

A RARE CAUSE OF DYSPHAGIA THAT IS TREATABLE WITH SURGERY: A SINGLE-CENTER CASE SERIES OF FORESTIER'S DISEASE

DİŞFAJİNİN CERRAHİ İLE TEDAVİ EDİLEBİLİR NADİR BİR SEBEBİ: FORESTIER HASTALIĞININ TEK MERKEZLİ VAKA SERİSİ

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ABSTRACT

Objective: Diffuse idiopathic skeletal hyperostosis (DISH), Forestier's disease, is a systemic disease characterized by the abnormal ossification and calcification of the paravertebral ligaments and muscles in front of the vertebral bodies. It rarely causes clinical symptoms but sometimes produces dysphagia. This study will evaluate the effectiveness of the anterior cervical approach in patients with Forestier's disease who suffer from dysphagia.

Material and Method: In this series, a retrospective analysis was performed on five patients referred to our clinic with dysphagia. Clinical, demographic, and surgical features were evaluated.

Result: All of the cases were male. Their mean age was 63.6 (range 56-69). The involved regions were C3-4; C2-3; C6-7; C3-7; C3-D3. All the patients who have dysphagia were treated via the anterolateral cervical approach. There were no neurological deficits or complaints postoperatively. All the patients stated that they benefited from surgery.

Conclusion: Osteophyte resection by an anterolateral cervical approach is a safe and effective treatment option for dysphagia in Forestier's disease.

Keywords: Cervical spine, diffuse idiopathic skeletal hyperostosis, dysphagia, osteophyte

ÖZET

Amaç: Difüz idiopatik iskelet hiperosteozu (DİİH), Forestier hastalığı, vertebra korpuslarının önündeki paravertebral ligaman ve kasların anormal kalsifikasyonu ile karakterize sistemik bir hastalıktır. Klinik semptomlara nadiren sebep olurlar ve bu semptomlar arasında disfaji bulunmaktadır.

Gereç ve Yöntem: Bu vaka serisinde, kliniğimizde disfaji ile prezente olan beş DİİH tanılı hasta retrospektif olarak incelenmiştir. Klinik, demografik ve cerrahi bilgiler hastane kayıtlarından elde edilerek değerlendirilmiştir.

Bulgular: Tüm vakalar erkek ve ortalama yaş 63,6 (56-69) yılıdır. Etkilenen bölgelerin C3-4; C2-3; C6-7; C3-7; C3-D3 olduğu saptandı. Tüm vakalarda anterolateral servikal yaklaşım uygulandı. Postoperatif yeni şikayet ve nörolojik defisit ile karşılaşılmadı. Tüm hastalar disfaji şikayetlerinin ameliyattan sonra gerilediklerini belirttiler.

Sonuç: Anterolateral servikal yaklaşım Forestier hastalığında vertebra korpus anteriorunda bulunan osteofit rezeksiyonunda uygun ve güvenli bir seçenektir.

Anahtar Kelimeler: Difüz idiopatik iskelet hiperosteozu, disfaji, osteofit, servikal vertebra

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INTRODUCTION

Diffuse idiopathic skeletal hyperostosis (DISH), a Forestier's disease, is a systemic disease characterized by the abnormal ossification and calcification of the paravertebral ligaments and muscles in front of the vertebral bodies. Forestier and Rotes-Querol first described the disease in 1950 (1,2).

The etiology is not well known. Ninety-seven percent of the cases occur in the thoracic spine, in the lumbar spine 90%, in the cervical spine 78%, and in all three segments 70%. Forestier's disease may be seen in all genders and races but usually in patients in their 60s (3). It rarely causes clinical symptoms but sometimes leads to dysphagia, pain, stiffness, reduced articular motion, and dysphonia (4-6).

Forestier's disease is managed conservatively. Surgical resection through the anterior approach provides resolution of the dysphagia (7). The study reports on five cases of dysphagia due to the osteophytes of the cervical levels.

MATERIAL and METHODS

Study design

In this series, a retrospective analysis was performed on patients with Forestier's disease referred to our clinic with dysphagia between 2005-2021. The minimum follow-up period was 12 months.

Patient population

This study included five patients who underwent anterolateral cervical resection of osteophytes to treat dysphagia caused by Forestier's disease. The diagnosis was made according to the criteria suggested by Resnick and Niwayama (8). All the patients exhibited dysphagia persisting for more than 12 months.

