

A Case of Cervical Anterior Longitudinal Ligament Ossification Causing Dysphagia and Lung Aspiration

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Abstract

Ossified anterior longitudinal ligament (OALL) in the cervical spine can cause dysphagia and dyspnea, although these symptoms are rare. We report a 67-year-old man presenting dyspnea and dysphagia caused by OALL, which was successfully treated by surgical intervention. The patient had a history of a few months of gradually increasing dysphagia and repeated pneumonia. At the time of admission, he manifested respiratory distress with stridor. Computed tomography (CT) of the chest showed consolidation in the right lung. Esophagography revealed obstruction of the esophageal passage by external compression with a pool at the laryngopharynx. Cervical CT revealed large anterior degenerative osteophytic spurs between C2 and C5, resulting in esophageal compression. The OALL was removed. The patient's postoperative course was unremarkable and his symptoms improved. Emergency physicians should consider surgical decompression for cases of marked projection of esophagus-obstructing cervical osteophytes as seen in our case. Among the several causes of dysphagia and aspiration pneumonia, severe osteophyte formation in the cervical spine should be considered.

Keywords: Ossified anterior longitudinal ligament, Dysphagia, Aspiration pneumonia

Introduction

Aspiration pneumonia, a common disease seen in the emergency department, is a major form of healthcare-associated, community-acquired pneumonia and a predominant cause of death among geriatric populations. In elderly people, saliva swallowing impairment and airway secretions mis-swallowing are chief causes of aspiration pneumonia. Particularly, dysphagia, or difficulty in swallowing, is a significant risk factor for developing aspiration pneumonia.

In elderly patients, osteophyte compression due to ossified anterior longitudinal ligament (OALL) has been identified as a cause of dysphagia when osteophytes are extremely big and are found in the cricoid cartilage region. Cervical spondylosis, including OALL, is a relatively prevalent disorder affecting mainly elderly people (it is found in approximately 20-30% of the elderly population) that often presents with excessive formation of bone [1,2]. Among these patients with OALL, aspiration pneumonia is frequently associated with hypopharyngeal compression [3]. Although dysphagia, aspiration, and OALL are common presenting problems, the association among these disorders has not been well recognized by emergency physicians and the management of symptomatic OALL remains controversial.

Here, we report a case of aspiration pneumonia and dysphagia caused by OALL, and discuss recent concepts of symptomatic OALL and aspiration pneumonia. Our experience may help emergency physicians determine the cause of pneumonia and the appropriate intervention, including surgical excision of the osteophytes. When patients present aspiration pneumonia or dysphagia, emergency physicians should be aware of the possibility of the concurrence of spinal spondylosis.

Case Presentation

A 67-year-old man was referred to our department complaining of head trauma and syncope. When he was admitted to our department, he had been experiencing an unceasing low-grade fever for five days with coarse crackles. His past medical history comprised coronary heart disease (percutaneous coronary intervention before eight

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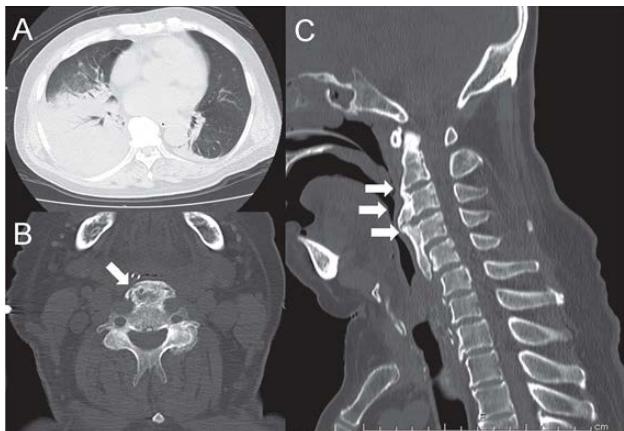


Figure A: Chest computed tomography showing aspiration pneumonia. **B:** Cervical axial CT at C4 level demonstrated OALL. **C:** Cervical sagittal CT noted osteophytic spurs in the anterior aspect of the C2-5 vertebrae (white arrow), in close approximation to the inlet of the esophagus.

years), chronic renal failure, and diabetes mellitus. He had suffered several incidents of choking sensation on swallowing. Cervical spinal movement was full and painless. Neurological examination revealed no sensory disturbance or muscle weakness. Dyspnea, cough, and fever (38.5°C) were present. Blood tests demonstrated deteriorated renal functions (serum urea nitrogen of 58.9 mg/dL, creatinine of 4.08 mg/dL) and inflammatory disease status (white blood cell count of $9360/\text{mm}^3$, C-reactive protein of 18.11 mg/dL). The patient was negative for antinuclear antibody and rheumatoid factor.

