



CASE REPORT

Myelopathy associated with instability consequent to resection of ossification of anterior longitudinal ligament in DISH

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Abstract

Purpose The presence of prominent OALL (ossification of anterior longitudinal ligament) in the anterior cervical spine has been implicated as a cause of dysphagia. Surgical resection of the OALL is considered effective for the management of diffuse idiopathic skeletal hyperostosis (DISH)-related dysphagia. Although many reports have been published on DISH-related dysphagia, no cases of postoperative cervical instability have been reported thus far. We present a case in which the patient developed myelopathy associated with instability consequent to resection of OALL in DISH.

Methods A 62-year-old man presented with progressive dysphagia that persisted for a year. The patient's symptoms were successfully resolved by resection of OALL. Five years after the surgery, the dysphagia resurfaced and was found to be caused by the regrowth of the OALL. A repeat surgery was performed, and the dysphagia disappeared. Eleven months after the second surgery, he visited the hospital with progressive quadripareisis and pain in the cervical region.

Results Nine-month follow-up radiologic study revealed cervical instability at the level of C5–6 resulting in myelopathy. The patient underwent decompressive laminectomy and posterior fusion surgery.

Conclusion Surgical resection of DISH-related dysphagia typically yields excellent outcomes, but our experience in this case highlights the possibility of OALL regrowth and subsequent cervical instability after resection of OALL.

Keywords Diffuse idiopathic skeletal hyperostosis · Resection of OALL · Cervical instability · Myelopathy

Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) is a rare, idiopathic spinal disease characterized by the “flowing” ossification of the anterior longitudinal ligament of the spine (OALL) [1]. The diagnosis of DISH is made when ligaments of the anterolateral spine are ossified over at least four contiguous segments, with flowing vertebral bony bridges [2]. DISH of the cervical spine has been reported to cause dysphagia [3–7]. Surgical resection of the OALL is recommended when conservative treatment is not indicated or patients have a severe dysphagia [8]. Many surgical reports about DISH-related dysphagia have been described in the literature; however, no papers have been published thus far on postoperative cervical instability. We present a case in which the patient developed cervical instability after resection of OALL for DISH-related dysphagia and discuss the cause of instability.

Case report

A 62-year-old man visited our hospital with gradually progressing dysphagia persisting for a year. Cervical computed tomography (CT) and magnetic resonance imaging (MRI) showed an elongated ossification of the anterior vertebral bodies from C2 to C6, which was more prominent between C3 and C5 (Fig. 1). The patient underwent resection of OALL at C3–4–5–6 via the Smith-Robinson approach. Immediate postoperative radiography and CT showed considerable removal of the OALL (Fig. 2). The patient had marked improvement in dysphagia.

