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Ossified Posterior Longitudinal Ligament With Massive Ossification of the Anterior Longitudinal Ligament Causing Dysphagia in a Diffuse Idiopathic Skeletal Hyperostosis Patient

Kazuhiro Murayama, MD, Shinichi Inoue, MD, Toshiya Tachibana, MD, Keishi Maruo, MD, Fumihiro Arizumi, MD, Shotaro Tsuji, MD, and Shinichi Yoshiya, MD

Abstract: Descriptive case report.

To report a case of a diffuse idiopathic skeletal hyperostosis (DISH) patient with both massive ossification of the anterior longitudinal ligament (OALL) leading to severe dysphagia as well as ossification of the posterior longitudinal ligament (OPLL) causing mild cervical myelopathy, warranting not only an anterior approach but also a posterior one.

Although DISH can cause massive OALL in the cervical spine, severe dysphagia resulting from DISH is a rare occurrence. OALLs are frequently associated with OPLL. Treatment for a DISH patient with OPLL in setting of OALL-caused dysphagia is largely unknown.

A 70-year-old man presented with severe dysphagia with mild cervical myelopathy. Neurological examination showed mild spastic paralysis and hyper reflex in his lower extremities. Plane radiographs and computed tomography of the cervical spine revealed a discontinuous massive OALL at C4-5 and continuous type OPLL at C2-6. Magnetic resonance imaging revealed pronounced spinal cord compression due to OPLL at C4-5. Esophagram demonstrated extrinsic compression secondary to OALL at C4-5.

We performed posterior decompressive laminectomy with posterior lateral mass screw fixation, as well as both resection of OALL and interbody fusion at C4-5 by the anterior approach. We performed posterior decompressive laminectomy with posterior lateral mass screw fixation, as well as both resection of OALL and interbody fusion at C4-5 by the anterior approach. Severe dysphagia markedly improved without any complications.

We considered that this patient not only required osteophyctomy and fusion by the anterior approach but also required decompression and spinal fusion by the posterior approach to prevent both deterioration of cervical myelopathy and recurrence of OALL after surgery.

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Abbreviations: CT = computed tomography, DISH = diffuse idiopathic skeletal hyperostosis, MRI = magnetic resonance imaging, OALL = ossification of the anterior longitudinal ligament, OPLL = ossification of the posterior longitudinal ligament.

INTRODUCTION

In 1950, Forestier described a peculiar pattern of axial new bone formation characterized by flowing ossification along the anterior aspect of the vertebral column, which they termed ankylosing hyperostosis of the spine (also known as Forestier disease).¹ In 1976, Resnick et al² described diffuse idiopathic skeletal hyperostosis (DISH) as a common disease that leads to ossification of ligaments and entheses of the spine as well as peripheral skeleton. The spinal form of DISH is characterized by ossification of the anterior longitudinal ligament (OALL) with involvement of the cervical spine in approximately 76% of patients.^{3,4} It occurs primarily in the elderly and predominantly in men.⁵ Some authors have reported that dysphagia resulting from DISH of the cervical spine is a rare occurrence, because it is estimated that 3% of individuals >40 years of age have DISH and, of these, only 0.1% to 6% will develop dysphagia.⁶⁻⁹

Surgical resection of OALL has been reported to be an effective treatment for cases of severe dysphagia.¹⁰⁻¹⁵ In general, OALLs are frequently associated with ossification of the posterior longitudinal ligament (OPLL).¹⁶⁻¹⁹ However, clinical features of patients complaining dysphasia complicated with these combined morbidities (OALL and OPLL) have been only sporadically reported in literatures, and its optimal treatment option has not been clarified. We present a case of a patient with DISH with both massive OALL leading to severe dysphagia and OPLL causing mild cervical myelopathy, necessitating not only an anterior approach but also a posterior one.

CASE REPORT

The review board of our institute approved this study, and an appropriate written informed consent was obtained from the patient. The study was authorized by the local ethical committee and was performed in accordance with the Ethical standards of the 1964 Declaration of Helsinki as revised in 2000.

A 70-year-old man presented with severe dysphagia with mild cervical myelopathy. His past medical history showed atrial fibrillation. The dysphagia gradually deteriorated in the past 6 months, and he had difficulty in swallowing solid food. Pharyngeal neoplasm was excluded by pharyngoscopy (Figure 1). Neurological examination revealed mild spastic paralysis and hyper reflex in his lower and upper extremities, though he could still walk without a cane. Babinski's reflex was

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From the Department of Orthopaedic Surgery, Hyogo College of Medicine, Nishinomiya, Japan.

