The surgical management of dysphagia secondary to diffuse idiopathic spinal hyperostosis

Lui Jonathan YC [a]; Sayal P[f] ; Prezerakos G[b]; Russo, V[c]; Choi D[d]; Casey A[e];.

[a]Medical Student, University College London, London, United Kingdom. BSc. Yew.lui.10@ucl.ac.uk

[b]Consultant, Dept. of Neurosurgery, The National Hospital for Neurology & Neurosurgery, London, United Kingdom, FRCS MD. George.prezerakos@nhs.net

[c]Consultant, Dept. of Neurosurgery, The National Hospital for Neurology & Neurosurgery, London, United Kingdom. Vittorio.russo@uclh.nhs.uk

[d]Consultant, Dept. of Neurosurgery, The National Hospital for Neurology & Neurosurgery, London, United Kingdom, FRCS PhD. David.choi@uclh.nhs.uk.

[e]Consultant, Dept. of Neurosurgery, The National Hospital for Neurology & Neurosurgery, London, United Kingdom, FRCS. Adrian.casey@uclh.nhs.uk

[f]Consultant, Dept. of Neurosurgery, The National Hospital for Neurology & Neurosurgery, London, United Kingdom, FRCS. Parag.sayal@nhs.net

Corresponding author:
Mr Jonathan Lui
The Victor Horsley Department of Neurosurgery
The National Hospital for Neurology & Neurosurgery
Queen Square, London, WC1N 3BG
Phone: 0044 7737034311
Email: yew.lui.10@ucl.ac.uk

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Running Title: Surgical management of dysphagia secondary to diffuse idiopathic skeletal hyperostosis

Abstract

Objective

This study reviews the management pathway and surgical outcomes of patients referred to and operated on at a tertiary neurosurgical centre, for dysphagia associated with anterolateral cervical hyperostosis (ACH) in diffuse idiopathic skeletal hyperostosis (DISH).

Methods

Electronic patient records for 6 patients who had undergone anterior cervical osteophytectomy for dysphagia secondary to ACH were reviewed. ACH diagnosis was made by an Ear, Nose and Throat (ENT) specialist and patients were referred to a neurosurgical-led multidisciplinary team (MDT) for review. A senior radiologist performed imaging measurements and vertebral level localization was confirmed via barium-swallow video-flouroscopy. Speech and language therapists (SLTs) determined suitability of pre-operative conservative management. Patients were followed-up post-operatively with clinical and radiological assessments.

Results

6 patients (Male to female ratio, 6:0; mean age, 59 years) were referred to a tertiary neurosurgical centre with DISH related dysphagia, an average of 25 months after ENT review (range, 14-36 months) between 2005 and 2016. The vertebral levels implicated in dysphagia ranged from C2 to T1 with a median of 4 vertebral levels involved. The most frequently affected vertebral levels were C4-6 (all 6 patients). The average height of the most prominent osteophyte was 15.9mm (range 12.0-20.0mm).

Patients underwent elective cervical osteophytectomy on average 10.8 months after neurosurgical review (range, 3-36 months). One patient had a post-operative haematoma needing evacuation and prolonged hospital stay. The average duration of follow-up was 42.3 months. All our patients maintained good symptomatic resolution without osteophyte recurrence.

Conclusions

All our patients experienced significant and sustained clinical improvement. Anterior cervical osteophytectomy consistently leads to improvement in symptomatic ACH patients without recurrence. Early referral to a neurosurgical multi-disciplinary team (MDT) is indicated in ACH related dysphagia, once conservative management has failed.

Key Words (8)

- Diffuse Idiopathic Skeletal Hyperostosis (DISH)
- Anterolateral cervical hyperostosis
- Dysphagia
- Dysphonia
- Anterior cervical osteophytectomy

Highlights

- Anterior cervical osteophytectomy successful for DISH related dysphagia
- Early referral for DISH related dysphagia to multidisciplinary surgical review
- DISH - an important differential in obstructive dysphagia and airway obstruction

