Postsurgical recurrence of osteophytes causing dysphagia in patients with diffuse idiopathic skeletal hyperostosis

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Received: 6 August 2008 / Revised: 20 July 2009 / Accepted: 4 August 2009 / Published online: 28 August 2009
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Abstract Although cervical anterior osteophytes accompanying diffuse idiopathic skeletal hyperostosis (DISH) are generally asymptomatic, large osteophytes sometimes cause swallowing disorders. Surgical resection of the osteophyte has been reported to be an effective treatment; however, little study has been given to the recurrences of osteophytes. A prospective study was performed for seven patients who underwent surgical resection of cervical anterior osteophytes for the treatment of recalcitrant dysphagia caused by osteophytes that accompanied DISH. The seven patients were six men and one woman ranging in age from 55 to 78 years (mean age = 65 years). After a mean postoperative follow-up period of 9 years (range: 6–13 years), surgical outcomes were evaluated by symptom severity and plain radiographs of the cervical spine. On all operated intervertebral segments, the effect of postoperative intervertebral mobility (range of movement >1 degree) on the incidence of recurrent osteophytic formation (width >2 mm) was analyzed by Fisher's exact test. Complete relief of the dysphagia was obtained within one month postoperatively in five patients, while it was delayed for 3 months in two patients.

All of the patients developed recurrent cervical osteophytic formation, with an average increase rate of approximately 1 mm/year following surgical resection. Of the 20 operated intervertebral segments, the incidence of recurrent osteophytes was significantly higher ($P = 0.0013$) in the 16 segments with mobility than in the four segments without mobility. Five of the seven patients remained asymptomatic, although radiological recurrence of osteophytes was seen at the final follow-up. The two remaining patients complained of moderate dysphagia 10 and 11 years after surgery, respectively; one of these two required re-operation due to progressive dysphagia 11 years postoperatively. In patients with cervical DISH and dysphagia, surgical resection of osteophytes resulted in a high likelihood of the recurrence of osteophytes. Therefore, attending surgeons should continue to follow these patients postoperatively for more than 10 years in order to assess the regrowth of osteophytes that may contribute to recurrent symptoms.

Keywords Dysphagia · Diffuse idiopathic skeletal hyperostosis · Cervical spine · Osteophyte · Recurrence

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Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) is an ossifying diathesis characterized by spinal and peripheral enthesopathy. It was first described as senile ankylosing hyperostosis of the spine by Forestier and Rotes-Querol [2] in 1950. In the 1970s, Resnick et al. [9, 10] coined the term DISH for this systemic entity. They further advocated following three diagnostic criteria: (1) the presence of flowing ossification of the anterior longitudinal ligament (OALL) of at least four contiguous vertebral bodies, (2) the preservation of intervertebral disc height, and (3) the absence of

apophyseal joint bony ankylosis and sacroiliac joint erosion, sclerosis, or intra-articular osseous fusion [9, 10].

Cervical anterior osteophytes accompanying DISH are most frequently asymptomatic. Large osteophytes, however, do cause swallowing disorders through a variety of mechanisms, including: (1) direct mechanical compression of the pharynx and esophagus [9], (2) disturbances of normal epiglottis tilt over the laryngeal inlet by the osteophytes at C3–C4 level [5, 14], (3) inflammatory reactions in the tissues around the esophagus [1, 16], and (4) cricopharyngeal spasm [2].

In 1926, Moshe [8] was the first to report dysphagia secondary to cervical osteophytes. Clinical studies have shown that 17–28% of patients with DISH manifested symptoms of dysphagia due to cervical osteophytes [6, 9]. Conservative treatment has been indicated for the initial management of these cases [2, 16]. Surgical resection of the osteophyte has been reported to be an effective treatment for severe cases and/or cases with airway obstruction [1, 3, 4, 7, 12–14].

Many surgical reports about DISH-related dysphagia have been described in the literature [1, 3, 4, 7, 12, 13, 16]: however, few of these include postsurgical results for more than 2 years. Except for a few reports in the Japanese literature [3, 13], little study has been given to the regrowth of osteophytes after surgical resection. Therefore, from 1991, we started to observe long-term postoperative courses of DISH patients with recalcitrant dysphagia who underwent surgical resection of osteophytes. The purpose of this study was to examine these surgical outcomes, with particular attention given to the recurrence of those osteophytes contributing to dysphagia.

