiovascular effects and lithiasis are also often present in these patients (2,3).

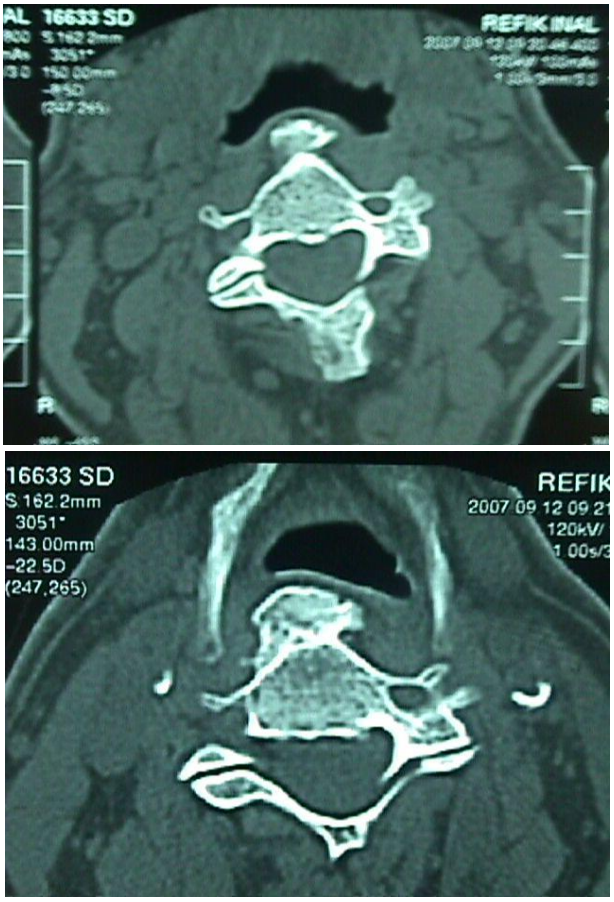

Figure-1.a-1b. It is seen anterior ossification extending from C3 to C6 and impinging on the hypopharynx in the CT.

It is reported that 2-4% of the general population may have hyperostotic changes and these changes are present in approximately 25% of the diabetic population (4). One half of all patients with DISH will have manifest diabetes or impaired glucose tolerance. DISH may be associated with obesity, hyperlipidemia and hyperuricemia, in addition to its association with diabetes. It does not show an association with the duration of diabetes (4). DISH occurs more commonly in males (65%) than in females (35%) (6). The reported patient is non-diabetic, non-hypertensive and slim. The blood levels of lipid and urea are within normal limits, which make this case interesting.

DISH diagnostic criteria are as follows: 1-Flowing calcifications and ossifications along the anterolateral aspect of at least 4 contiguous vertebral bodies with or without osteophytes. 2-Preservation of disc height in the involved areas and an absence of excessive disc disease. 3-Absence of bony ankylosis of facet joints and absence of sacroiliac erosion, sclerosis, or bony fusion, although narrowing and sclerosis of facet joints are acceptable (6).

Dysphagia is a common presenting complaint in otolaryngology practice, and there are many causes. DISH is a rare cause of dysphagia (7). The proportion of cases revealed by dysphagia has ranged across studies from 0.1% to 28% (1). The dysphagia is usually marked, present for solid foods, improved by anterior flexion of the neck, and worsened by extension of the neck. Concomitant symptoms may include a foreign body sensation, odynophagia, reflex otalgia, salivary stasis, sleep apnea, and aspiration (1). In addition to dysphagia, DISH has been reported to cause laryngeal stridor, dyspnea, snoring and hoarseness. Other important symptoms associated with DISH are stiffness and pain in the back, pain related to tendinitis, myelopathy related to core compression associated with the ossification of the posterior longitudinal ligament, and pain related to vertebral complications such as fracture or subluxation (7,8).

In DISH, patients are frequently asymptomatic, but may have stiffness of joints and radiological changes are generally much more severe than might be predicted from the symptoms. The most commonly affected vertebrae are the lumbar spine. The cervical spine is less involved (4).

The conditions that can mimic DISH are ankylosing spondylitis or cervical spondylosis (4). DISH is distinct from ankylosing spondylitis in which the involvement of the sacroiliac joint excludes DISH. DISH also is distinct from marginal osteophytes that form in response to degenerative disk disease. Patients with DISH infrequently demonstrate disk height reduction or vacuum changes (6).

The management includes three components, physical, pharmacological, and surgical. Physical therapy may improve range of motion in patients with spinal stiffness. Nonsteroidal anti inflammatory agents or glucocorticoid bolus therapy in combination with a muscle relaxant can be used to improve the symptoms. Dietary precautions aimed at minimizing gastroesophageal reflex are in order in patients with dysphagia. Spinal pain may require analgesic therapy. Surgery is needed to prevent aspiration pneumonia in patients with severe dysphagia related to a markedly prominent ossification. Anterior decompression with stabilization by an interbody cage can be performed. A concomitant space-occupying pharyngeal lesion should be looked for before initiating the treatment of DISH (1).

Although it is a rare cause of dysphagia, especially in the elderly dysphagic patients, DISH also should be considered as a possible cause of dysphagia.

References

1. Dioré K, Facon D, Marsot-Dupuch K, Bobin S. An underrecognized cause of dysphagia: Forestier's disease. French ORL 2005;(88):124-7.
2. Verlaan JJ, Oner FC, Maat GJ. Diffuse idiopathic skeletal hyperostosis in ancient clergymen. Eur Spine J 2007;16(8):1129-35.
3. Colina M, Govoni M, De Leonardis F, Trotta F. Diffuse idiopathic skeletal hyperostosis (DISH). Reumatismo 2006;58(2):104-11.
4. Ghosh B, Kishore S, Vijay V, Ramachandran A. JAPI 2004; (52):994-6.
5. Kiss C, Szilagyi M, Pasky A, Poor G. Risk factors for diffuse idiopathic skeletal hyperostosis: A case-control study. Rheumatology 2002; 41(1):27-30.
6. Nakhoda K, Greene G. Imaging in diffuse idiopathic skeletal hyperostosis [Updated: Feb 22, 2012]. Available from: *http://emedicine.medscape.com/article/388973-overview*.
7. Aydin E, Akdogan V, Akkuzu B, Kirbas I, Ozgirgin ON. Six cases of Forestier syndrome, a rare cause of dysphagia. Acta Otolaryngol 2006;(126):775-8.
8. Altay M, Sahin M, Atabay M, ve ark. Diyabetiklerde diffüz idiyopatik skeletal hiperostozis gelişimi artmış mıdır? Türkiye Klinikleri J Med Sci 2006;26(1):15-9.