1.1 Introduction

Diffuse idiopathic spinal hyperostosis (DISH) is a common albeit under-diagnosed condition. Resnick and Niwayama classified DISH as; the presence of flowing calcification and ossification along the anterolateral aspects of at least 4 contiguous vertebral bodies, with a relative preservation of disc height, in the absence of extensive degenerative disease or ankylosis.[1] The prevalence of DISH is reported to be as high as 27.1%, increasing with age and demonstrating a male preponderance of up to 6:1.[2, 3]

DISH affecting the cervical vertebrae can be referred to as anterolateral cervical hyperostosis (ACH). ACH is seen in up to 78% of DISH cases[1] and is associated with clinical symptoms including: reduced range of movement, dysphagia, dysphonia and dyspnoea.[3] Multiple pathophysiological mechanisms have been used to explain ACH symptomatology, including; direct mass effect, neuropathy and inflammation of adjacent soft-tissue structures.[3, 4, 5, 6] The incidence of dysphagia secondary to ACH, ranges from 0.1-33%, with 8-10% requiring surgical treatment.[4, 6]

Conservative management options for ACH-related symptoms include diet modification, speech and swallow therapy, and anti-inflammatory medication.[7,8] In extreme cases, enteral feeding, gastrostomy and tracheostomy may be indicated[8]. Surgical management in the form of anterior cervical osteophytectomy is considered to be highly effective[5, 7, 8] and has been recommended on: (1) failure of conservative management, (2) increased dysphagia with weight loss or (3) the appearance of laryngeal signs.[9]

Based on our institutional experience, this study reports on the surgical management and outcomes of patients diagnosed with dysphagia secondary to ACH.

1.2 Materials and methods

A retrospective review of the hospital records of patients undergoing anterior cervical osteophytectomy for DISH related dysphagia at a tertiary referral academic institution was performed.

Six patients were identified between 2005 and 2017. All patients were referred by their primary physician for ENT review. ENT specialists diagnosed dysphagia due to DISH after ruling out other causes of dysphagia, including intrinsic oesophageal dysfunction. The average time from initial ENT review to neurosurgical review was 25 months (14-36mo).

Patients were subsequently referred to neurosurgeons, who led a multidisciplinary review of these patients. A senior radiologist confirmed the diagnosis of DISH and established ACH was implicated in dysphagia, with the use of the IMPAX radiological suite (Phillips) (spine radiographs, CT and MRI) and barium videofluoroscopy.

1.3 Results and Discussion

All 6 patients were male, aged between 43 and 74 years (see appendix A1). 4 patients were of South Asian ethnicity and 2 were Caucasian. 2 were obese (BMI>30), the only significant risk factor suggested for DISH[10]. 5 patients had a history of hypertension, 3 had type 2 diabetes mellitus, and 2 had a smoking history. One patient presented without any known co-morbidities.

All patients presented with dysphagia and at least one further clinical manifestation of ACH (see table 1). The severity of dysphagia was classified as mild, if able to swallow solid food; moderate, if only able to swallow liquids; and severe, if unable to swallow fluids at all.[3] 5 patients complained of moderate dysphagia and one patient with severe dysphagia.

Subjective reporting was used to determine the presence or absence of other clinical manifestations associated with cervical DISH. Of these other clinical manifestations, 3 patients suffered from subjective reduced range of neck movement, weight loss, odynophagia or dysphonia. 2 patients suffered from dyspnoea and sleep apnoea. One of the patients had sleep apnoea independent of obesity, suggesting a direct relation to DISH.

Table 1: Pre- and post-operative severity of dysphagia, other extraspinal manifestations of cervical DISH and follow-up for osteophyte recurrence

Patient	Dysphagia		Reduced range of neck movement		Dyspnoea		Dysphonia	
	Pre-op	Post-op	Pre-op	Pre-op	Pre-op	Post-op	Pre-op	Post-op
1	Moderate	N	Y	N	Y	N	Y	N
2	Severe	N	Y	N	Y	N	N	N
3	Moderate	Occasionally mild	Y	Y*	N	N	Y	N
4	Moderate	N	N	N	N	N	Y	Y*
5	Moderate	N	N	N	N	N	N	N
6	Moderate	N	N	N	N	N	N	N