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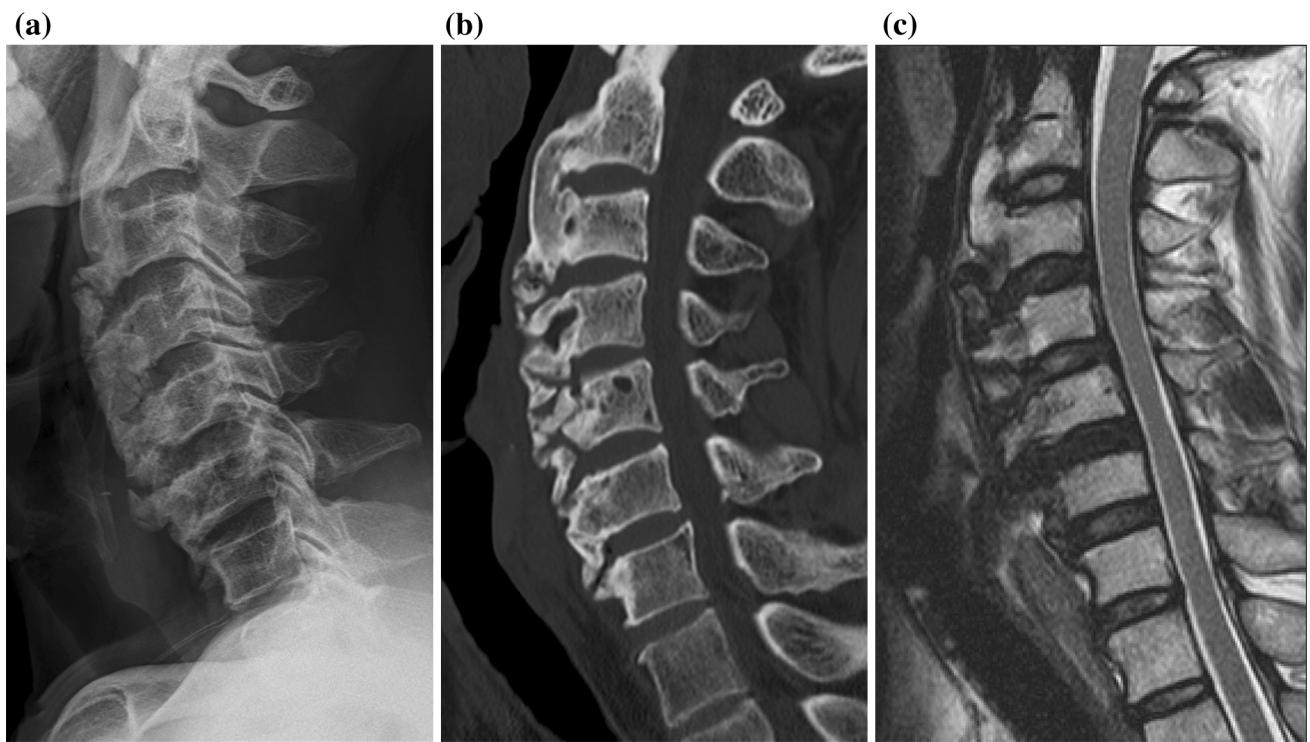
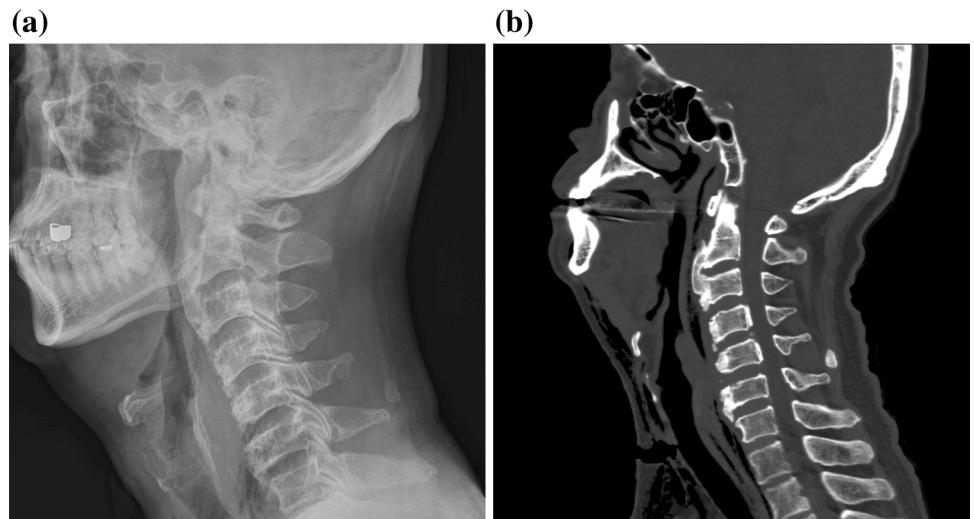


Fig. 1 Preoperative cervical simple X-ray (a), CT (b), and MRI (c) showed severe OALL from C2 to C7

Fig. 2 Postoperative lateral cervical spine X-rays (a) and CT (b) showed considerable removal of the OALL



Five years after the surgery, the patient experienced a recurrence of the dysphagia. Cervical radiography and CT showed regrowth of OALL at C3–6 (Fig. 3) and there was no instability on dynamic radiography (Fig. 4). Revision resection of OALL was performed without fusion. Immediate postoperative radiographs showed no significant residual OALL (Fig. 5) and complete resolution of the dysphagia.

Nine months after the second operation, the patient visited our hospital with posterior neck pain, bilateral

numbness of the upper limbs, and quadriplegia. Dynamic radiography revealed cervical instability at C5–6 (Fig. 6a, b), while CT showed more prominent ossification of the posterior longitudinal ligament (OPLL) at C3–5 (Fig. 6c). Further, cervical MRI showed a change in signal intensity of the spinal cord, thereby indicating cervical myelopathy at C5–6 (Fig. 6d). We performed decompressive laminectomy and posterior fusion at C3–6. After surgery, the patient reported improvement in neck pain, arm pain, and weakness.

