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Surgical Intervention of Dysphagia Caused by Ossification of Anterior Longitudinal Ligament: A Case Report

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1. Abstract

Ossification of the Anterior Longitudinal Ligament (OALL) in the cervical spine is a common entity but rarely causes dysphagia, dyspnea, and dysphonia. We report an OALL case which causes such symptoms. A 47-year-old female patient had a complaint of progressive difficulty swallowing for two months. A cervical X-ray and Computerized Tomography (CT) scan were taken afterward, which showed OALL at the C3-7 level. She then had esophageal endoscopy to rule out other dysphagia-related disorders. The patient underwent anterior osteotomy via anterior cervical approach with significant relief of dysphagia postoperatively.

1.1. Conclusion: Surgical management in symptomatic OALL improves dysphagia and prevents its secondary complications.

2. Introduction

Hypertrophic abnormalities and osteophytes, including ossification of the Anterior Longitudinal Ligament (OALL), could be found in degenerative cervical spine or Diffuse Idiopathic Skeletal Hyperostosis (DISH), also known as Forestier's disease [1]. DISH is a noninflammatory joint disorder characterized by calcification and ossification of spinal ligaments and entheses, resulting in the development of osteophytes across the anterolateral side of the spine. DISH is diagnosed by ossification along the anterior aspect of at least four adjacent vertebrae without obvious signs of the involved intervertebral disc or apophyseal degenerative modification, and absence of facet joint ankyloses [2].

More than 75% of people 65 years of age and older have some degrees of cervical spondylosis, including anterior cervical osterophytes [3-5]. Its incidence ranges from 20% to 30% of the elderly population, but the disease rarely produces dysphagia [6]. The dysphagia caused by cervical OALL, although not commonly seen, is a primary complaint especially when osteophytes are extraordinarily large [2]. The ideal solution of symptomatic OALL remains controversial. While the majority of patients have been successfully managed by observation and medication, a surgical approach also shows benefits on some occasions [7].

This study illustrates an uncommon cervical OALL case causing dysphagia without any evidence of comorbid ossification of the posterior longitudinal ligament. The patient has made a successful recovery after the osteotomy. We will focus on the diagnostic criteria and the management strategy of this entity.

3. Case Presentation

A 47-year-old female was brought to the hospital with a complaint of solid food swallowing difficulty for 2 months. She denied hoarseness, dyspnea, sore throat, acid reflux, or unintentional weight loss. She did not have any neurological deficits. The blood work result was noncontributory.

The lateral view of the cervical spine radiograph revealed an extensive and continuous calcification area at the anterior longitudinal ligament along the anterolateral aspect of vertebral bodies from C3 to C7 (Figure 1A). The 3-dimensional (3D) reconstructed

CT scan of the cervical spine showed ossification of the anterior longitudinal ligament at C3-C7 vertebral levels (Figure 2).

The esophageal endoscopy demonstrated the protrusion of the posterior pharyngeal wall causing partial obstruction of the esophageal inlet and deviating the esophagus towards the right (Figure 3). Also, there was no evidence of intrinsic lesions within the esophageal lumen. The barium radiography has also yielded the same impression.

The patient was diagnosed with OALL complicated with an esophageal stricture. She consequently underwent a left-sided anterior cervical approach to the spine. Intraoperatively, we confirmed the esophagus had been pushed towards the right side by a large OALL at C3-C6 vertebral levels. Fortunately, there was no adhesion between the esophagus and OALL; hence, we could retract the esophagus to the right quite easily. The OALL was largely removed with an osteotome and then smoothed using a high-speed drill under operative microscope magnification (Figure 4). The histological result was chronic ligamentitis with osteochondral metaplasia (Figure 5).

The patient has experienced an improvement in swallowing since the second day after surgery. The postoperative radiograph showed the OALL has been near-complete removed (Figure 1B).

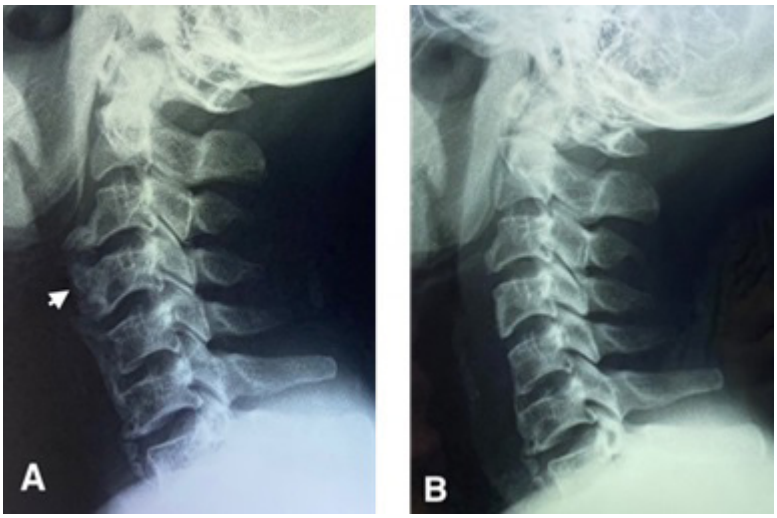

Figure 1: A: The pre-operation lateral plain radioghrap showed an extensive calcification extent from C3 toC7 (arrow), B: Post-operation image revealed the calcification were removed almost totally.