Patient	Odynophagia		Sleep apnoea		Weight loss		Osteophyte recurrence at last follow-up
	Pre-op	Post-op	Pre-op	Post-op	Pre-op	Post-op	
1	N	N	Y	N	Y	N	N
2	Y	N	N	N	N	N	N
3	N	N	N	N	Y	N	N
4	N	N	N	N	N	N	N
5	Y	N	N	N	Y	N	N
6	Y	N	Y^	Y^	N	N	N

Y= symptoms reported, N = no symptoms reported, *relative improvement reported, ^symptom unrelated to ACH

CT and MRI scans were used to determine the extent of ACH.[2] The vertebral levels implicated in dysphagia ranged from C2 to C7 with a median of 3 vertebral levels involved (See Table 2). The most frequently affected vertebral levels were C4-6 (all patients), with C4 and C5 most commonly implicated in dysphagia. The average height of the most prominent osteophyte was 15.9mm (range 12.0-20.0mm).

Patients waited on average 10.8 months from initial neurosurgical review to surgery. This however includes one patient (patient 3) who opted for additional speech and language therapy (SALT) prior to surgery, with a time from neurosurgical review to surgery of 36 months.

Table 2. Radiological characteristics as identified by swallow studies and CT/MRI.

Patient	Cervical and Thoracic vertebral levels affected by DISH (No. of vertebrae)	Level of oesophageal compression identified via video-flouroscopy	Maximum height of osteophyte(mm)
1	C3-T1 (7)	C4-6	17
2	C2-T1 (8)	C2-5	20
3	C3-6 (4)	C3-5	16.5
4	C4-6 (3)	C4, 5	16
5	C4-7 (4)	C5-7	14
6	C4-6 (3)	C5	12

All 6 patients underwent anterior cervical osteophytectomy (Smith Robinson approach) without spinal fusion. One patient (Patient 2) underwent a planned pre-operative awake tracheostomy due to significant DISH-related airway obstruction. One patient (Patient 3) suffered from a post-operative complication - a haematoma requiring evacuation and admission to an intensive care unit

Clinically significant improvement was recorded in all patients at last follow-up (see table 1). Patient 3 reported occasionally mild dysphagia with solids and improved, albeit continued, reduced range of neck movement. Patient 4 reported relative improvement of dysphonia. The average follow-up for patients was 42.3 months (12-150mo). No patients have demonstrated recurrence of osteophytes associated with DISH.

Our surgical technique uses the standard Smith Robinson approach. We use a generous horizontal mid-cervical skin crease incision. Platysma is split longitudinally. This allows generous access from C2 down to C7-T1 and as illustrated in the 2 cases below, we were able to achieve satisfactory exposure through a horizontal incision. By elevating the longus colli, the lateral aspects of the osteophytes are exposed. We used a drill to create a trough lateral to the osteophytes, using Image intensifier as guidance to drill the elatively avascular osteophytes and not enter the vascular vertebral body or the disc space. Then using an Osteotome, we are able to remove the osteophytes, with subsequent meticulous hemostasis using bone wax and hemostatic sealant to avoid post-operative hematoma in the surgical bed.

Illustrative cases detailing the methodology and results of two patients are demonstrated below.

1.3.1 Illustrative case 1 (Patient 1)

A 47 year-old caucasian male presented with recent dysphonia, progressive dysphagia of solids and liquids, weight loss, sleep apnoea, and limited range of neck movement over 36 months. Fibre-optic naso-endoscopy showed significant pharyngeal impingement due to osteophytes. MRI cervical spine (Figure 1a) and CT (Figures 1b and 1c) demonstrated multi-level anterior cervical osteophytes without disc degeneration at C3-T1. The height of the most prominent osteophyte was 17mm. Video-fluoroscopy (Figure 2) showed deflection of barium at C4-6.