Correspondence: Shinichi Inoue, Department of Orthopaedic Surgery, Hyogo College of Medicine, 1-1 Mukogawa-cho, Nishinomiya, Hyogo 663-8501, Japan (e-mail: inoshin@hyo-med.ac.jp).

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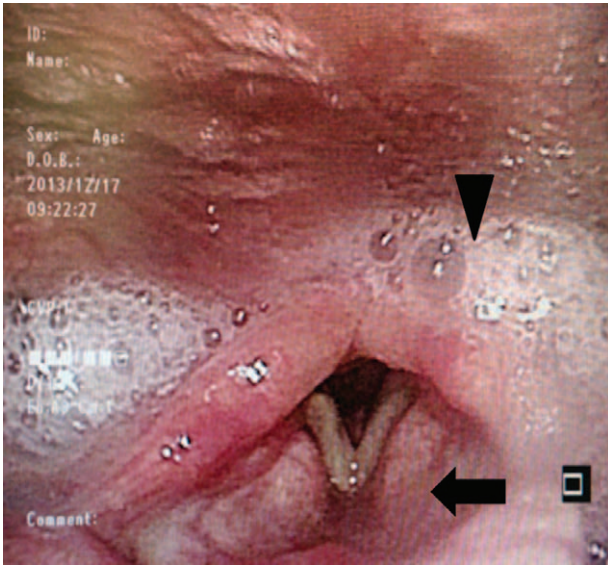


FIGURE 1. Preoperative endoscope showed stagnation of the saliva in piriform recess (arrow head) and protrusion of the posterior wall of the pharynx (arrow) without other abnormalities.



FIGURE 2. A lateral preoperative radiograph reveals massive anterior cervical osteophyte at C4-5.

not observed. Plane radiographs revealed extensive ossification ventral to the cervical spine (Figure 2). Computed tomography (CT) of the cervical spine demonstrated a discontinuous massive OALL at C4-5 and continuous type OPLL at C2-6. Axial CT image at C4-5 level presented protrusion of osteophyte compressing esophagus (Figure 3). The thickness of OALL at C4-5 was 18 mm. Magnetic resonance imaging (MRI) revealed pronounced spinal cord compression due to OPLL at C4-5



FIGURE 3. Preoperative computed tomography (CT). (A) CT of the cervical spine on the sagittal plane shows the discontinuous massive OALL at C4-5 and continuous type OPLL at C2-6. (B) CT of the cervical spine on the axial plane shows OALL compressing the esophagus and OPLL occupying the spinal canal at C4-5. OPLL = ossification of the posterior longitudinal ligament. OALL = ossification of the anterior longitudinal ligament