Radiological evaluation

In all the patients, lateral plain radiographs showed anterior osteophytes. All the patients were evaluated by X-rays, computerized tomography (CT) scanning, and magnetic resonance imaging (MRI) of the cervical and thoracic vertebrae.

Surgical technique

The patient is positioned with a shoulder support pillow enabling a 15 degree extension of the neck. Anteroposterior (AP) and lateral X-rays are used for determining the level and planning the incision. The incision is made longitudinal and parallel to the medial border of the sternocleidomastoid (SCM) muscle. The longitudinal incision allows a broader exposure of the cervical spine. After incising the platysma muscle, the medial border of the SCM muscle is determined and retracted laterally. Additional attention is paid to palpating the carotid artery and retracting it laterally under the SCM muscle. The esophagus is carefully dissected and retracted medially,

avoiding any further compression. A marker is placed, and X-ray control of the levels is achieved. Once the correct levels are identified, and osteophytes are exposed, the high-speed drill is used to drill the osteophytes. Care should be taken not to disrupt the vertebral bodies and the annulus fibrosus. The lateral extent of the osteophytes can be removed. Bone bleeding is handled with bone wax. Soft tissues are spared through the procedure. After adequate decompression, a drain can be left in the surgical site. The wound is closed in layers.

Postoperative period and follow-up evaluation

On the first postoperative day, the patients were mobilized, and the drain was removed. Oral intake was begun in the 6th hour. As the symptoms resolved, the patient was put on a regular diet. A cervical collar was not required. Patients were evaluated in the outpatient clinic for one week, one month, six months, and one year after surgery. AP and lateral X-rays, CT scans, and MRI were obtained at each visit (Figure 1). This study was approved by Istanbul Faculty of Medicine Clinical Research Ethics Committee (Date: 03.02.2023, No: 03).

RESULTS

All of the cases were male, and the mean age was 63.6 (range 56-69). In the five patients included in the study, the involved regions were C3-4, C2-3, C6-7, C3-7, and C3-D3. All the patients who have dysphagia were treated via the anterolateral cervical approach. A longitudinal cervical incision along the SCM muscle allowed adequate pathology exposure even for more laterally extending osteophytes. The mean operation duration was 132 minutes (102-156). The surgery was feasible for all cases.

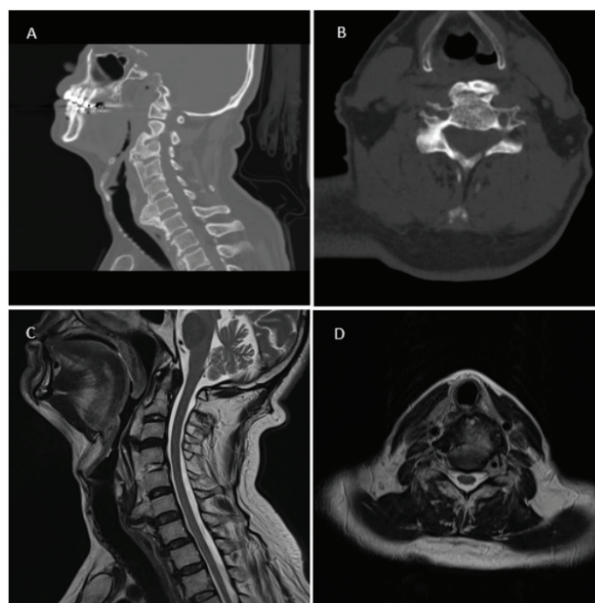


Figure 1: Preoperative CT (A, B) and MRI (C, D) scans of a patient with DISH presented with dysphagia

The total removal of the osteophytes was appreciated in postoperative imaging (Figures 2 and 3). There were no neurological deficits or complaints postoperatively. All the patients stated that they benefited from the surgery.

The minimum follow-up period was 12 months, with an average of 17 months. The patients tolerated the diet during their hospital stay. However, dysphagia resolved completely after two weeks. Complete removal of the osteophytes was seen in early postoperative radiographs. Radiographs at the first-year follow-up showed minimal regrowth of the osteophytes in all patients, but the patients had no symptoms. There were no instability findings.