Fig. 3 Lateral cervical spine X-rays (**a**) and CT (**b**) showed regrowth of the OALL on C3–4–5–6 5 years after the first surgery. Also, mild OPLL was shown in C3–5

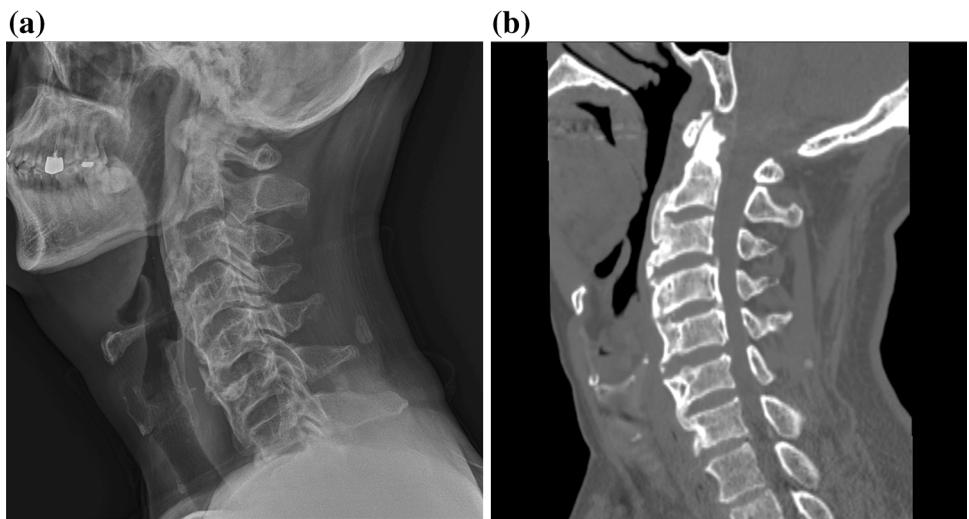


Fig. 4 Dynamic cervical spine X-ray (**a, b**) showed regrowth of the OALL on C3–4–5–6 and there was no instability at 5 years after the first surgery

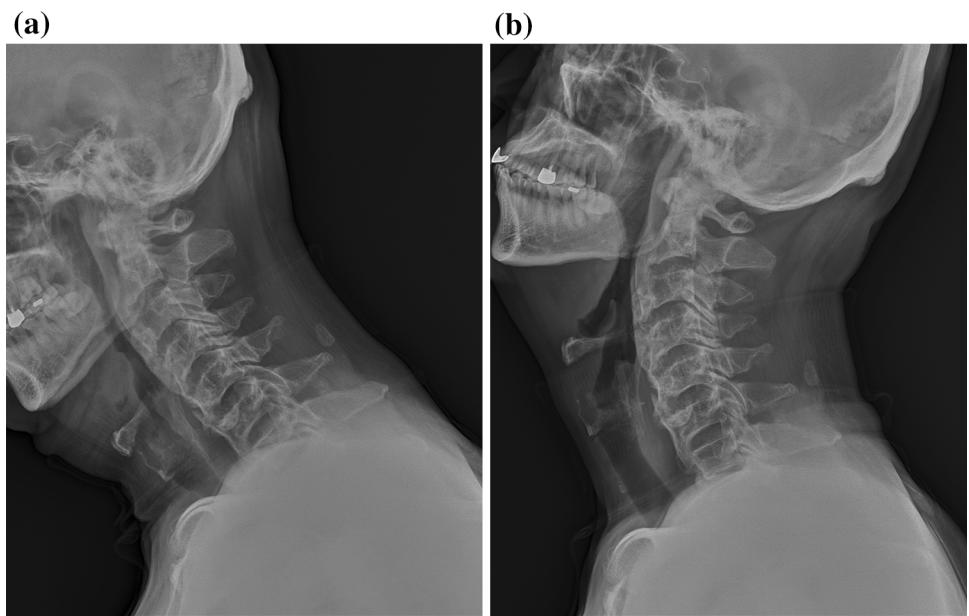


Fig. 5 Immediate second postoperative X-ray (**a**) and CT (**b**) showed significant removal of OALL on C3–6