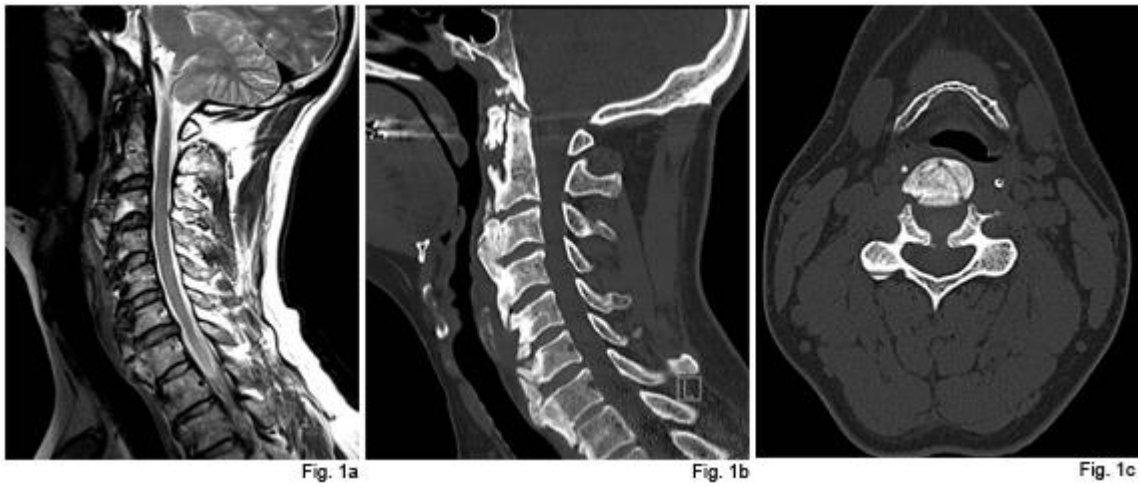

Figure 1(1.5 column fitting): Pre-operative imaging for Patient 1 demonstrating C3-T1 cervical DISH involvement in a) sagittal MRI b) sagittal CT and c) axial CT imaging.

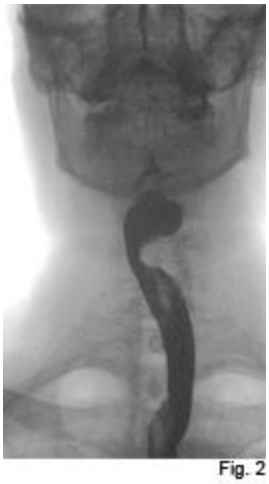

Figure 2 (Single column fitting): Video fluoroscopy barium-125 swallow on Patient 1 demonstrating bolus displacement in the oesophagus by osteophytes at C4-6.

Following anterior cervical osteophytectomy, post-operative radiographs (figure 3a) and CT (Figure 3b and 3c) showed significant resolution of osteophytes. On last post-operative follow-up at 24 months, the patient showed significant improvement in each of the presenting symptoms including the sleep apnoea and no osteophyte recurrence.

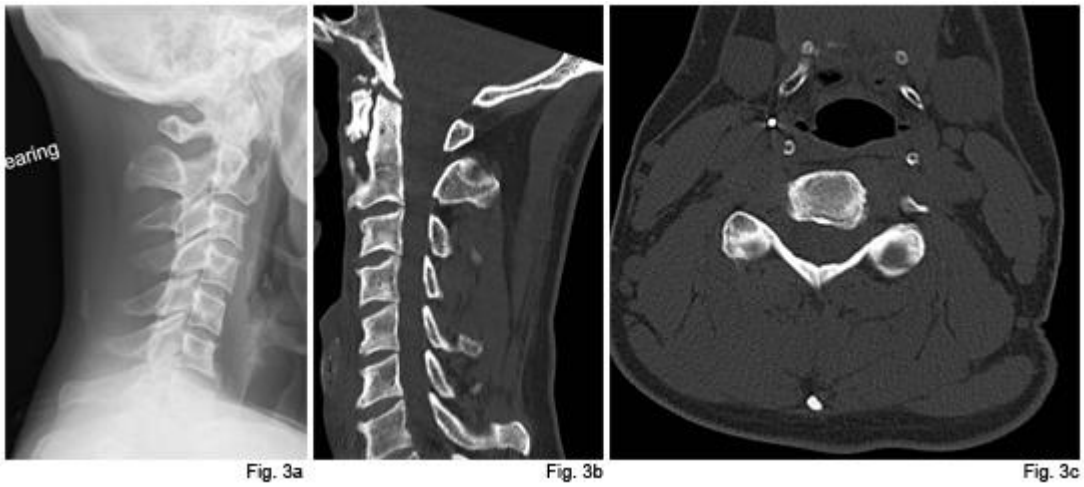

Figure 3 (1.5 column fitting): Post-operative imaging demonstrating resolution of osteophytosis in a) lateral radiograph b) sagittal CT and c) transverse CT imaging.