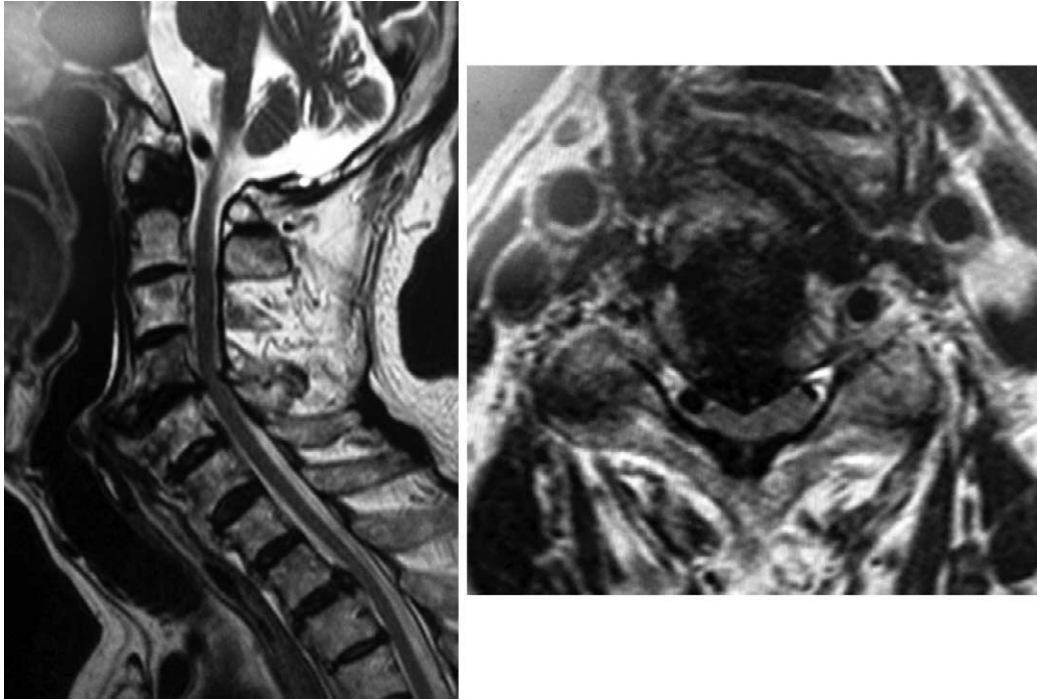


FIGURE 4. Preoperative magnetic resonance imaging (MRI). MRI of the cervical spine on the sagittal plane and the axial plane shows pronounced spinal cord compression due to ossification of the posterior longitudinal ligament at C4-5.

(Figure 4). His esophagram revealed extrinsic compression secondary to OALL at C4-5 (Figure 5). Because 6-month medications failed and his dysphagia caused gradual weight loss, we considered surgical intervention. Under general endotracheal anesthesia, we firstly performed posterior decompressive laminectomy with posterior lateral mass screw fixation. Immediately after this procedure, both OALL and the disc at C4-5 were removed by the anterior approach. Subsequent interbody fusion with a polyetheretherketone cage was

performed (Figure 6). These procedures were performed as 1 stage operation. The patient was mobilized using a semi-rigid collar after surgery. The dysphagia considerably improved without any complications. No deterioration of myelopathy or recurrence of dysphagia was detected at the 1 year follow-up, and a postoperative MRI and CT at this time showed adequate decompression with solid fusion at C4-5 (Figures 7 and 8).

DISCUSSION

We successfully treated a patient with DISH with OPLL who underwent combined surgery for decompression and stabilization of both OALL and OPLL. Because of the frequent association of OALL with OPLL in general, the same mechanism may enhance ossification of these spinal ligaments. Song et al¹⁸ reported that OPLL coexisted with OALL in 11 of 17 patients (65%); however, most patients showed asymptomatic OALL. The case in the present report who not only had myelopathy due to OPLL but also had dysphagia due to OALL is rare because till date, only 2 cases presenting with cervical myelopathy due to OPLL with symptomatic OALL have been reported.¹⁸ Epstein¹⁷ described staged surgeries in which a cervical laminectomy without fusion for OPLL was performed by the anterior approach for the excision of OALL. Chacko and Daniel¹⁹ reported that the oblique corpectomy by the anterior approach is a technique for patients with asymptomatic OALL in the setting of cervical myelopathy due to OPLL. Therefore, different surgical procedures were employed in previously reported cases, and the treatment strategy for patients who complain dysphagia caused by symptomatic OALL/DISH complicated with myelopathy due to OPLL is still unclear.

In the present case, the thickness of OALL on CT was an important contributing factor to dysphagia. Song et al¹⁸ demonstrated the mean thickness was 13.5 mm for patients with



FIGURE 5. Preoperative esophagram revealed extrinsic compression secondary to ossification of the anterior longitudinal ligament at C4-5.