Materials and methods

Patient population (Table 1)

This study included a total of seven consecutive patients who underwent surgical resection of cervical anterior

osteophytes between 1991 and 1997 for the treatment of recalcitrant dysphagia caused by large osteophytes accompanying DISH. The seven patients were six men and one woman ranging in age from 55 to 78 years (mean age = 65 years). All patients gave their informed consent to participate in this prospective study. All patients exhibited recalcitrant dysphagia persisting for more than 6 months despite appropriate conservative treatment including diet modification and a regimen of anti-inflammatory drugs and muscle relaxants. Four of the seven patients presented with odynophagia. Only Case 7 had aspiration accompanied by spontaneous cough during swallowing of solids. Serologic tests for HLA-B27 antigen were negative and serum levels of C-reactive protein and rheumatoid factor were normal in all patients.

Physical examination

In all patients, the physical examinations were remarkable only for a decreased range of movement in the cervical spine.

Plain radiographs, CT scanning, and MR imaging

In all seven patients, lateral plain radiographs of the thoracic spine showed flowing ossification of the anterior longitudinal ligament along the anterior vertebral bodies, consistent with the diagnoses of DISH [9, 10]. On plain radiographs, hyperostosis at the ligamentous attachments [9, 10] was observed on the pelvis (seven patients), patella (three patients), calcaneus (three patients), and elbow (two patients). All patients had large osteophytes along the anterior aspects of the cervical vertebrae. Two patients (Cases 5 and 6) had a protruding osteophyte complex at only one intervertebral segment. In the other five patients (Cases 1–4 and 7), the OALL extended along multiple segments, producing beak-like osteophytes at one or more segments (Table 1). All patients were assessed by

Table 1 Preoperative clinical and radiological data in 7 surgically treated patients

Case	Age (years)	Sex	Symptoms	Duration of dysphagia (months)	Number of affected segments	Width of the largest osteophyte (mm)	OPLL
1	55	M	Moderate dysphagia	22	2 (C4–5, 5–6)	20	No
2	57	F	Moderate dysphagia, odynophagia	45	4 (C3–4, 4–5, 5–6, 6–7)	15	No
3	63	M	Moderate dysphagia	11	3 (C4–5, 5–6, 6–7)	16	Yes, C2–5
4	64	M	Severe dysphagia, odynophagia	25	4 (C4–5, 5–6, C7–Th1)	17	No
5	66	M	Moderate dysphagia, odynophagia	12	1 (C4–5)	14	No
6	70	M	Moderate dysphagia	7	1 (C5–6)	14	No
7	78	M	Severe dysphagia, odynophagia, aspiration	78	5 (C2–3, 3–4, 4–5, 5–6, 6–7)	18	No

OPLL ossification of the posterior longitudinal ligament

computerized tomography (CT) scanning and magnetic resonance (MR) imaging of the cervical region. Ossification of the posterior longitudinal ligament (OPLL) was identified by CT scanning of the cervical spine only in Case 3, who was asymptomatic. In this patient, MR imaging demonstrated a slight compression of the dural sac by the OPLL mass.

Indirect laryngoscopy and swallowing studies

Indirect laryngoscopy and video fluoroscopic-modified barium swallow studies were performed by an otolaryngologist. In all patients, indirect laryngoscopy revealed a submucosal protrusion in the pharynx and video fluoroscopic swallow studies demonstrated extrinsic compression of the pharynx or esophagus caused by the osteophytes. A moderate reduction of the epiglottic tilt, due to C3–C4 osteophyte, was found in Cases 2 and 7. Pharyngeal residue in the valleculae or pyriform sinuses after swallowing were observed in all patients. Aspiration of the contrast medium was found without coughing (silent aspiration) in three patients (Cases 1, 3, and 4), with coughing in one patient (Case 7).

Diagnosis for cause of dysphagia

None of the patients had coexisting diseases that might affect swallowing function, such as cancer of the lung, tongue, larynx, pharynx or esophagus, or stroke, achalasia, or Parkinson's disease. In all patients, the protruding cervical osteophytes accompanying DISH were diagnosed as the cause of dysphagia.

Severity of dysphagia

The degree of dysphagia was graded as none, mild, moderate, or severe. Mild dysphagia was defined as an abnormal sensation (e.g., foreign-body sensation or mild painful sensation) in the pharynx during swallowing of solids or liquids. Those patients with moderate dysphagia had difficulty in swallowing solid boluses, although the patients could swallow small amounts of liquid without difficulty. Patients graded with severe dysphagia were unable to swallow a small solid bolus, or experienced aspiration and coughing during swallowing of solids or liquids. Dysphagia grades were moderate in five patients and severe in two patients (Table 1).