DISCUSSION

DISH is a rare condition characterized by paravertebral ligament calcification and ossification in the ante-

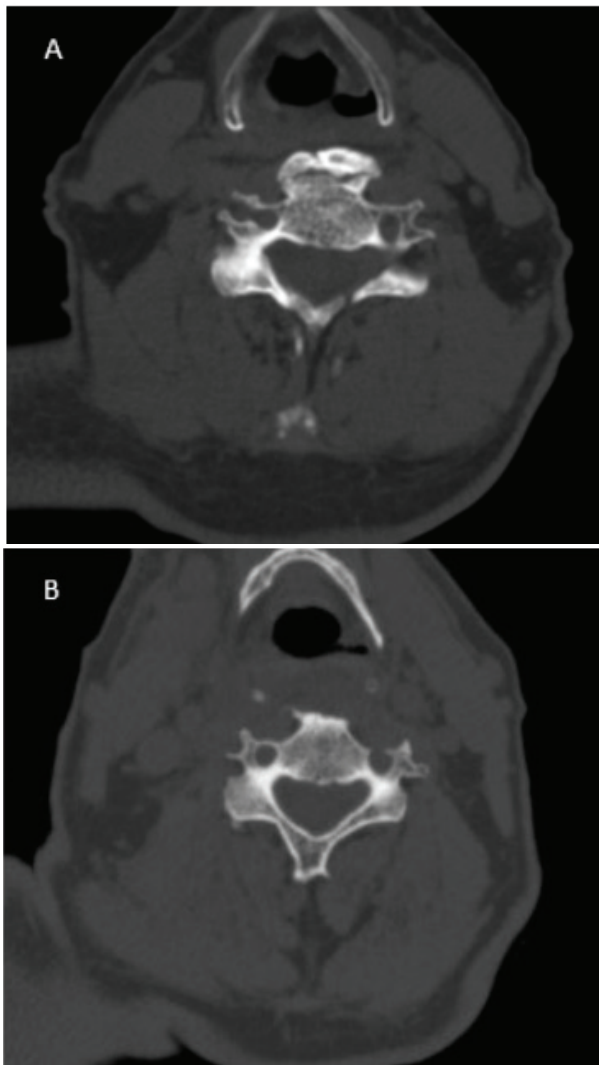


Figure 2: Preoperative (A) and postoperative (B) axial CT scans show total removal of anterior cervical osteophyte

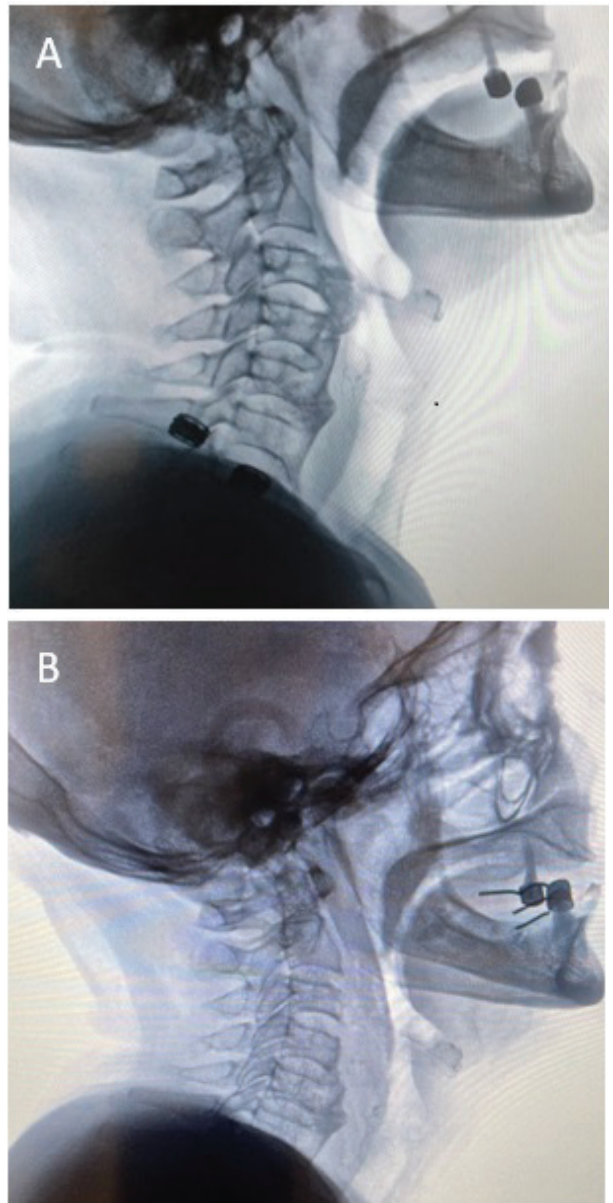


Figure 3: Lateral cervical X-ray of a patient with diffuse idiopathic skeletal hyperostosis (A). Same patient after undergoing anterolateral cervical osteophyte resection (B)

rior parts of the vertebral bodies. It may be seen in all genders, but the frequency is higher in males, and the incidence increases with age (9,10). It is more common in Mediterranean countries, especially Türkiye and Italy (11). The etiology is not known clearly. Mechanical factors such as the proximity of the aorta causing bony bridging, genetic factors, environmental exposures like fluoride and retinol, and drugs like vitamin A derivatives are some of the culprits (12). Recent studies also discovered the correlation between Forestier's disease and various metabolic disorders such as obesity, hyperlipidemia, hyperinsulinemia, and hypertension (13).