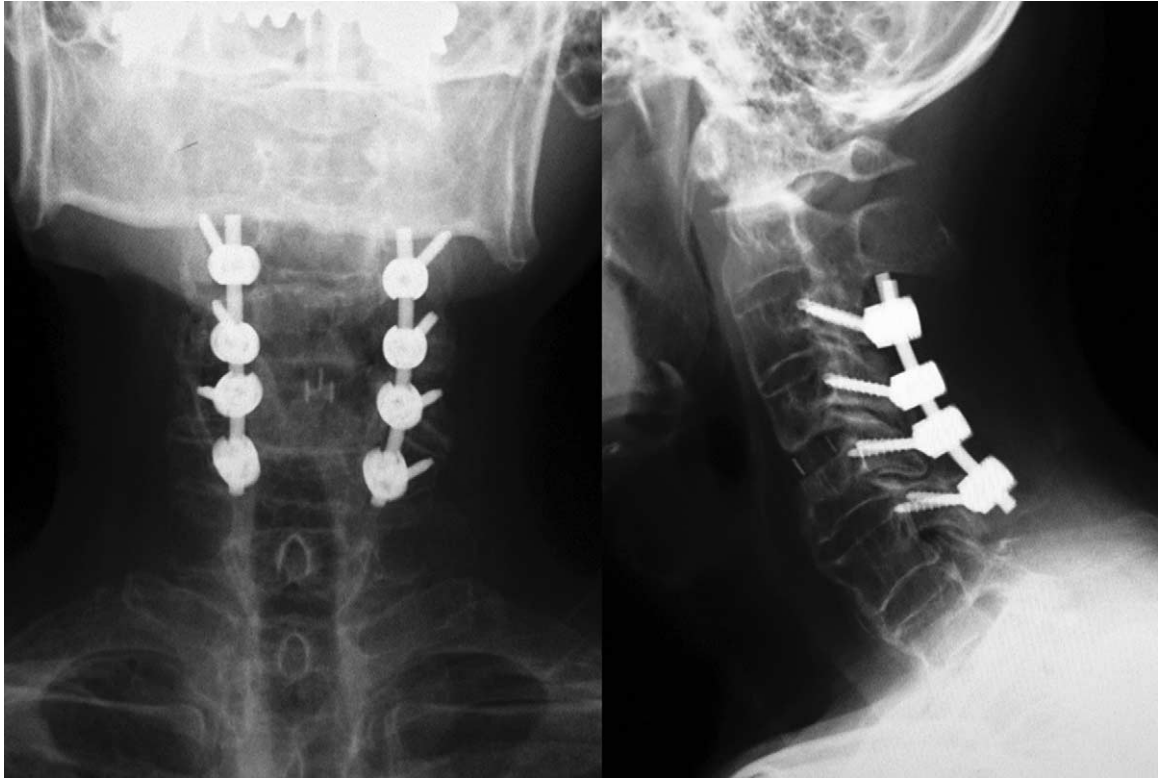


FIGURE 6. Postoperative radiograph of the cervical spine from the anteroposterior (left) and lateral (right) views.

symptomatic OALL and 6.5 mm for those with asymptomatic OALL. In the present case, the thickness of OALL was 18 mm, and it caused severe dysphagia. Carlson et al⁹ hypothesized direct compression of the aerodigestive tract and associated nerves, as well as local inflammation that leads to mucosal edema, adhesion formation, fibrosis, and cricopharyngeus

muscle spasm-induced dysphagia and respiratory complaints. In particular, adhesion or fibrosis of the esophagus due to chronic inflammation must be considered in the treatment of patients with OALL.

The initial treatment of patients with symptomatic OALL should be conservative therapy, which includes

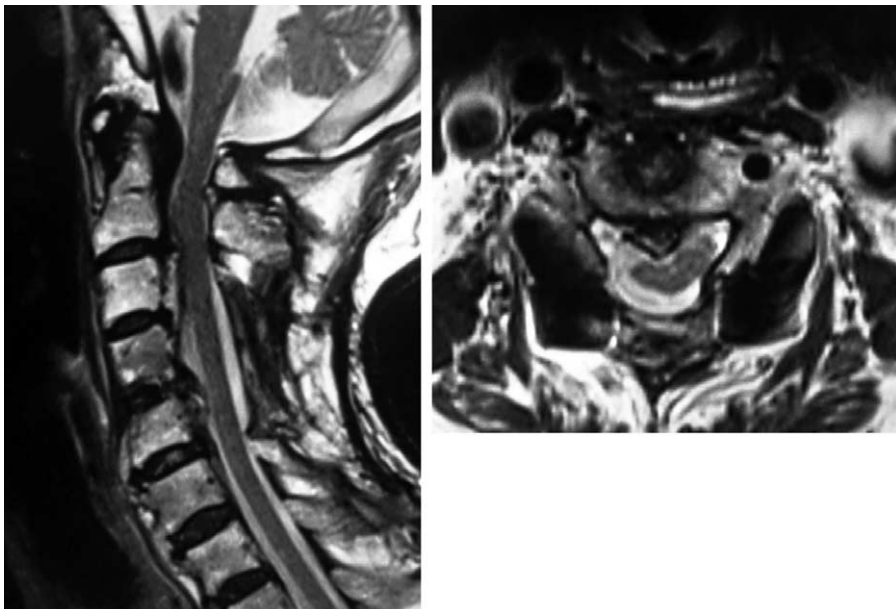


FIGURE 7. Postoperative magnetic resonance imaging of the cervical spine on the sagittal plane (left) and the axial plane at C4-5.