Because of his presentation, his lateral neck was X-rayed to rule out any ingested foreign body and disclosed osteophytes at the C2-5 level. The chest X-ray and chest computed tomography (CT) showed increased opacification in the lower lobe of the right lung field, which suggests infective consolidation (Figure). Gastroendoscopy was performed and esophageal lesion was ruled out. Lateral video fluoroscopy revealed a disturbance in the esophageal phase and reflux at the level of the beak-like projection. Cervical CT noted osteophytic spurs in the anterior aspect of the C2-5 vertebrae, in close approximation to the inlet of the esophagus. There was minimal cervical cord compression at the C2-5-disc level. Magnetic resonance imaging excluded spinal cord and soft tissue involvement. Based on the results of examination, cervical osteophyte was considered the most likely cause of this patient's symptoms, which included dysphagia and repeated aspiration pneumonia. Tazobactam/piperacillin (9 gram/day) was intravenously administered for aspiration pneumonia. Since the patient refused surgical intervention, he was initially conservatively managed and received dietary advice from the nutritionist, as well as anti-inflammatory therapy. However, with increasing symptoms, the patient was reassessed the next year for surgery. Surgical intervention for this case consisted of removing the offending anterior osteophytes on vertebral levels C2-5. The patient's postoperative course was unremarkable and his symptoms improved.

Discussion

Cervical osteophytes can arise as a result of spondylosis, Forestier's disease, spinal osteoarthritis, intervertebral disc

degeneration, herniated calcified nucleus pulposus, or congenital bone bar, or following trauma or infection [4]. These osteophytes are usually asymptomatic; however, they may sometimes cause dysphagia and, less often, dysphonia. The concept of diffuse idiopathic skeletal hyperostosis (DISH) is also proposed and has enduringly been considered a radiological entity showing flowing ossification neighboring the anterior and lateral borders of a minimum of four contiguous vertebral bodies, disc space maintenance, and a paucity of bony ankylosis and deterioration of the sacroiliac and apophyseal joints. In most cases, this entity presents an unremarkable clinical course, but the dysphagia seen in our patient has also been recorded in previous publications. Our case can be referred to as a case of OALL associated with DISH presenting as dysphagia.

A recent cross-sectional large-scale investigation studying Japanese elderly people showed that aspiration pneumonia risk factors were dehydration, dysphagia, dementia, and sputum suctioning [5]. Aspiration pneumonia was observed in 75% of subjects with osteophytes bigger than 10 mm and in 34% with osteophytes less than or equal to 10 mm. Other diseases affected swallowing function in 88% of those subjects with small osteophytes who aspirated [3]. Usually, osteophytes that cause dysphagia are found in the interspace of the C5 spinal segment. However, tracheal aspiration and prevention of epiglottic retroversion can sometimes be caused by osteophytes at the C3-4 level and accompanied by a choking sensation [6]. In our patient, the site of the osteophyte at the pharyngo-esophageal junction was the most appropriate position to interfere with the swallowing mechanism.

As expected, aspiration and retention are more frequently found with larger osteophyte size. However, smaller osteophytes can create clinically relevant pharyngeal residue and aspiration if concomitantly occurring with other clinical conditions. Sanders reported a patient with respiratory failure and death caused by osteophytic tracheal compression. Treatment of cervical OALL-induced dysphagia should be conservative with dietary modifications [7]. Sedation, muscle relaxants, and anti-inflammatory drugs and with an appropriate soft diet have proven effective. Most patients can be conservatively managed with a team approach, a strategy that guarantees that the dysphagic patient receives a detailed evaluation and rehabilitation for the swallowing disorder. However, surgical decompression should be considered when marked projection of esophagus-obstructing OALL is seen as in our case [8]. Of course, examination of the esophagus is necessary and other causes of dysphagia should be excluded. Our patient was able to maintain normal nutrition and avoid weight loss despite the long duration of the dysphagia. However, episodes of repeated pneumonia can indicate surgical intervention. Our patient's postoperative course was normal and the patient's symptoms improved. No aspiration pneumonia had occurred by two-year follow up.

Conclusion

A case of aspiration pneumonia and dysphagia caused by OALL is presented. The association between dysphagia and cervical spondylosis should be accepted with caution; adequate investigation and intervention is required to avoid misdiagnosis. Treatment for dysphagia caused by OALL is regarded as conservative. However, surgical decompression should be

considered for cases of marked projection of esophagus-obstructing OALL as seen in our case.

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