CASE REPORT

Symptomatic dyspnoea addressed by excision of ossified anterior longitudinal ligament

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SUMMARY

Ossification of the anterior longitudinal ligament (OALL) in cervical spine is known to cause dysphagia. However, dyspnoea and obstructive sleep apnoea (OSA) due to OALL is a rare entity. A 50-year-old man presented to our clinic 2 years after anterior cervical discectomy and fusion (ACDF) with complaints of dysphagia, dyspnoea and difficulty in sleeping supine. The clinico-neurological examination of patient was normal without any long tract signs. The diagnosis of OALL was made on plain lateral radiographs. Ultrasonic bone cutter was used to convert sessile osteophyte mass into a pedunculated mass. It was then disconnected from the anterior aspect of vertebral bodies with a chisel. The patient showed immediate relief from dysphagia and OSA. Dyspnoea improved over a week and the postoperative change in voice responded well to speech therapy. To the best of our knowledge, this is the first report of dyspnoea due to OALL after ACDF.

BACKGROUND

Ossification of the anterior longitudinal ligament (OALL) unlike the ossified posterior longitudinal ligament is a less common entity. It has not been widely described in the literature as it is rarely symptomatic.^{1–5} It has been considered as a part of Forestier's disease or diffuse idiopathic skeletal hyperostosis (DISH). Resnick and colleagues first coined the term DISH for Forestier's disease.^{6,7}

Dysphagia due to OALL has been confused previously with that of osteophytes secondary to degenerative disc disease. A clear distinction between these two conditions was not made due to poor recognition of ossification of the spinal ligaments.^{8–12} The presence of calcification or ossification bridging the anterolateral aspect of four contiguous vertebral bodies and relative preservation of disc height in the affected areas distinguish OALL from degenerative disc disease.³

Dysphagia due to DISH of cervical spine is common and various case reports have been published. However, dyspnoea leading to difficulty in performing activities of daily living and obstructive sleep apnoea (OSA) is relatively rare. We describe a case report of a patient who presented to our clinic 2 years after anterior cervical discectomy and fusion with dyspnoea, dysphagia and difficulty in sleeping supine along with difficulty in doing his routine activities.

CASE PRESENTATION

A 50-year-old man who underwent anterior cervical discectomy and fusion presented with complaints of 4-month duration of dyspnoea, dysphagia and inability to sleep supine. He had difficulty in breathing and could barely walk for more than 500 metres due to dyspnoea. He had difficulty in swallowing both liquid and solids. The clinico-neurological examination of patient was normal without any long tract signs. Conservative treatment was offered in the form of anti-inflammatory drugs, steroids, muscle relaxants and postural change while sleeping and swallowing. But there was no relief.

INVESTIGATIONS

The diagnosis of OALL was made on plain lateral radiographs of the cervical spine (figure 1). There was hyperostosis of the anterior longitudinal ligament at several levels with preservation of the disc height at the symptomatic level. The degree of anterior projection and the shape of the OALL were best visualised on CT scans (figure 2). Axial CT scans revealed prominent anterior bridging syndesmophytes with associated mass effect on the right posterior aspect of the hypopharynx (figure 3). MRI was effective in evaluating and excluding any associated compression of the cord.

Flexible laryngoscopy revealed laryngeal inlet obstruction by bulge in posterior wall of right oropharynx (figure 4). Obstruction was worse with complete neck flexion and lateral neck flexion, while it was better with neck extension. However, vocal cords were normal. No abnormal growth detected on flexible laryngoscopy. The sleep study showed moderate OSA and apnoea hyperpnoea index (AHI) was 28. The findings of sleep study suggested moderate respiratory obstruction which was supported by a low saturation of 87% at room air.

TREATMENT**Operative procedure**

A mini open anterior Smith-Robinson approach was used for removal of the OALL. We used a right transverse skin incision over the C4 vertebral body; the platysma was exposed and a thick flap of the platysma was raised. Sternocleidomastoid muscle and its medial border were identified. A plane was created medial to sternocleidomastoid and major vessels were retracted laterally.