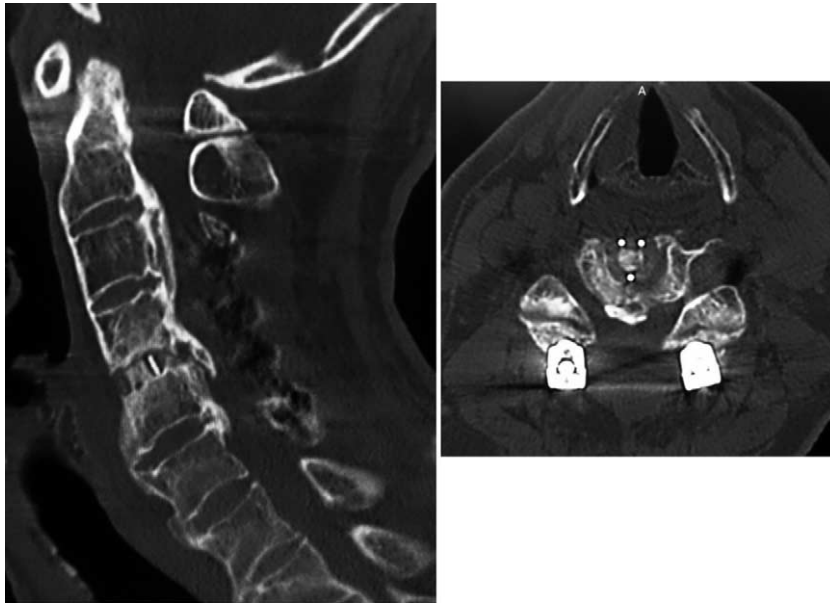


FIGURE 8. Postoperative computed tomography of the cervical spine on the sagittal plane (left) and the axial plane at C4-5.

anti-inflammatory medication, steroids, muscle relaxants, and postural changes when swallowing. Dysphagia, which is severe or resistant to conservative therapy, should be considered during surgical treatment. Resection of symptomatic OALL alone by the anterior approach is commonly performed, and many reports have shown that dysphagia resolved well after surgery.^{10–15} However, some reports have described patients with recurrence OALL associated with dysphagia. Miyamoto et al¹¹ reported that the postsurgical recurrence of OALL-caused dysphagia in patients with DISH was at an average rate of approximately 1 mm/year and that the incidence of recurrence in segments with mobility was significantly than that in segments without mobility. In the present case, because preoperative CT revealed discontinuous OALL at C4-5, postoperative recurrence caused by instability at C4-5 was thought to be a possible sequel after resection of OALL alone. Hwang et al²⁰ suggested considering a surgical strategy using a solid fusion procedure after osteophy-tectomy in order to prevent segmental instability and recurrence.

In the present case, preoperative CT and MRI revealed not only OALL at C4-5 but also a continuous type OPLL at C2-6 with apparent spinal cord compression at C4-5 that caused mild cervical myelopathy. Simple resection of OALL by the anterior approach may lead to deterioration of cervical myelopathy due to subsequent segmental instability. In general, the corpectomy by the anterior approach is recommended for segmental type OPLL, but it has limited indication for continuous OPLL. Although anterior spinal fusion by plate system is one of the surgical options, the cervical alignment change during surgery may increase spinal cord compression at C4-5. Moreover, mechanical irritation by an implant may cause chronic inflammation leading to perforation of the inherently fragile esophagus wall. Therefore, we believe that the patient reported here required not only osteophy-tectomy and fusion by the anterior approach but also decompression and spinal fusion by the posterior approach to prevent both deterioration of cervical myelopathy and recurrence of OALL after surgery. Following the surgery, dysphagia considerably improved. Although 1-year follow-up period is relatively short, we predict that clinical

condition at 1 year should be stable thereafter based on the clinical course and image findings showing solid fusion at the fusion site.