Surgical procedures

Following induction of general anesthesia and nasotracheal fiber optic intubation, all seven patients underwent resection of cervical anterior osteophytes without fusion. All the

procedures were performed using a left-side anterior approach and Casper self-retraction systems. There was a slight to moderate adhesion between the anterior aspect of the cervical osteophyte and the surrounding connective tissue. Therefore, dissection was performed very carefully. The osteophytes were removed down to the level of the anterior cortices of the vertebral bodies by using a rongeur and high-speed air drill with diamond burs. An intra-operative lateral radiograph was obtained to confirm the adequacy of resection. None of these patients received external stabilization. The numbers of operated intervertebral segments were one (Cases 5 and 6), two (Case 1), three (Case 3), four (Cases 2 and 4), and five (Case 7). There were 20 operated segments in all (Table 1).

Patient assessment

All seven patients underwent a minimum of 6 years' follow-up (mean = 9 years, range: 6–13 years). Clinical and radiological assessments were performed preoperatively and were continued monthly for 6 months postoperatively, then at 6-month intervals. The clinical assessment included a self-assessment questionnaire on the severity of dysphagia. For radiological assessment, plain radiographs of the cervical spine, including anteroposterior, lateral, and flexion-extension views, were obtained. On lateral X-ray films, the width from the anterior edge of the vertebral body to the tip of the osteophytes (sagittal diameter) was measured at each intervertebral segment from C2–C3 to C7–Th1 (six segments; Fig. 1). The measurement was corrected for the

Fig. 1 The measurement for the size of the osteophytes. A straight line was drawn at the anterior margin of the vertebral body. Then, another line was drawn perpendicularly to the straight line at the vertebral anterior margin. Then, the distance between the tip of the osteophyte and the vertebral anterior margin was measured

degree of magnification of the radiograph. The mean pre-operative width of each patient's largest osteophyte was 16 mm (range = 14–20 mm). Symptomatic recurrence was defined as dysphagia for solids or liquids occurring postoperatively. Radiological recurrence was defined as the width of the recurrent osteophyte increasing by more than 2 mm in postoperative lateral radiographs. The range of movement (ROM) at each intervertebral segment from C2–C3 to C7–Th1 was measured by flexion-extension views. If the ROM was more than one degree, the segment was considered mobile. A bridging osteophyte across the segment or ROM ≤ 1 degree at the segment were the criteria used to confirm immobilization of the segment.

The relationship between postoperative intervertebral mobility and changes in recurrent osteophytes

In the seven patients, the effect of postoperative intervertebral mobility (ROM > 1 degree at 1-year follow-up) of the 20 operated segments on the incidence of recurrent osteophytic formation (width > 2 mm at the final follow-up) was analyzed by Fisher's exact test.

Results

Clinical outcome (Table 2)

There were no incidences of esophageal injury or recurrent laryngeal nerve paralysis. All patients were given nothing by mouth for several days postoperatively to avoid aspiration. Supportive intravenous fluids were given during this period. Five of the seven patients returned to a regular diet within one week following surgery and obtained complete relief of dysphagia within one month postoperatively. Two patients (Cases 4 and 7) had minimal initial improvement followed by a substantial, but slow resolution of dysphagia and required diet modification for the following 3 months.

At the one-year follow-up evaluation, the five patients with a speedy recovery, and Case 4, had recovered normal swallowing. Case 7 obtained a significant decrease of aspiration accompanied by a cough, but remained moderately dysphagic. At the final follow-up, this patient, who was the oldest and had the longest duration of dysphagia, still presented with mild dysphagia.

Five of the seven patients did not have symptomatic recurrence during the follow-up period. However, two patients (Cases 1 and 2) complained of recurrent dysphagia at their 11- and 10-year follow-up evaluation, respectively. Case 2 required re-operation includes resection of recurrent osteophytes, a two-level discectomy and fusion due to progressive dysphagia 11 years following the initial surgery, and was symptom-free 2 years later. Intraoperative finding at the re-operation revealed firm adhesion between the connective tissue and the osteophyte. Case 1 complained of moderate dysphagia when swallowing a solid bolus at the 12-year follow-up.