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Figure 1 Comparison of cervical spine lateral preop (A), immediate postsurgery (B) and 1 year post-op (C): showing reduction of ossification of the anterior longitudinal ligament at C3–C4.

Medially, there were adhesions between the oesophagus and the OALL, which were separated using a microdissector. After retracting the trachea and oesophagus, the ossified mass was incised and the anterior surface was smoothed using Kerrison rongeurs. The deeper dissection was carried out using an ultrasonic bone scalpel (Misonix, Farmingdale, New York, USA) to avoid any injury to the surrounding vital structures like oesophagus, trachea, recurrent laryngeal nerve, carotid vessels and the cervical sympathetic chain.

The ultrasonic bone cutter was an ideal instrument in the present case considering the narrow window of exposure, limited corridor of vision and vital structures in the vicinity. The bone scalpel (Misonix) is an ultrasonic surgical device that cuts through osseous structures without causing significant bleeding and without significant damage to the adjacent soft tissue structures. This bone scalpel was used to gradually turn the sessile osteophyte mass into a pedunculated mass with minimal blood loss. After pedunculating the mass, it was gradually disconnected with a chisel from the anterior aspect of vertebral bodies.

Haemostasis was achieved and wound was closed in layers subsequently over a low suction drain.

OUTCOME AND FOLLOW-UP

Postoperative progress

The dysphagia recovered immediately and he was able to swallow both liquid and solid food without pain in the early postoperative period. OSA improved after surgery and sleep study at the end of second week after surgery showed AHI < 5% and 97% saturation at room air. Patient was unable to sing and generate high pitch voice following surgery for about 6 weeks. He was reviewed by otolaryngologists and an indirect laryngoscopy was performed. The larynx and hypopharynx were normal, bilateral vocal cords were mobile and there was no glottic gap on phonation but there was poor mucosal fold lengthening on high pitch phonation. The voice disorder raised the suspicion of neuropraxia of external branch of superior laryngeal nerve. After a couple of sessions of speech and voice therapy, he regained his normal pitch. [Figures 1–3](#) show comparison of preoperative and postoperative cervical spine

Figure 2 Comparison of cervical spine sagittal CT scans preop (A) and 1 year postop (B): showing reduction of the continuous massive ossification of the anterior longitudinal ligament at C3–C4.

Figure 3 Comparison of cervical spine axial CT scans at C3 level, pre-op (A) and 1 year post-op (B): showing reduction of the continuous massive ossification of the anterior longitudinal ligament at C3 with no osseous pressure on hypopharynx.

Figure 4 Comparison of laryngoscopic images showing larynx: open in supine position (A) and complete closure in forward flexion of neck (B).

radiographs and CT scans. There was a marked reduction of prevertebral shadow at the levels of C3 and C4 (table 1). The patient was followed for a period of 1 year after surgery.

DISCUSSION

Forestier's disease was first described in 1950 by Forestier and Rotes-Querol as senile ankylosing hyperostosis of the spine. In the 1970s, Resnick and colleagues defined it as DISH and directed attention to extraspinal ossification seen in this disease and the systemic nature of the ossification.^{2 6 7} They also established radiological criteria to separate Forestier's disease from osteophytes due to similar disease processes.^{3 6 7} This includes: (1) flowing calcification/ossification along anterolateral aspect of four contiguous vertebral bodies; (2) relative preservation of intervertebral disc height in affected areas and (3) the absence of apophyseal joint ankylosis and sacroiliac joint sclerosis/fusion.

OALL is generally considered as a part of DISH and is classified into three types on the radiological basis.⁴ Segmental type is defined as partial or total ossification over a vertebral body without involving the disc space. Continuous type showed ossification over many disc spaces. Mixed type is a combination of the segmental and continuous types. It generally affects elderly males above the age of 60 but the disease is usually asymptomatic. In symptomatic patients with OALL, they generally present with dysphagia resulting from the compression of oesophagus.^{1 4 13 14} The compression of oesophagus is very well seen by a barium swallow as an obstruction caused by the OALL. The compression of the oesophagus may not be limited to cervical spine but also extends to thoracic spine.³ Constant chronic compression on the oesophagus and trachea can result in erosion of the mucosal layer of the oesophagus resulting in adhesion or fibrosis and cricopharyngeal spasm due to chronic irritation.^{1 13 14} Cases have also been reported with patients having aspiration of food resulting in aspiration pneumonia due to the obstruction of the surrounding structures.¹⁵ OALL associated with neurological complaints are very rare. The other documented symptoms seen in OALL are spinal cord compression and peripheral nerve entrapment.³