In conclusion, the simultaneous occurrence of DISH, OALL, and OPLL has been reported previously; however, a DISH patient who had cervical myelopathy due to OPLL that coexisted with a symptomatic OALL is uncommon. Therefore, planning of surgical treatment should be made based on all pathologies of DISH, OALL, and OPLL.

REFERENCES

1. Forestier J, Lagier R. Ankylosing hyperostosis of the spine. *Clin Orthop Relat Res.* 1971;74:65–83.
2. Resnick D, Niwayama G. Radiographic and pathologic features of spinal involvement in diffuse idiopathic skeletal hyperostosis (DISH). *Radiology.* 1976;119:559–568.
3. Meyer PR Jr. Diffuse idiopathic skeletal hyperostosis in the cervical spine. *Clin Orthop Relat Res.* 1999;359:49–57.
4. Mader R. Clinical manifestations of diffuse idiopathic skeletal hyperostosis of the cervical spine. *Semin Arthritis Rheum.* 2002;32:130–135.
5. Mader R. Diffuse idiopathic skeletal hyperostosis: a distinct clinical entity. *Isr Med Assoc J.* 2003;5:506–508.
6. Kmucha ST, Cravens RB Jr. DISH syndrome and its role in dysphagia. *Otolaryngol Head Neck Surg.* 1994;110:431–436.
7. Aydin E, Akdogan V, Akkuzu B, et al. Six cases of Forestier syndrome, a rare cause of dysphagia. *Acta Otolaryngol.* 2006;126:775–778.
8. Giger R, Dulguerov P, Payer M. Anterior cervical osteophytes causing dysphagia and dyspnea: an uncommon entity revisited. *Dysphagia.* 2006;21:259–263.
9. Carlson ML, Archibald DJ, Graner DE, et al. Surgical management of dysphagia and airway obstruction in patients with prominent ventral cervical osteophytes. *Dysphagia.* 2011;26:34–40.
10. De Jesus-Monge WE, Cruz-Cuevas EI. Dysphagia and lung aspiration secondary to anterior cervical osteophytes: a case report and review of the literature. *Ethn Dis.* 2008;18(Suppl 2):S2-137–S2-140.

11. Miyamoto K, Sugiyama S, Hosoe H, et al. Postsurgical recurrence of osteophytes causing dysphagia in patients with diffuse idiopathic skeletal hyperostosis. *Eur Spine J.* 2009;18:1652–1658.
12. Urrutia J, Bono CM. Long-term results of surgical treatment of dysphagia secondary to cervical diffuse idiopathic skeletal hyperostosis. *Spine J.* 2009;9:e13–e17.
13. Oppenlander ME, Orringer DA, La Marca F, et al. Dysphagia due to anterior cervical hyperosteo-phytosis. *Surg Neurol.* 2009;72:266–270 discussion 270–271.
14. Mizuno J, Nakagawa H, Song J. Symptomatic ossification of the anterior longitudinal ligament with stenosis of the cervical spine: a report of seven cases. *J Bone Joint Surg Br.* 2005;87:1375–1379.
15. Verlaan JJ, Boswijk PF, de Ru JA, et al. Diffuse idiopathic skeletal hyperostosis of the cervical spine: an underestimated cause of dysphagia and airway obstruction. *Spine J.* 2011;11:1058–1067.
16. Ehara S, Shimamura T, Nakamura R, et al. Paravertebral ligamentous ossification: DISH, OPLL and OLF. *Eur J Radiol.* 1998;27:196–205.
17. Epstein NE. Simultaneous cervical diffuse idiopathic skeletal hyperostosis and ossification of the posterior longitudinal ligament resulting in dysphagia or myelopathy in two geriatric North Americans. *Surg Neurol.* 2000;53:427–431 discussion 431.
18. Song J, Mizuno J, Nakagawa H. Clinical and radiological analysis of ossification of the anterior longitudinal ligament causing dysphagia and hoarseness. *Neurosurgery.* 2006;58:913–919 discussion 913–919.
19. Chacko AG, Daniel RT. Multilevel cervical oblique corpectomy in the treatment of ossified posterior longitudinal ligament in the presence of ossified anterior longitudinal ligament. *Spine (Phila Pa 1976).* 2007;32:E575–E580.
20. Hwang JS, Chough CK, Joo WI. Giant anterior cervical osteophyte leading to dysphagia. *Korean J Spine.* 2013;10:200–202.