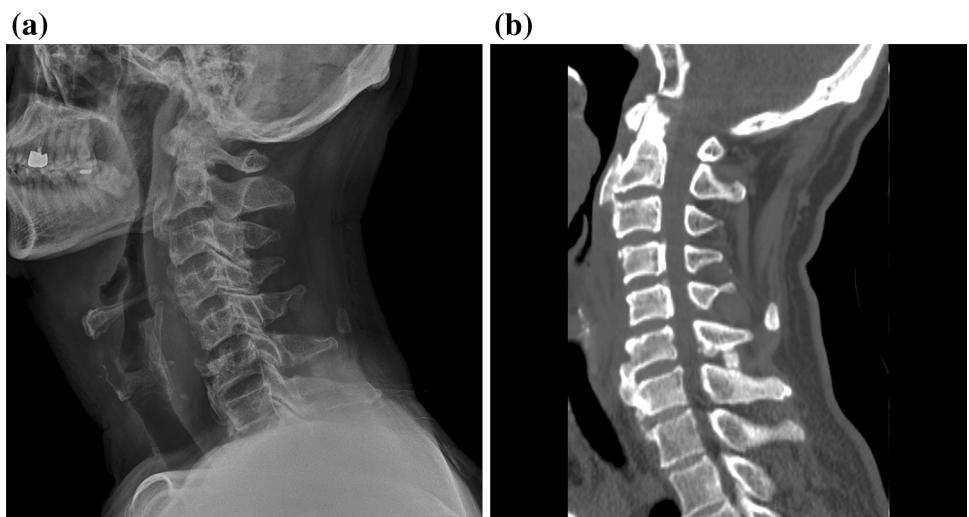
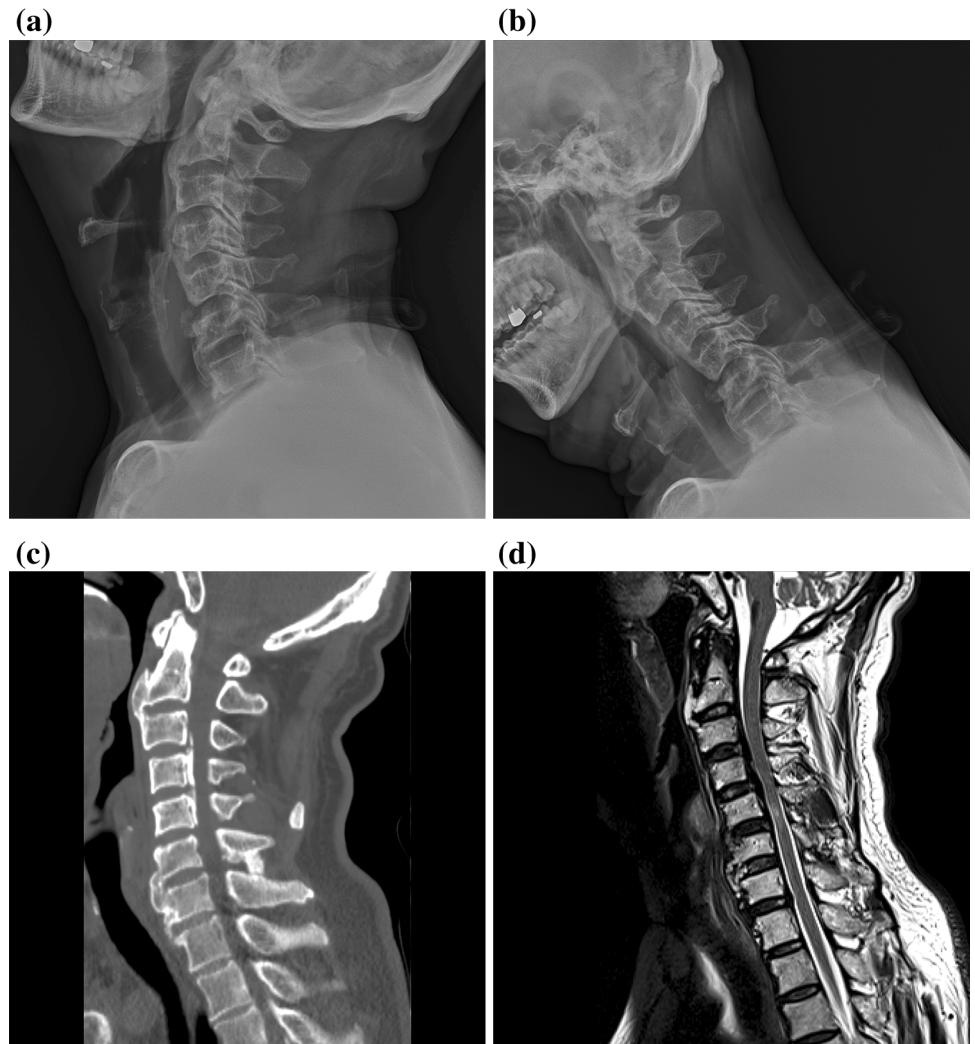


