

Olgu Sunumu

Diffuse Idiopathic Skeletal Hyperostosis Causing Dysphagia Due to a Giant Cervical Osteophyte

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Diffuse idiopathic skeletal hyperostosis (DISH) is a common disease in which tendons and capsules are subject to calcification leading to hyperostosis and functional impairment. Diagnosis of DISH is established by the presence of both spinal and extra-spinal radiographic characteristics. Typical appearance of the cervical spine includes irregular and pointed osseous osteophytic appositions of the superior and inferior vertebral margin.

Large anterior osteophytes of the cervical spine are common in diffuse idiopathic skeletal hyperostosis (DISH) and 17% of patients with DISH have been reported to describe some levels of dysphagia. In this case report, we describe a surgically treated 88-year-old man who presented with dysphagia and painful swallowing caused by a huge anterior cervical osteophyte due to DISH. The osteophyte was removed surgically and dysphagia resolved immediately.

Key words: DISH, osteophyte, cervical, dysphagia

J Nervous Sys Surgery 2014; 4(3):147-152

Diffüz İdyopatik Skeletal Hiperostozda Dev Servikal Osteofit ve Disfaji

Diffüz idyopatik skeletal hiperostoz (DISH) tendon ve eklem kapsüllerinin kalsifikasyona maruz kaldığı hiperostoz ve işlevsel bozukluğa neden olan yaygın bir hastalıktır. DISH tanısı hem spinal hem extra-spinal karakteristik radyolojik bulgularla konur. Servikal omurganın tipik görüntüsünde süperior and inferior vertebral yüzeylerde osteofit şeklinde birbirine karşılıklı olarak büyüyen düzensiz osseöz çıkıntılar ortaya çıkar.

Büyük anterior servikal osteofitler diffüz idyopatik skeletal hiperostozda sık görülür ve DISH tanısı konmuş hastaların %17'si belirli bir derecede disfaji tanımlar.

Biz bu bildiride, DISH tanısı konan ve dev bir anterior servikal osteofitin neden olduğu disfaji ve ağırlı yutkunma yakınmasıyla başvuran 88 yaşında bir erkek olguyu sunuyoruz. Hastanın osteofiti cerrahi olarak tedavi edildi ve disfajisi postop dönemde dramatik olarak düzeldi.

Anahtar kelimeler: DISH, osteofit, servikal, disfaji

J Nervous Sys Surgery 2014; 4(3):147-152

† Where applicable, all procedures were followed in accordance with the responsible committee on human experimentation and with the Helsinki Declaration of 1975 and subsequent revisions. Informed consent was received from the patient involved in this case study.

Alındığı tarih: 06.04.2015

Kabul tarihi: 11.08.2015

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Diffuse idiopathic skeletal hyperostosis (DISH) is a common disease in which tendons and capsules are subject to calcification leading to hyperostosis and functional impairment. Diagnosis of DISH is established by the presence of both spinal and extra-spinal radiographic characteristics. Typical appearance of the cervical spine includes irregular and

pointed osseous appositions of the superior and inferior vertebral margin ⁽⁹⁾.

Large anterior osteophytes of the cervical spine may be seen in diffuse idiopathic skeletal hyperostosis (DISH) and 17% of patients with DISH have been reported to describe some levels of dysphagia ^(1,5). However, anterior cervical osteophytes are common and occur in 20-30% of the normally aging elderly population and generally remain asymptomatic ⁽⁴⁾. Although rare, patients with anterior cervical osteophytes may complain of dysphagia particularly when the osteophytes are extraordinarily large ⁽⁵⁾. Also, various local structural lesions such as oropharyngeal tumors,

vascular pathologies, retropharyngeal abscesses, and anterior cervical osteophytes may lead to mechanical esophageal dysphagia ^(1,5,12,15).

In this case report, we describe a surgically treated 88 year old man with dysphagia caused by a huge anterior cervical osteophyte due to DISH.

CASE REPORT

An 88-year-old male patient presented at our neurosurgical outpatient clinic with complaints of dysphagia and pain in his throat radiating to his face whenever he attempted to swallow solid food. His pain and difficulty in swallowing had



Figure 1. Preoperative study of the patient's spine: A giant osteophyte is noted anteriorly at the cervical spine. Continuous calcification of anterior longitudinal ligament throughout the thoracic spine was detected. Study of the sacroiliac joints revealed no involvement suggesting the diagnosis of DISH. Ossification of the prostatic gland was a coincidental finding.