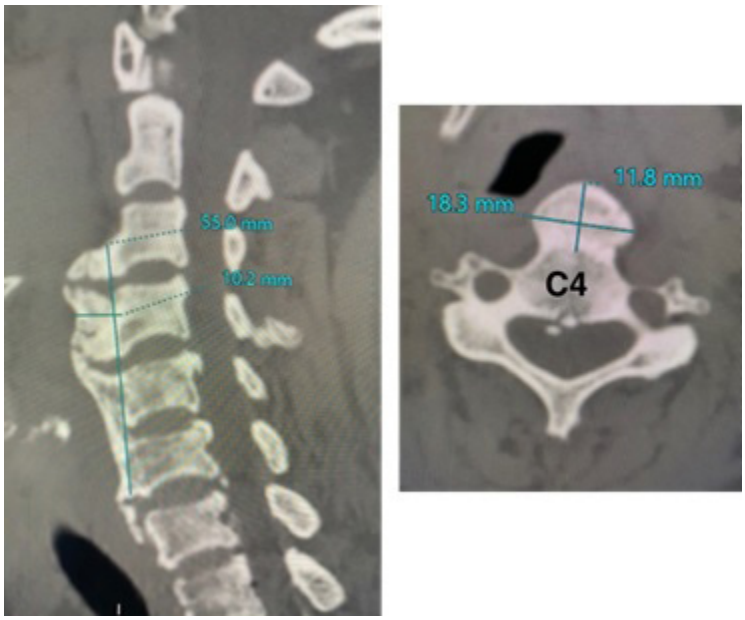

Figure 2: Cervical CT scan showed the calcification at anteriolateral aspect of C3-C7.

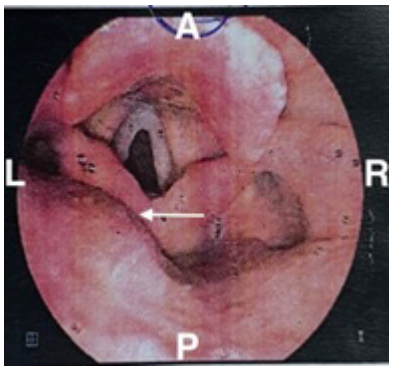

Figure 3: The esophageal endoscopy revealed the OALL protruded the posterior pharyngeal wall from the left (arrow)

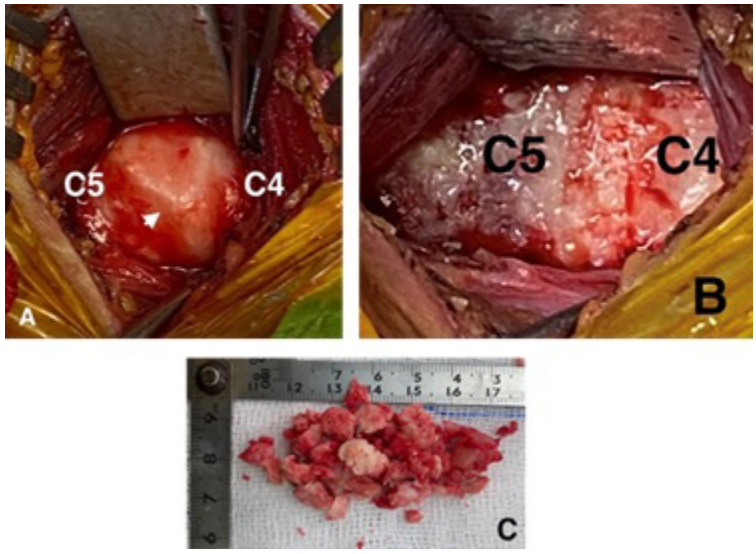

Figure 4: Intra-operation images A: The OALL (head of arrow), B: The ossification was taken out, C: The calcification

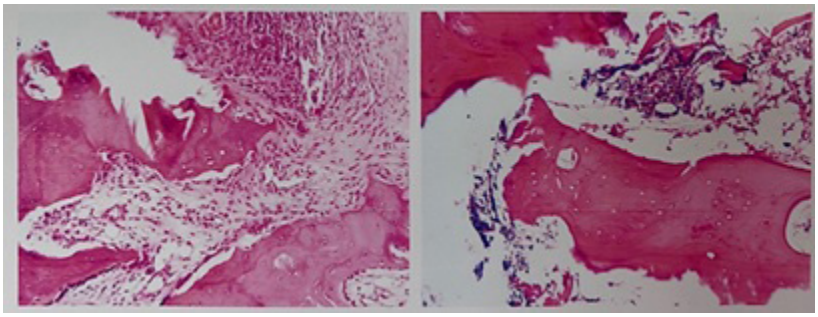

Figure 5: Histological result was chronic ligamentitis with osteochondral metaplasia