1.3.2 Illustrative case 2 (Patient 2)

A 60 year-old male Caucasian, suffered from dysphagia, odynophagia, dyspnoea that increased on neck flexion and limited range of neck movement. Fibre-optic naso-endoscopy showed significant narrowing of laryngeal inlet and pharyngeal impingement due to osteophytes. Radiograph cervical spine (figure 4a) showed extensive contiguous osteophytes from C2-T1. Radiograph lumbar spine (figure 4b) revealed anterior osteophytes confirming diagnosis of DISH. On CT cervical spine (figures 5a and 5b) there was extensive flowing anterior osteophytes, the largest measuring 20 mm. Level of compression on swallow studies was C2-C5.

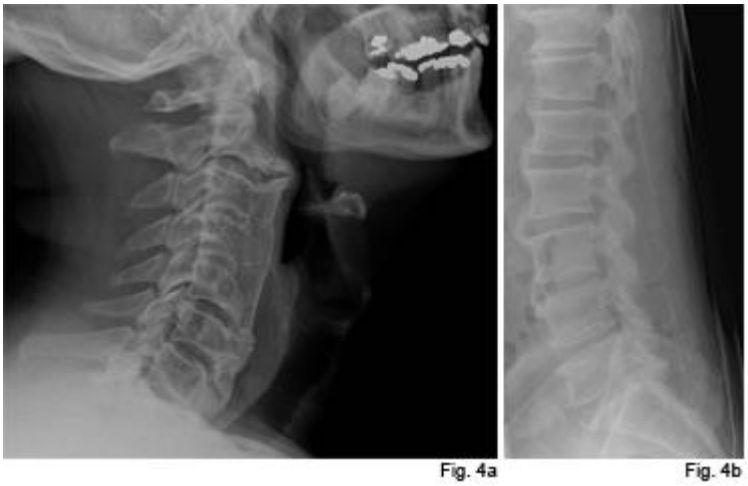

Figure 4(1.5 column fitting) : Pre-operative radiograph in a) sagittal section demonstrating C2-T1 anterior vertebral hyperostosis and b) lumbar section, demonstrating lumbar hyperostosis.

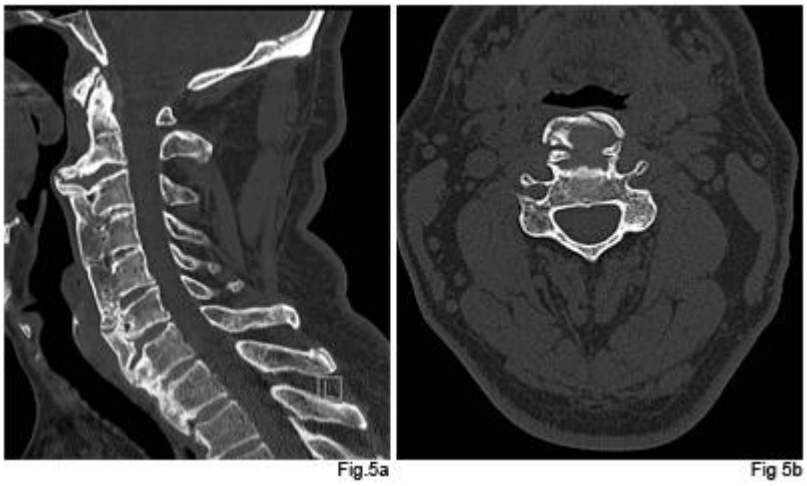

Figure 5 (1.5 column fitting): Pre-operative CT imaging with C2-6 anterior osteophytes and C2-4 tracheal compression in a) saggital and b) transverse sections

Oro/naso-tracheal intubation for surgery was not possible due to osteophyte-related airway inlet narrowing. Awake tracheostomy was performed followed by anterior cervical osteophytectomy. Post-operative CT (Figure 6a and b) confirmed significant reduction in laryngeal inlet obstruction. The tracheostomy was decanulated at 7 days and 2 weeks post-operatively the patient reported significant improvement in the pre-operative neck flexion related dyspnoea. Normal swallowing was reported at 3 months. Total follow-up was 26 months without recurrence of symptoms or osteophytes on imaging.