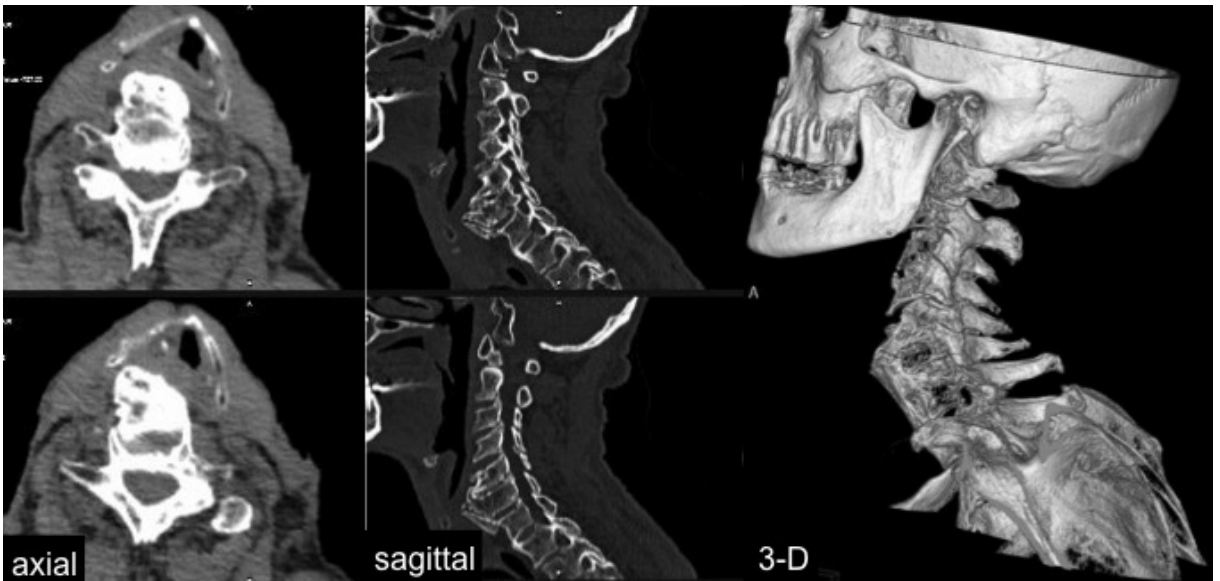


Figure 2. Preoperative Cervical CT scan.

