⟨Case Report⟩

Surgical intervention of dysphagia caused by Ossification of the Anterior Longitudinal Ligament

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ABSTRACT Background: Ossification of the anterior longitudinal ligament (OALL) of the cervical spine is a common condition but rarely results in dysphagia. Here, we present a case of dysphagia caused by OALL, successfully managed with surgical intervention.

Case: A 79-year-old female patient presented with dysphagia, having been unable to ingest any medication for one year. She had a medical history of diabetes mellitus. The patient was 153 cm tall, weighed 84 kg, and was severely obese, with a BMI of 35.9. Cervical radiography and simple CT revealed OALL at C4-7. Preoperative Videofluoroscopy(VF) demonstrated obstruction of oral medication passage due to OALL. Given the patient's difficulty in taking hypoglycemic medications and poor glycemic control, surgical intervention was pursued. The patient underwent anterior osteotomy via the anterior cervical approach, resulting in significant postoperative alleviation of dysphagia. Subsequent radiography and CT confirmed the removal of ossification, with VF indicating improvement in dysphagia. No recurrence of OALL was observed one-year post-surgery.

Conclusions: We encountered a case of ossification of the anterior longitudinal ligament leading to dysphagia. While there is no clear-cut indication for surgery, we advocate surgical intervention in cases where activities of daily living are compromised due to dysphagia.

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Key words: Ossification of the Anterior Longitudinal Ligament (OALL), Dysphagia, Cervical

BACKGROUND

Ossification of the anterior longitudinal ligament (OALL) of the cervical spine often presents asymptomatically. However, severe ossification can lead to esophageal compression and tracheal stenosis. The first documentation of dysphagia caused by OALL dates to 1950 by Forestier¹⁾. In

this report, we present a case of dysphagia resulting from ossification of the anterior longitudinal ligament of the cervical spine, which was successfully managed through surgical intervention.

CASE REPORT

The patient is a 79-year-old woman who presented

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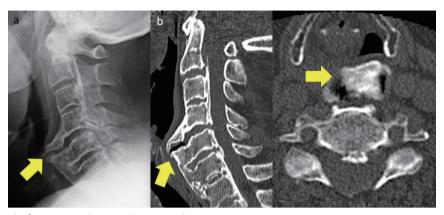


Fig. 1. a: Preoperative X-ray, b: Preoperative CT



Fig. 2. Preoperative VF

with dysphagia as her chief complaint. One year prior, she visited our hospital due to difficulty in consuming food and medicine. Her medical history included stroke and diabetes mellitus. Her height, weight, and body mass index were 153 cm, 84 kg, and 35.9, respectively, indicating severe obesity. Physical examination revealed no neurological abnormalities in the extremities. The patient was only aware of dysphagia. Preoperative radiography and CT scans showed ossification of the anterior longitudinal ligament (OALL) at C4-7, and preoperative Videofluoroscopy (VF) indicated ossification obstructing the passage of oral medication (Fig. 1, 2). Head CT images showed no abnormalities (Fig. 3). Surgery was performed because the patient had difficulty taking hypoglycemic medications and had poor glycemic control. We approached the anterior cervical

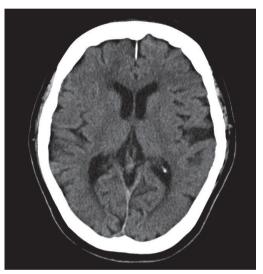


Fig. 3. Preoperative head CT

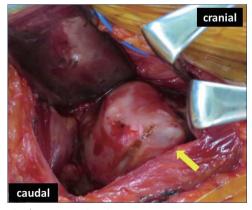


Fig. 4. Intra-operative picture

vertebrae and resected the ossification (Fig. 4). Anterior fusion was unnecessary due to the absence



Fig. 5. Postoperative X-ray

of instability. Postoperative radiographs showed successful removal of the ossification, and VF imaging indicated an improvement in dysphagia (Fig. 5). One year after surgery, there was no recurrence of dysphagia.

DISCUSSION

OALL occurs in approximately 42% of individuals over 65 years old, with an estimated frequency of dysphagia of $0.6\% - 1\%^{2}$ of patients requiring surgery³⁾. OALL is effectively treated by resecting the ossification using an anterior approach 4). However, resection of ossification has been reported to potentially exacerbate instability and lead to reoccurrence with ossification formation, resulting in dysphagia⁵⁾. The mechanism of ossification development is thought to involve stress at the site of ossification disconnection. Therefore, some reports suggest that anterior fixation should be performed because ossification resection alone may worsen instability 6). In this case, there was disconnected ossification between C4/5, but no instability was observed. One year after surgery,

there was no recurrence of dysphagia.

In this case, ossification occurred at C4-7, and physiological stenosis of the esophagus was observed at the C6 level. Therefore, the physiologic stenosis was compressed by the OALL, causing dysphagia. Additionally, dysphagia can result from sustained pressure, leading to edema and inflammation of the soft tissues, decreased esophageal elasticity, and nerve degeneration around the esophagus 7). If dysphagia is not due to compression, removing the ossification may not improve the symptoms. Other causes of dysphagia include stroke and esophageal cancer and diabetic neuropathy, which should be considered as they occur more frequently than OALL. Thus, careful preoperative examination is necessary. The patient had a history of stroke, but the onset was more than 10 years ago and there were no sequelae such as paralysis or dysphagia. And the patient had no symptoms such as hoarseness due to recurrent laryngeal nerve paralysis caused by diabetic neuropathy. We considered dysphagia due to stroke or diabetes unlikely based on medical history. Preoperative VF did not show aspiration of water. However it showed passage disturbances due to ossification, so we operated. An essential aspect of treating dysphagia due to OALL is confirming that the cause is not another disease and ensuring there is no instability when performing ossification resection. Although there is no clear indication for surgery for OALL, it is recommended in cases of difficulty to take medications due to dysphagia or in cases of low nutrition.

CONCLUSION

We encountered a case of ossification of the anterior longitudinal ligament causing dysphagia. Although there is no clear indication for surgery, we believe that surgery should be performed when the patients had difficulty taking medications or low nutrition due to dysphagia.

CONFLICTS OF INTEREST

The authors state that they have no conflicts of interest.

REFERENCES

- Foresteier J, Rotes-Querol J: Senile ankylosing hyperostosis of the spine. Ann Rheum Dis. 1950; 9(4): 321-330.
- Diogo S, Francisco B, Marta S, Jose M: Daniel L. Diffuse Idiopathic Skeletal Hyperostosis (DISH)-Phagia. Cureus. 2023; 15(10).
- 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.

- 4) Duc DTT, Quoc BN, Van TT, Thai DD, Quoc VD, Duc PV: Surgical Intervention of Dysphagia Caused by Ossification of Anterior Longitudinal Ligament. Asian J Neurosurg. 2022 8; 17(3): 485-488.
- 5) McCafferty RR, Harrison MJ, Tamas LB: Ossification of the anterior longitudinal ligament and Forestier's disease. J Neurosurg 1995; 83: 13-17.
- 6) von der Hoeh NH, Voelker A, Jarvers JS: Results after the surgical treatment of anterior cervical hyperostosis causing dysphagia. Eur Spine J 2015; 24 Suppl 4: 0-93.
- 7) Oga M, Mashima T, Iwakuma T. Dysphagia complications in ankylosing spinal hyperostosis and ossification of the posterior longitudinal ligament. Roentgenographic findings of the developmental process of cervical osteophytes causing dysphagia. Spine (Phila Pa 1976) 1993; 1; 18(3): 391-394.