Radiological outcome (Table 2)

All seven patients developed radiological recurrence of cervical osteophytes (width >2 mm) within 4 years after the osteophyte resection. The recurrent osteophytes continued to grow in thickness at the operated mobile segments. The mean postoperative width of each patient's largest recurrent osteophyte was 9 mm (range: 4–15 mm) at the final follow-up evaluation.

The relationship between postoperative intervertebral mobility and changes in recurrent osteophytes (Table 3)

Of the 20 intervertebral segments that underwent resection of osteophytes, the incidence of recurrent osteophyte was significantly higher ($P = 0.0013$ by Fisher's exact test) at the segments with mobility (ROM > 1 degree) than in the segments without mobility (ROM ≤ 1 degree).

Table 2 Postoperative clinical and radiological data

Case	Follow-up (years)	Postoperative relief of symptoms (time period)	Radiological recurrence	Width of the largest recurrent osteophyte (mm)	Symptomatic recurrence	Additional surgery
1	12	Complete (immediate)	Yes	15	Yes	No
2	13	Complete (immediate)	Yes	12	Yes	Yes ^a
3	6	Complete (immediate)	Yes	5	No	No
4	10	Complete (postoperative 3 months)	Yes	10	No	No
5	6	Complete (immediate)	Yes	4	No	No
6	9	Complete (immediate)	Yes	5	No	No
7	7	Incomplete (postoperative 3 months)	Yes	9	No	No

^a Resection of recurrent osteophytes, a two-level discectomy and fusion using plate

Table 3 The incidence of recurrent osteophytes with and without postoperative intervertebral mobility

Postoperative intervertebral mobility	Recurrent osteophyte (number of segments)		Total
	Present	Absent	
Present (ROM > 1 degree)	14*	2	16
Absent (ROM ≤ 1 degree)	0*	4	4

* Significant difference ($P = 0.0013$) by Fisher's exact test

Clinical presentations

Case 1

A 55-year-old man presented with a 2-year history of dysphagia. Lateral radiography of the cervical spine demonstrated large anterior osteophytes bridged from the C4 to the C6 level (Fig. 2a). The C4–C6 osteophyte complex was removed by surgery (Fig. 2b). The patient had complete relief of dysphagia within one month. Dysphagia recurred 11 years later. The recurrent osteophytes, representing bony multisegmental bridges, extended from the C3 to the C7 level, where intervertebral mobility remained (Fig. 2c).

Case 2

A 57-year-old woman had a 4-year history of dysphagia. Lateral radiography of the cervical spine demonstrated anterior osteophytes bridged from the C3 to the C7 level (Fig. 3a). The patient underwent resection of the osteophytes (Fig. 3b) and had complete relief of dysphagia within one month. Ten years later, the patient again complained of dysphagia; recurrent osteophytes had formed at the C2–3, 3–4, and 6–7 mobile segments (Fig. 3c). Eleven years after the first surgery, resection of the recurrent osteophytes and a two-level discectomy with fusion using

plates were performed due to progressive dysphagia. The patient had no recurrence of osteophytes and dysphagia for 2 years after the second surgery (Fig. 3d).

Discussion

The present study reports the first long-term follow-up of a cohort of seven DISH patients with dysphagia who underwent surgical treatment. Actually, previously reported pathology of dysphasia due to anteriorly protruding deformity of the cervical spine can be classified into OALL [1, 5, 16] and osteophytosis related to cervical osteoarthritis [5, 6, 12, 13, 15, 16]. In the present study, large osteophyte at a still mobile intervertebral segment in a patient with OALL and DISH, suggesting that the osteophyte can be potentially an immature form of the OALL, were subjected for analyses. Due to the rare incidence of this pathological condition, the recruitment of seven patients took nearly 7 years and the final follow-up period varied from 6 to 13 years. The results consistently showed that these DISH patients developed a recurrence of cervical anterior osteophytes following surgical resection. Importantly, the presence of postoperative intervertebral mobility was found to be a significant risk factor in the recurrent formation of osteophytes [3]. Until now, about 100 cases with DISH-related dysphagia have been described in the literature [1, 3, 4, 7, 12, 13, 16]. Most of these surgical reports do not include outcomes from long-term postoperative follow-up and little study has been given to the recurrence of osteophytes. To the best of our knowledge, only three Japanese cases have been recorded reporting recurrence of cervical anterior osteophytes. Hirano et al. [13] reported that two patients developed asymptomatic recurrent osteophytic formation at the operated site 4.5 years after surgical resection. Suzuki et al. [3] also

Fig. 2 Case 1: **a** Preoperative lateral cervical spine X-ray film. **b** Immediately after surgery. **c** Eleven years after surgery

Fig. 3 Case 2: **a** Preoperative lateral cervical spine X-ray film. **b** Immediately after surgery. **c** Ten years after the first surgery. **d** Two years after the second surgery

reported that two patients developed a recurrence of osteophytes 3 and 4 years after surgical resection, respectively, and one of the two had recurrent dysphagia 11 years postoperatively.