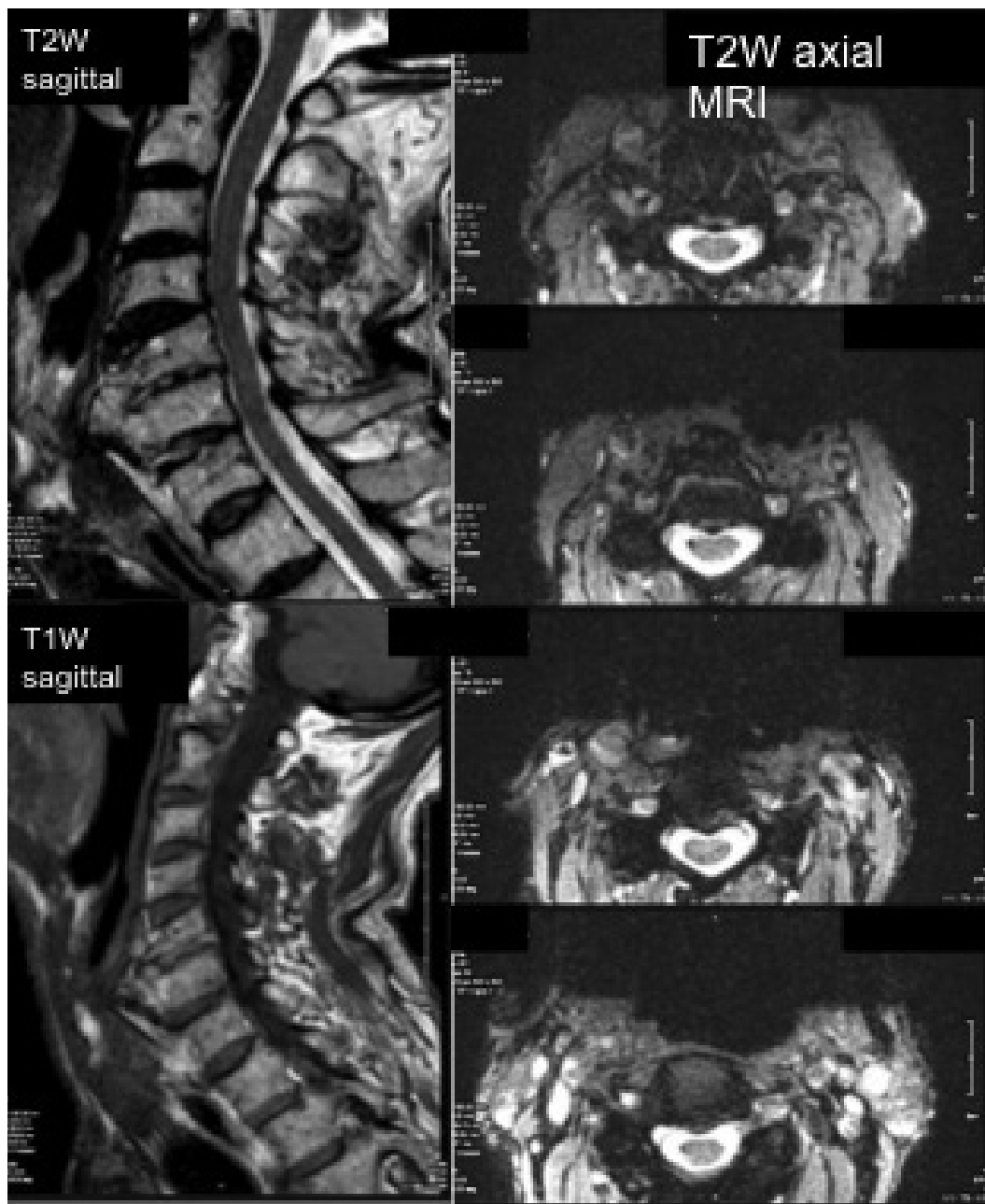


Figure 3. Preoperative MRI of the cervical spine. No compromise of neural tissue is present.

been gradually increasing for the last 3 years. He also reported a neck stiffness and hard mass in his throat that he was aware of for the last several years. On close inspection, a lateral deviation of the trachea to the left was noticeable. Also, a non-tender stone-hard mass was palpable anterior to his cervical spinal column at the level of C5-6 on the right side of trachea. No motor or sensory deficits were noted on his neurological ex-

amination. Routine blood tests were also normal. Plain radiographs of the cervical spine showed extensive ossification at the anterior longitudinal ligament along the anterolateral aspect of the vertebral bodies from C3 to T1, especially prominent at C5-6 level, suggesting DISH. Cervical computed tomography (CT) confirmed the giant anterior osteophyte (Figure 1-2) and showed continuous anterior longitudinal calcification

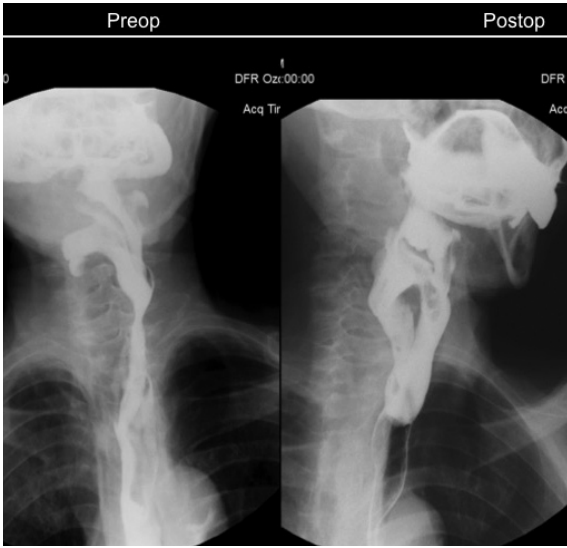


Figure 4. Barium swallow esophagography, postop graph shows the resolution of the filling defect seen in the preop graph.

culminating in a giant osteophyte at the C5-6 level and continuing over C7 and extending into the thoracic spine (Figure 2). Osteophytic multiple bony spurs were also noted in the radiographs and CT of the thoracic and lumbar spine. MRI of the cervical spine confirmed the presence of the giant anterior osteophyte and revealed its relation the ossified anterior longitudinal ligament as it extended into the thoracic column. MRI also showed no involvement of the posterior longitudinal ligament, some intervertebral disk protrusions and ligamentum flavum hypertrophy with

no neural compromise. There were no definite signs of instability in the cervical spine (Figure 3). Barium contrast swallow cinematography revealed a filling defect along the pharynx with lateral displacement of the esophagus at the level of the anterior osteophyte (Figure 4).

