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Case Report

Dysphagia after occipital cervical fusion for retro-odontoid pseudotumor with ossification of the anterior longitudinal ligament

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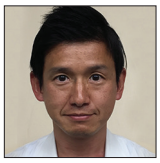

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ABSTRACT

Background: Ossification of the anterior longitudinal ligament (OALL) of the cervical spine is a relatively rare disease. If patients present with dysphagia, hoarseness, and/or dyspnea, they may require surgery.

Case Description: Over a 7-month period, a 55-year-old female with a history of cerebral palsy developed a progressive quadriparesis accompanied by diffuse sensory loss (i.e., clumsiness of the hand/legs and gait disturbance). The cervical spine X-rays showed atlanto-axial subluxation with instability, while the cervical MRI demonstrated "pseudotumor in the retro-odontoid" region. Following an occipital cervical fusion (C0-C2) surgery, her quadriparesis resolved. Nevertheless, she had persistent dysphagia that worsened over 6 months. Video fluoroscopy revealed severe mechanical stenosis of the pharynx, which was attributed to OALL extending from the C3-C6 levels. Following OALL resection through a right anterior approach utilizing diamond burrs and an ultrasonic bone curette, the dysphagia rapidly resolved.

Conclusion: We report a rare case of retro-odontoid pseudotumor successfully treated with a posterior C0-C2 cervical fusion. Additional symptomatic C3-C6 OALL, responsible for progressive dysphagia, was later managed with focal anterior OALL resection.

Keywords: Atlanto-axial subluxation, Dysphasia, Occipital cervical fusion, Symptomatic OALL, Video fluoroscopy

INTRODUCTION

Cervical ossification of the anterior longitudinal ligament (OALL), a subtype of diffuse idiopathic skeletal hyperostosis (DISH), rarely causes dysphagia requiring direct anterior cervical resection.[1,6,9] Here, we describe a 55-year-old female who following C0-C2 posterior fusion with C1 arch resection (i.e., for retro-odontoid "pseudotumor"), developed progressive dysphagia attributed to C3-C6 OALL that warranting direct anterior resection.

CASE DESCRIPTION

Over a 7-month period, a 55-year-old female with a history of athetoid cerebral palsy developed a progressive quadriparesis. Cervical X-rays showed atlanto-axial subluxation (AAS) with

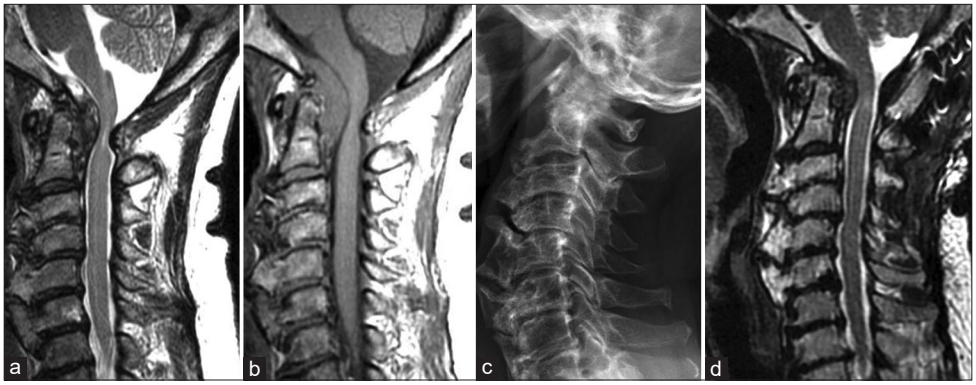

Figure 1: Preoperative MRI showed severe compression of spinal nerves due to retro-odontoid tumor (a: T2WI, b: T1WI). Cervical X-ray showed OALL like a bird's beak at the C3-6 level (c), and follow-up MRI 6 months after the first surgery clearly showed that the cerebrospinal fluid around the spinal cord had recovered and the retro-odontoid tumor was shrinking (d).