The radiological diagnostic criteria suggested by Resnick and Niwayama for diffuse idiopathic skeletal hyperostosis: the presence of following ossification of the anterior longitudinal ligament of at least four contiguous vertebral bodies, the preservation of intervertebral disc height, and the absence of other signs of degenerative spine joint diseases (8). The differential diagnosis of dysphagia includes tumors of the larynx, pharynx, mediastinum, spine, lung or esophagus, esophageal motility disorders, esophageal strictures, aberrant vessels, and esophageal diverticula. These pathologies can be masked by DISH and should be considered a possible coexistent pathology (14).

It is primarily seen in the C4-7 level in the cervical region (9). In the reviewed, the involved regions were C3-4, C2-3, C6-7, C3-7, and C3-D3. The most frequent symptoms seen in Forestier's disease are neck rigidity, pain, hoarseness, stridor, and dysphagia (15–17). Dysphagia is encountered in 0.6-1% of patients diagnosed with Forestier's disease. Dysphagia appears especially in lower cervical levels, mainly at C4-5 (7,18,19). Case 2 is a rare situation representing an upper cervical area, C2-3. The dysphagia results from pharyngeal and esophageal compression and inflammation in paravertebral and paroesophageal tissues. In addition, stridor, dyspnea, and aspiration may be seen in Forestier's disease (20). Extraspinal involvement, such as the pelvis, calcaneus, olecranon, and patella (20).

Computed tomography (CT) is the gold standard in diagnosis (8). The relationship between osteophytes and adjacent anatomical structures may be shown clearly. In the reviewed cases, a CT scan was used to show that compression on the esophagus causes dysphagia. We also used esophagoscopy, X-rays, and an MRI for evaluation. Some clinicians suggest using endoscopy, but this might be dangerous and has been a cause of esophageal perforation since esophageal wall can be thinned in involved locations (21,22). In our experience, these imaging modalities are sufficient to evaluate the bony structures and soft tissue compression, especially in the esophagus and trachea. The anterior osteophytosis usually involves multiple vertebral levels, but the compression and the related symptoms might not be in all levels.

In DISH cases, conservative treatments are recommended if the patient does not have symptoms and neurological deficits. Nonsteroid anti-inflammatory drugs, steroids, and myorelaxants are conservative treatment options. Surgical treatment can be an option in cases with progressive dysphagia and dyspnea (23–25). In the reviewed cases, an anterolateral cervical approach was used, providing adequate space for removing the osteophytes. There were no findings of instability, such as spondylolysthesis or fractures. In cases of suspected instability, this

approach also allows for applying anterior fusion. Other case series also showed the effectiveness of this approach regarding dysphagia in Forestier's disease. A case series study with nine patients showed the resolution of symptoms in all patients after the anterolateral cervical approach (26). In our experience, osteophylectomy at the symptomatic level is sufficient for patient satisfaction and produces less exposure and inflammation in the postoperative period. A case series of nine patients suggested osteophylectomy in the symptomatic segment, consistent with this study (20). Different approaches for osteophyte resection in Forestier's disease are described, such as the per-oral-transpharyngeal approach (27).

Complications like fistula formation, recurrent laryngeal nerve injury, and instability can occur. Infections in this region by oropharyngeal flora could be a devastating complication, especially if osteomyelitis occurs (28,29). In the presented cases, there were no significant complications. The anterolateral cervical approach is a feasible technique for osteophyte removal and provides symptom relief caused by compression.

CONCLUSION

Forestier's disease presenting with dysphagia is very rare. Surgery may be considered when the symptoms are severe and progressive. Osteophyte resection via an anterolateral cervical approach is a safe and effective option for Forestier's disease.

Ethics Committee Approval: This study was approved by Istanbul Faculty of Medicine Clinical Research Ethics Committee (Date: 03.02.2023, No: 03).

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