Few case reports have been documented in literature reporting the patients with OALL presented with dysphagia.^{1 4 16} However, patient with dyspnoea and OSA due to OALL following an

anterior cervical spine surgery is a rare clinical scenario. Rarely, OALL can result in OSA leading to fatal consequences in patients who have very high AHI.¹⁷ The proposed mechanisms for dyspnoea in patients of OALL are retro cricoid inflammation inducing laryngeal oedema, vocal cord immobility or stridor.^{18–23}

Investigations for diagnosis of OALL should include lateral cervical spinal X-ray, CT, MRI, barium swallow, laryngoscopy and a sleep study if patient presents with difficulty in sleeping.³ The first line of management is usually conservative therapy, that is, anti-inflammatory drugs, muscle relaxants, steroids and postural changes while sleeping and swallowing. Approximately, less than 10% of patients presenting with OALL who are generally resistant to conservative management require surgical decompression.³

Song *et al* demonstrated that the mean thickness of bony barr was 13.5 mm for patients with symptomatic and 6.5 mm for those with asymptomatic OALL.²⁴ Miyamoto *et al* in his study hypothesised that there is an average increase of OALL by approximately 1 mm per year. It is significantly higher in segments which are mobile when compared with fused segments.²⁵ In the present case, the thickness of OALL was of 12.7 mm and 17.3 mm, respectively, at C3 and C4 on the trachea resulting in difficulty in breathing. The thickness after surgery recorded at C3 and C4 was 6.18 mm and 6.6 mm, respectively. There was no increase in the OALL or difficulty in breathing, sleeping or swallowing at 1-year follow-up.

For symptomatic OALL, surgery is performed mainly via the anterior (Smith-Robinson approach) or transpharyngeal approaches.^{26–28} However, patients should be aware of the potentially severe complications of surgical treatment such as haematoma, Horner's syndrome, recurrent nerve palsy, superior laryngeal nerve palsy and oesophageal injury.²⁹ Complications following surgery, which generally include dysphagia, dysphonia and dyspnoea, should be discussed with patients prior to surgery. The anterior cervical approach carries a risk of dysphonia or dysphagia, at rates of 35% and 18%, respectively, after surgery.³⁰ A thorough clinical examination, radiological assessment and laryngoscopic assessment should be done prior to surgery. It aids in minimising the intraoperative and postoperative complications and also has medicolegal value.

Standard surgical technique uses chisel, osteotomes and high-speed burr. All these are sharp instruments increasing the risk of injury to the vital soft tissues structures in the neck as these are difficult to retract. The Misonix Bone Scalpel is an ultrasonic surgical device that oscillates in a linear piston like motion, with a frequency of 22 500 strokes per minute. The thickness of the blade used was 0.5 mm. It efficiently slices bone but spares elastic soft tissues in case of incidental contacts. Unlike the high-speed burr, it does not wrap around soft tissues. The efficient irrigation

Table 1 Prevertebral shadow thickness (mm)

Level	Immediate before surgery	2 weeks postsurgery	1 year postsurgery
C3	12.7	6.18	6.18
C4	17.3	6.60	6.60

Learning points

- We present a rare case of ossification of anterior longitudinal ligament presenting with dysphagia, dyspnoea and obstructive sleep apnoea.
- Ultrasonic surgical device can help to safely remove the bony overgrowth without damaging the surrounding vital structures and minimising the blood loss.
- Excision of ossification of the anterior longitudinal ligament can be an effective treatment for symptomatic patients who are resistant to conservative treatment.

of the device and coagulum due to protein denaturation minimise the blood loss.

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