The patient underwent surgery with a left-sided antero-lateral cervical approach. The pre-vertebral fascia was separated and the trachea and esophagus were retracted on the left side, with the carotid sheath on the right side. The C3-7 vertebrae were exposed. The giant anterior osteophyte was removed with rongeurs and a high speed drill until the anterior spinal surface from C3 to C7 was flat. A postoperative CT of the cervical spine demonstrated removal of the anterior cervical osteophyte and normalization of the anterior cervical curve (Figure 5).

Dysphagia improved immediately following surgery. The postoperative barium contrast swallow cinematography demonstrated resolution of the filling defect in the esophagus and no fistulas were detected that might have occurred during surgical dissection (Figure 4). The patient was discharged without any complications the following day.



Figure 5. Intraoperative view of the osteophyte formation. Tracheal deviation is notable. Also, please note the sparing of the intervertebral disk space.

DISCUSSION

DISH is a clinical syndrome characterized by the abnormal formation of osteophytes involving the spine, with ossification of the paraspinal muscles and ligaments. This condition was originally described by Forestier and Rotes-Querol in 1950 as a calcification of anterior longitudinal ligaments⁽³⁾. Subsequently, in 1975, Resnick defined the acronym of “DISH” to describe the syndrome⁽¹⁰⁾. Two years later, together with Niwayama, he also encoded the diagnostic criteria for Forestier’s disease⁽¹⁰⁾.

These included: 1. Longitudinal anterior ligament calcification involving at least four contiguous vertebrae; 2. Absence of degenerative changes of the affected disks; 3. Absence of ankylosis of the joints and sacroiliac arthrosis phenomena. Among the abnormal conditions occurring in patients suffering from DISH, the elevated levels of insulin-like growth factor-I and hyperinsulinemia are observed. Dickkopf-1 (DKK-1), a recently identified inhibitor of osteoblast differentiation, may be a key player in the formation of osseous appositions⁽¹¹⁾.

DISH is a common condition in the aging spine, often associated with large anterior osteophytes of the cervical spine, which may cause symptoms⁽⁶⁾. It is estimated that 3% of individuals over the

age of 40 have DISH and 0.1-6% of those will develop dysphagia⁽²⁾. The possible mechanism of dysphagia is mostly mechanical compression to esophagus. However pharyngo-esophageal irritation, which induces peri-esophageal edema, inflammation, and a local inflammatory reaction resulting in cricopharyngeal spasm and esophageal denervation, have also been suggested^(7,8,12).

Lateral cervical radiography and CT imaging are important for diagnosis in patients with DISH and/or cervical osteophyte-induced dysphagia. An ossified anterior longitudinal ligament continuously stretching over at least four vertebrae is a prerequisite for diagnosis. Cervical MRI may show additional pathologies in the soft tissues including cervical spondylosis and is helpful in surgical planning. 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⁽¹⁶⁾. A lateral neck radiograph during barium swallow esophagogram is generally adequate to demonstrate narrowing of the esophagus due to the vertebral osteophytes⁽⁵⁾. In our case, we repeated the procedure postoperatively both to demonstrate the correction of the esophageal passage as well as to rule out esophageal fistulas that might have developed during the surgical procedure.



Figure 6. Postoperative CT scan of the cervical spine. Normal cervical anterior curvature is achieved.

The cervical osteophytes in most DISH patients may be treated conservatively as they are asymptomatic. However, in the presence of dysphagia, surgical removal of the large osteophytes should be considered. In our case, the patient also complained of severe throat pain radiating to his face when he attempted to swallow solid food. Therefore the surgical decision was made to provide relief for both the pain and dysphagia.

Surgical resection of the osteophyte has been reported to be an effective treatment for severe cases and/or cases with airway obstruction^(7,13,14,16). Similar to our case, most cases are treated by resection of the osteophyte without spinal fusion. Avoiding spinal fusion is advantageous due to no implant-associated complication and less operation time as some of these patients present at advanced age with other associated medical conditions. On the other hand, recurrence of the osteophyte formation may occur and presence of postoperative intervertebral mobility was reported to pose a significant risk for recurrent formation of osteophytes⁽¹⁴⁾. Therefore, intersegmental mobility in the cervical spine should be considered during surgical planning, especially in relatively younger patients who are expected to have longer life expectancy. Similar to our patient, dramatic improvement in dysphagia soon after surgery can be expected in most cases with relief of the mechanical obstruction on the esophagus. A slower resolution could be due to more gradual abatement of esophageal inflammation or spasm.

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