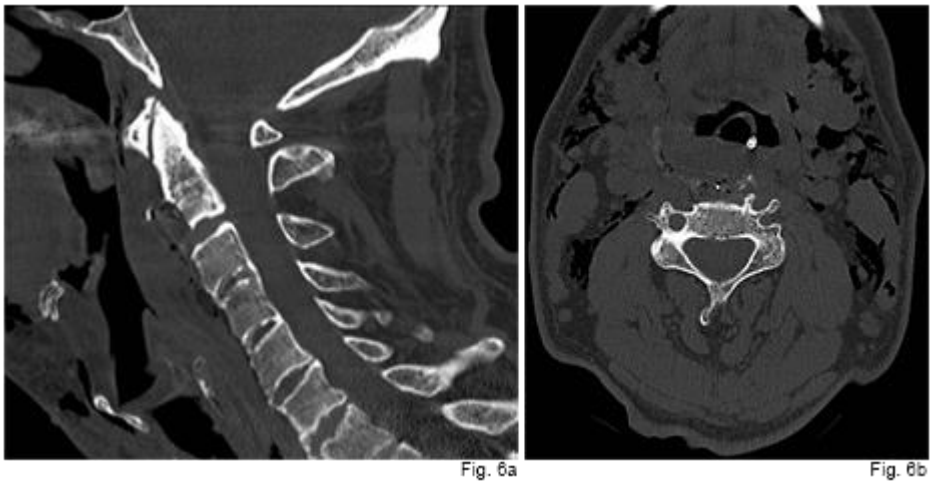

Figure 6 (1.5 column fitting): Immediate post-operative CT demonstrating resolution of anterior ostephytes and tracheal obstruction in a) saggital and b) transverse sections.

1.4 Discussion

Our study supports findings that the prevalence of cervical DISH or anterolateral cervical hyperostosis (ACH), increases with age (mean age 68.9±9.9 years) and that there is a strong male predominance (up to 6:1).[3]

The prevalence of DISH ranges from 3.8-38.7% in retrospective imaging and autopsy reviews of undiagnosed hospitalised populations.[2, 7,11] A reliable estimation of population prevalence is difficult, as the natural course of the disease is benign.[12] Whilst the aetiology of DISH remains unclear, age, sex and obesity are established risk factors.[5, 10] The prevalence of DISH and need for treatment can thus be expected to rise.[13] Ongoing classification of DISH may also increase the patient population.[14] The Resnick and Niwayama criteria, which specifies involvement of at least 4 contiguous vertebral bodies,[1] may in fact classify the end-stage of the disease.[14] More recent classification systems identify earlier phases of DISH involving 2 vertebral levels.[12] Improved classification and diagnosis of the disease may enable better characterisation of the development of dysphagia and earlier identification of surgical candidates. Involvement of the thoracic spine alone has also been suggested to be diagnostic for DISH,[4, 12] with lumbar and cervical involvement documented in up to 93% and 78% of cases respectively.[1]

ACH is associated with a number of clinical symptoms: 73% of patients may suffer from reduced range of movement, 60% with cervical pain and 33% from dysphagia.[4, 6, 15] Other ACH associated symptoms include airway obstruction, dysphonia and odynophagia.[3] There is no association between osteophyte size and symptoms.[3]

The pathophysiology of ACH associated symptoms are accounted for by a number of mechanisms: (1) mechanical obstruction by an enlarged osteophyte compressing or deviating the oesopheal and tracheal lumens, (2) osteophyte growth where the oesophagus is anatomically anchored (level of cricoid cartilage and diaphragm); (3) inflammation, fibrosis and stenosis of the soft-tissue due to irritation from the hyperostosis; (4) osteophyte associated musle spasm; (5) direct restriction of epiglottis and laryngeal movement and (6) indentation of the pharynx by an osteophyte resulting in retention in the piriform sinus.[5, 7]

These pathophysiological mechanisms concur with epidemiological findings in support of a chronic timecourse for the disease.[12] However, dysphagia and dyspnoea associated with ACH may present acutely and sub-acutely.[3] Hypotheses suggest that chronic hyperostosis is relatively well tolerated, until a ‘trigger event’ such as choking,

aspiration, or minor cervical trauma, overwhelmes the already challenged compensatory reserves.[3] There is also a propensity for low-intensity trauma causing fractures in patients with ACH.[13]