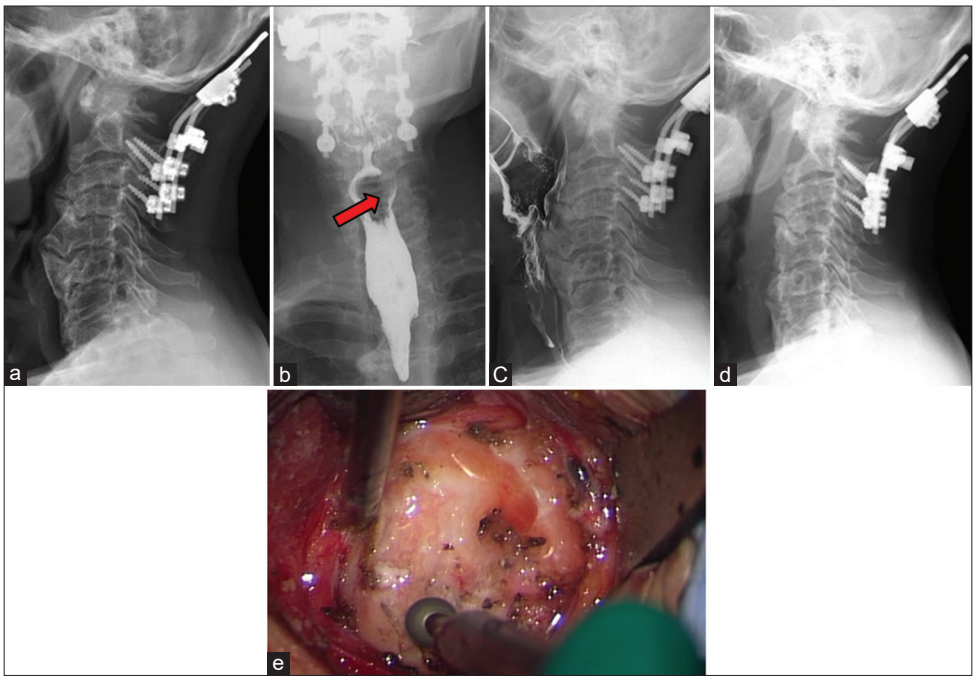

Figure 2: Aggressive OALL is prominent on cervical X-ray after the first surgery (a). Preoperative video fluoroscopy revealed a filling defect due to OALL at the C5/6 level (b,c). The red arrow in Figure 2b indicates contrast loss. Cervical X-ray after OALL resection showing adequate smoothening of the OALL (d). In the operative field, the OALL is flattened with a diamond burr (e).

instability. The cervical MR showed a retro-odontoid "pseudotumor" focally compressing the ventral cord, while the cervical CT showed OALL extending from C3-C6. For her progressive myelopathy and retro-odontoid mass with AAS, she underwent that a C0-C2 posterior fusion with C1 was arch resection. Postoperatively, although her quadriparesis resolved, she exhibited increasing dysphagia., The follow-up MRI demonstrated regression of the retro-odontoid pseudotumor [Figure 1], but video fluoroscopy confirmed C5-C6 OALL resulting in esophageal obstruction. Therefore, 6 months after the original surgery, she underwent anterior C3-C6 OALL resection using a right anterior approach, diamond burrs, and an ultrasonic bone curette [Figure 2]. Postoperatively, the patient's dysphagia improved quickly.

DISCUSSION

OALL, also called Forestier's disease, is considered a subtype of DISH.[2,8] The mean thickness of symptomatic OALL in the axial plane of CT images is reported to be 13.5 mm, which is significantly thicker than that of asymptomatic OALL (mean 6.5 mm).[9] Although surgery for symptomatic OALL has been reported to be effective in improving dysphagia,[1,6] the indication for surgery for OALL cannot be determined by

Table 1 : Previously reported studies on dysphasia associated with occipital cervical fusion or with OALL.

Authors	Number of patients with dysphagia	OALL presence	O-C fusion	Risk factors for dysphagia
Miyata *et al*.	4/29 (13.8%)	(–)	(+)	O-C2 angle of –10° or less compared to preoperative
Izeki *et al*.	3/13 (23.0%)	(–)	(+)	O-C2 angle less than preoperative
Kaneyama *et al*.	6/38 (15.8%)	(–)	(+)	PIA less than 90°
Nishimura *et al*.	11/23 (47.8%)	(+)	(–)	Large maximum thickness of OALL, small cervical ROM, small O-C2 angle, and PIA<90°

OC fusion: Occipital cervical fusion, PIA: Pharyngeal inlet angle

CT imaging alone. In the present case, the preoperative CT confirmed the presence of OALL at the C3-C6 level, but the patient had no dysphagia. Since dysphagia after craniocervical fusion can be severe, the optimal O-C2 angle to avoid complications has been variously reported.[3,5] Kaneyama *et al*. showed that dysphagia after OCF is associated with narrowing of the pharyngeal space due to craniocervical malalignment and described a pharyngeal inlet angle (PIA) of <90° as a risk factor for dysphagia.[4] Nishimura *et al*. investigated patients with symptomatic and asymptomatic OALL and found that large maximum thickness of OALL, small cervical range of motion, small O-C2 angle, and a PIA of <90° are a risk factor for dysphagia.[7] In the present case, the PIA was <90°, which was consistent with the risk factors described [Table 1]. Here, although the optimal O-C2 angle was maintained during surgery under C-arm fluoroscopy, the ultimate fixation angle was not appropriate and thus contributed to the patient's dysphagia due to OALL. To avoid postoperative dysphagia, OCF with further extension of the neck to keep the PIA below 90° should be considered. Further, preoperative VF should have been performed to evaluate swallowing function while determining the optimal angle when performing the OCF.

CONCLUSION

Patients with retro-odontoid "pseudotumor" may require a posterior C0-C2 fusion. Here, the preoperative workup should include both an MR and CT to determine if there is any accompanying subaxial cervical OALL that may become symptomatic following the C0-C2 fusion and later require direct anterior OALL resection.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent.

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Nil.

Conflicts of interest

There are no conflicts of interest.

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