Fig. 6 **a, b** At 9 months after 2nd operation, there was cervical instability in C5–6 on dynamic X-ray. **c** Spine CT showed more prominent OPLL on C3–5 compared to CT 9 months prior. **d** MRI showed cord signal change at C5–6



Discussion

Prominent OALL can lead to compression of the esophagus or the trachea, and the patient may present with symptoms such as dysphagia, airway obstruction, stridor [6, 9]. For patients with progressive dysphagia refractory to conservative treatment or severe dysphagia, surgical resection of OALL may be recommended as a definitive treatment [3–5, 10]. In most case reports of surgical treatment, the standard, Smith-Robinson approach for resection of OALL without fusion was described [4, 10–12]. Recurrence of DISH after surgical resection is rare and if it causes dysphagia, re-operation may be considered [4, 13]. Therefore, long-term follow-up of the patients is necessary to check for the recurrence of OALL.

Many surgical reports on DISH-related dysphagia are available in the literature; however, no cases of postoperative cervical instability have been reported thus far. In our case, after the first operation, OPLL was detected (Fig. 2), but it was mild and of the segmental type. However, at 5 years after the first operation, OPLL was more prominent at C3–5

(Fig. 3). Because there was no instability on dynamic radiography (Fig. 4), we decided only resection of OALL without fusion for recurrent massive OALL. During the second surgery, we performed complete resection of OALL at C3–6, which seems to have given rise to the mobility of the C5–6 segment. On dynamic radiography of 9 months after the second surgery showed more than 11 degrees sagittal plane angulation at C5–6. At 9 months after the second surgery, the OPLL was thicker and C6–7 segment had remnant DISH with posterior bone bridge formation in the spinous process. We hypothesized that motion stress could be focused on C5–6 between fused C3–5 and C6–7 segments. This phenomenon is similar to that occurring in adjacent segment disease [14]. DISH is frequently associated with OPLL [15–18]. If other cervical spine segments are fused by OPLL and remnant OALL are present after resection of OALL, there would be mechanical loading at the non-fused segment(s) subject to excessive resection of OALL. Therefore, if fusion of other segments by OPLL is detected in preoperative imaging studies, the surgeon should take care to

avoid excessive osteophyectomy at the non-fused OPLL segment(s) to prevent segmental instability. As mentioned above, most DISH cases were treated by resection of OALL without fusion. That is beneficial because of no implant-related complication and less operative time [19]. In a review of the current literature, few cases with resection of OALL and fusion have been reported [5, 19]. Miyamoto et al. [4] demonstrated that the presence of postoperative intervertebral mobility was found to be a significant risk factor in the recurrent of OALL. And then adding cervical fusion to the resection of OALL for DISH patient could significantly lower the incidence of postoperative recurrent OALL in the fused intervertebral segments [4]. In second operation, because that was recurrent massive OALL and there was a risk of stress being concentrated in C5–6, fusion after resection of OALL could have been considered as an option. However, to determine the optimal selection of fusion level, it seems that detailed study is needed considering various variables. Considering the significant morbidity of cervical instability, we recommend that careful resection of the OALL is necessary to minimize the risk of this complication. And surgeon might be considered fusion after resection of OALL to prevent recurrence of OALL and segmental instability, especially in cases when other segment immobility by OPLL or OALL was identified.

Conclusion

Surgical resection of DISH-related dysphagia shows excellent outcomes, but cervical instability could occur after total resection of OALL. Especially, postoperative instability should be concerned in the presence of spinal diseases associated with vertebral movement, such as OPLL.

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Compliance with ethical standards

Conflict of interest None of the authors has any potential conflict of interest.

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