In our series, five of the seven patients had complete relief of dysphagia within 1 month postoperatively, while two patients obtained gradual improvement over the following 3 months. Similarly, in the studies of DISH-induced dysphagia reported by Stuart [1], McCafferty et al. [7], and Weinshel et al. [18], some patients required several months to recover. The early resolution of symptoms might be a result of eliminating the mechanical obstruction of the esophagus. A slower resolution could be due to more gradual abatement of periesophageal inflammation. In our study, one of the seven patients obtained an only incomplete relief of dysphagia, but had no evidence of concurrent diseases affecting swallowing. This patient was the oldest and had the longest history of dysphagia. Long-term mechanical compression may be associated with the incomplete resolution of swallowing disorders owing to the development of irreversible changes in the tissues around the esophagus [1].

While five out of seven patients with radiological recurrence remained asymptomatic, two of the seven, who were followed for more than 10 years, complained of recurrent dysphagia. The five asymptomatic patients need longer-term follow-up to provide conclusive evidence of the presence or absence of symptomatic recurrence. A dynamic video fluoroscopic study demonstrated that dysphagia accompanied by aspiration was common in patients with osteophytes larger than 10 mm and rare in those with smaller osteophytes [5]. Several pathophysiology of aspiration in patients with DISH have been reported. Osteophytes arising from the anterior face of C3/4 and C4/5 levels are shown to cause intradeglutitive aspiration of Xuids by restricting the laryngeal closure by the epiglottis

during deglutition [11]. Additionally, osteophytes at C5/6 and C6/7 levels are shown to complicate the deglutition of food on the respective side of the pharynx, consequently resulting in a retention of solid food in the pharynx with the possibility of postdeglutitive aspiration [11]. In our study, the mean postoperative increase in size of the largest recurrent osteophyte in each patient was about 1 mm/year. It seems possible that most patients will return to their preoperative condition 14–20 years after surgery because the size of the largest recurrent osteophyte will reach 14–20 mm, which is equal to the preoperative size. In elderly patients, a simple resection procedure might be a good option as it is less likely that symptomatic recurrence would occur in their remaining years. For patients less than 70 years old, who undergo surgical resection, the recurrence of dysphagia will be an issue. Since it was demonstrated that the incidence of postoperative recurrent osteophytes was significantly lower in the fused intervertebral segments, from 1998, we started to add cervical fusions to the simple resection procedures of the osteophytes for patients under 70 years of age. Additionally, since NSAIDS has a preventive effect on heterotopic ossification [17], application of NSAIDS might have a preventive effect on postoperative recurrence of osteophytes especially in patients, who are operated, but not fused. We speculate that the long-term follow-up of those patients would provide a new clue to the prevention of the recurrence of osteophytes in the future.

There are several limitations in the present study. First, the osteophytes seen in the present patients associated with DISH can be either an incomplete form of OALL or a bony production coming from an accompanying osteoarthritis. Second, among the subjects, the differences between those who aspirate and those who did not aspirate have not been elucidated, yet. Future study with more number of patients should be focused on this issue. Third, not all the subjects

underwent the postop swallowing studies. In order to accurately evaluate the short- and long-term results of operations, postop swallowing studies should have been performed periodically. Fourth, it is known that DISH is frequently seen in the context of diabetes. However, since medical records of all cases are not available now, description of such comorbidities in all cases is impossible. Description of those information would have been useful in understanding this pathological condition.

Conclusions

DISH patients with dysphagia are at great risk for recurrence of osteophytes following surgical resection. The surgeons should continue to follow the patients for more than 10 years postoperatively to assess the recurrence of osteophytes and dysphagia.

Acknowledgments We thank Dr. Fuminari Kuze, Department of Otolaryngology, Gifu University School of Medicine, Gifu, Japan, for his contributions regarding the evaluation of swallowing disorders.

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