Dysphagia occurs with both solids and liquids, depending on cervical location and disease progression[3]. Decline in nutritional status with unintentional weight loss secondary to dysphagia is frequent, as reported in half of our patients. Symptoms caused by airway obstruction include: dyspnea, stridor, cough, choking, pneumonia, and even death.[9, 16]

Conservative management options for ACH associated symptoms include diet modification and speech and swallow therapy. Anti-inflammatory, anti-reflux, antibiotic, steroid, muscle relaxant and sedative medications are also indicated.[7,8] Enteral feeding and gastrostomy may be required in refractory dysphagia.[8] Tracheostomy may be indicated in acute respiratory distress and if the larynx cannot be adequately visualized before surgery.

Our study supports the use of anterior cervical osteophytectomy in patients with dysphagia and other indicated clinical symptoms of ACH. Anterior cervical osteophytectomy is considered to be highly effective and has been recommended on: (1) failure of conservative management, (2) increased dysphagia with unintentional weight loss, (3) upper airway obstruction.[5, 7-9, 17]

It has been reported that 8-10% of patients with ACH related dysphagia require surgical treatment.[15] However this may be a skewed estimate as DISH may be under-recognised as a cause of dysphagia. Patients usually demonstrate significant symptomatic improvement within 3 months post-operatively.[17, 18] Reported risks of surgery include: haematoma; resection or compression of the superior and/or inferior laryngeal nerves, the hypoglossal nerve or its descending branch and the cervical sympathetic nerve; pharyngocutaneous fistula and oesophageal perfortation and infection.[5, 9] In our study, one patient suffered from a haematoma requiring evacuation. The application of bone wax may reduce the risk of haematoma.

Given the effectiveness of surgery and the predisposition to acute presentations, we recommend prompt referral to a neurosurgical MDT on diagnosis of ACH associated dysphagia. A meta-analysis of ACH related dysphagia, found that the mean duration of dysphagia and/or airway obstruction symptoms before diagnosis of cervical DISH was 500 days (ranging from 1 to 3010 days).[3] In our study, mean time from diagnosis to surgical referral was 25 months. This may stem from under-recognition of the surgical benefits and prolonged pursuit of conservative measures. Particularly in the elderly, DISH should be considered early on in dysphagia, and once other commoner causes have been ruled, prompt referral made for surgical review.

Miyamoto et al. found an average increased rate of hyperostosis of approximately 1mm/year following resection, with 2 (out of 7) patients requiring re-operating due to progressive dysphagia at 11 years. However, the rate of recurrence and indication for vertebral fusion to avoid re-operating remains unclear.[8, 17, 19] In our case series the longest patient-follow up was 12.5 years without recurrence, and all other patients remain in remission. This may indicate that osteophyte recurrence is not necessarily the norm and therefore extensive vertebral fusion is not necessary.

1.5 Conclusions

In six patients with dysphagia related ACH in DISH, treated at a tertiary neurosurgical centre from 2005 to 2016, anterior cervical osteophytectomy led to maintained symptom resolution over a mean follow-up duration of 42.3 months. Patients with ACH and mild symptoms may be managed conservatively via a multidisciplinary approach including ENT and SALT. However, in patients with moderate to severe symptoms and patients with failed conservative treatment, early neurosurgical review is recommended, given the symptomatic improvement following surgery. Patients can expect to recover and maintain normal swallowing and vocal function within 3 months of surgery. The need for surgical fusion and long-term follow-up due to recurrent osteophytes remains debatable.

Appendices

Table A1: Patient demographics and associated medical history

Patient	Age, Sex	Asian/ Caucasian	Smoking status	HTN*	T2DM**	BMI>30
1	46, M	C	Ex	Yes (Y)	No (N)	N
2	58, M	C	Non	Y	N	Y
3	74, M	A	Non	Y	Y	N
4	68, M	A	Smoker	Y	Y	N
5	65, M	A	Non	N	N	N
6	43, M	A	Smoker	Y	Y	Y

*Hypertension **Type 2 diabetes mellitus

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