4. Discussion

Osteophyte and cervical OALL originating from degenerative changes are usually asymptomatic. The most common symptom in symptomatic OALL is progressive dysphagia with solid food. In severe cases, patients can even experience difficulty swallowing liquids. Malnutrition and severe weight loss have been frequently reported with this entity [8].

Upper respiratory tract compression may lead to dyspnea, stridor, and cough. Other sequelae include musculoskeletal and neurological symptoms, sleep apnea, aspiration pneumonia, and sudden death due to complete airway obstruction [6]. There are several etiologies potentially leading to the dysphagia sequelae in addition to a direct esopharyngeal compression [7]. In OALL, dysphagia related to an osteophyte can be explained by various mechanisms, including: (1) the esophageal lumen being compressed by a huge osteophyte; (2) Periesophageal edema from the physical irritation by osteophytes; (3) a small osteophyte but crucially located at the fixed segment of the esophagus; (4) pain and muscle spasm from the irritation (5) the combination of any of those above mechanisms [6]. In our case, there was no significant adhesion between the OALL and the esophagus, representing a non-inflammatory esophageal compression.

A plain radiograph and CT scan can efficiently detect and differentiate OALL caused by DISH from those caused by degenerative osteophytes [9]. DISH is known as one of the most frequent causes of anterior cervical hyperosteophytosis resulting in dysphagia that

requires surgical management [10, 11]. Carlson ML et al. reported that 3% of the population over 40 years of age have DISH and 0.1-6% of them will develop dysphagia [12]. In this case, we made a diagnosis of DISH because the CT imaging of our patient showed the mixture of globular OALL with a beak-like projection at the C3-5 level, and the C5-T2 vertebral bodies were fused by the anterior osteophyte, the disc height has been relatively preserved, and facet joints were not ankylosis.

Esophageal endoscopy has an important role in ruling out other dysphagia-related casualties such as cancer, neurological deficit, diverticula, pharyngeal or esophageal stenosis [13]. If Lateral Video Fluoroscopy (LVF) or esophageal endoscopy suggests evidence of pharyngeal compression, CT imaging is recommended to further investigate potential OALL and other disturbances.

The management of dysphagia in cervical OALL includes medication-based or surgical approaches. We can reasonably postpone treatment in asymptomatic patients. In patients with mild dysphagia, swallowing therapy and anti-inflammatory agents may be beneficial to control the symptoms and decrease the risk of aspiration [2, 6]. However, patients may need surgery when these conservative treatments are ineffective or the osteophytes enlarge significantly. The recent review article reported that only 35 (20.7%) of 169 OALL cases were treated by nonoperative methods [14]. Surgical interventions were used for the rest of the cases and the most common operative technique was removing ossification without spinal fusion [14]. In severe cases, OALL removal may instantly resolve dysphagia. However, taking the ossification area out alone portends the long-term risk of spinal instability and does not prevent recurrence in the future [15]. In symptomatic patients, especially those with a mechanical obstruction, timely surgical management would tremendously cease the chronic inflammatory and local fibrosis process. A delayed intervention may result in a less responsive outcome [7].

Complications were regarding the anterior approach for osteophyte resection include vocal cord palsy, Horner syndrome, and esophageal/tracheal perforation or fistula [10].

A thorough evaluation should be conducted on any patient with dysphagia. Other etiologies of this problem except cervical OALL include tumors in the esophagus, lungs, or larynx, esophageal motility disorders, esophageal webs, benign strictures, esophagitis, and infections [6]. Physicians should always indicate esophageal and laryngeal endoscopy to precisely rule out other causes of dysphagia in patients with OALL; otherwise, a misdiagnosis could lead to disastrous sequelae. Alex B. Valadka et al. reported a 68-year-old male presenting with severe dysphagia, which was initially being concluded as a complication of cervical OALL; the patient then has been through a surgical resection without any symptom improvement postoperatively. Ten days later, the laryngoscopy showed a friable mass in the right vallecula, which turned out to be squamous cell carcinoma [16]. Again, cervical OALL should only be confirmed after eliminating all other potential differential diagnoses.

5. Conclusion

We suggest evaluating cervical X-rays of patients with dysphagia when we could not find any causes coming from the gastrointestinal system. Nevertheless, cervical OALL is an exclusive diagnosis and can only be confirmed after a detailed investigation. In patients with severe symptoms or in those who have failed with medication-based treatment, early osteophyte resection yields a promising outcome.

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