Surgical management of Diffuse Idiopathic Skeletal Hyperostosis (DISH) causing secondary dysphagia (Narrative review)

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Abstract

Objectives: To summarize the current evidence on surgical treatment for large bridging osteophytes of the anterior cervical spine from Diffuse Idiopathic Skeletal Hyperostosis (DISH). Overview of Literature. In the current review, the surgical treatment of secondary dysphagia from DISH was the most useful treatment. We propose a treatment algorithm for management of this condition because currently there are only case reports and retrospective studies available.

Methods. Literature search was performed using the MeSH terms “Anterior Cervical Osteophyte,” “Diffuse Idiopathic Skeletal Hyperostosis (DISH),” and “Dysphagia” and “Treatment” for articles published between January 2000 and February 2020. PubMed search identified 117 articles that met the initial screening criteria. Detailed analysis identified the 40 best matching articles, following which the full inclusion and exclusion criteria left 11 articles for this review.

Results. Incidence of secondary dysphagia was associated with DISH in elderly patients (average 65 years). The major clinical findings were dysphagia or respiratory compromise, with the most common level of bridging osteophytes of the cervical spine at C3–C5. There were 10 articles on surgical treatment involving anterior cervical osteophytectomy without fusion, 1 for multilevel cervical oblique corpectomy, 1 for anterior cervical discectomy with fusion plus plate, and 1 for anterior cervical osteophytectomy with stand-alone PEEK cage or plus plate. All the cases resulted in significant improvement without recurrence, with only 1 case having post-operative complications. Follow-up duration was 3–70.3 months. Conclusions. Surgical intervention for anterior cervical osteophytectomy appears to result in improved outcomes. However, there could be disadvantages concerning cervical spine motion if cervical osteophytectomy with cervical discectomy and fusion (ACDF) plus plate system is done.

Keywords
diffuse idiopathic skeletal hyperostosis, DISH, dysphagia

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Introduction

The anterior osteophyte is a bony hardening of the ligaments in areas where they attach to the spine. Diffuse Idiopathic Skeletal Hyperostosis (DISH) can be manifested as huge, multilevel anterior osteophytes of the cervical spine. The incidence of DISH is variable and ranges about 10–20% of the general population.¹,² However, DISH is generally asymptomatic. Studies have found that approximately 10%

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of patients older than 65 years commonly had anterior osteophytes of the cervical spine and more than 17% were found with DISH complaints of dysphagia. A previous literature review did not offer guidelines for the surgical treatment of these large anterior osteophytes of the cervical spine. This review article presents the current surgical treatment options for large anterior cervical osteophyte or DISH causing dysphagia.

The authors aim to summarize the current evidence on surgical treatment for large bridging osteophytes of the anterior cervical spine from Diffuse Idiopathic Skeletal Hyperostosis (DISH) that can cause dysphonia, dyspnea, and/or dysphagia. The authors propose a treatment algorithm for management because to date there have been only case reports and retrospective studies.

Methods

Literature search

Literature search was performed to collect the articles published in PubMed using the MeSH terms “Anterior Cervical Osteophyte,” “Diffuse Idiopathic Skeletal Hyperostosis (DISH),” “Dysphagia,” and “Treatment”. Articles published between January 2000 and February 2020 were selected for further screening. Additional manual searches were performed using relevant references in selected articles concerning anterior cervical osteophytes causing dysphagia.

Inclusion and exclusion criteria

Randomized controlled studies, observational studies, and case series were included in this review. There was no preference in the type of study (retrospective/prospective), and all studies had to be published in the English language. Case reports and technical reports were excluded. All the articles were screened for titles and abstracts. Selected articles were studied in detail, and the relevant data were recorded.

Data extraction

The following data were recorded: (1) Study ID: including author(s) and journal name; (2) year of publication; (3) type of study design; (4) study sample size: Number of patients; (5) average patient age (mean ± SD); (6) clinical findings of symptomatic DISH; (7) nationality; (8) radiologic findings (level of the cervical spine where the bridging osteophytes occurred); (9) surgical treatment: Anterior cervical osteophytectomy alone without fusion, multilevel cervical oblique corpectomy, anterior cervical disectomy without plating, anterior cervical discectomy and fusion (ACDF) plus plate, anterior cervical osteophytectomy + stand-alone polyetheretherketone (PEEK) cage without plating, and anterior cervical osteophytectomy + PEEK plus plate; (10) clinical results; and (11) follow-up duration.

Results

The PubMed search resulted in the identification of 117 articles that met the initial screening criteria. Detailed analysis found 40 matching articles; however, when applying the full inclusion and exclusion criteria, only 11 articles were determined to be eligible for this review. All articles reported an incidence of secondary dysphagia associated with DISH in elderly patients (average 65 years). Clinical findings were dysphagia or respiratory compromise with the most common level of bridging osteophytes of the cervical spine at C3–C5. The nationality of the authors showed 3 articles from USA, 3 articles from Germany, 2 articles from Japan, and 1 article each from India, Italy, and United Kingdom. Surgical treatment showed 10 articles for anterior cervical osteophytectomy without fusion, 1 for multilevel cervical oblique corpectomy, 1 for anterior cervical disectomy with ACDF (plus plate), and 1 article for anterior cervical osteophytectomy + stand-alone PEEK cage without plating and anterior cervical osteophytectomy + PEEK plus plate. All of the articles reported significantly improved clinical results following the treatment without recurrence; only 1 study reported post-operative complication, stroke, and Horner’s syndrome, which improved at 6 months. The follow-up duration was 3–70.3 months.

Demographic data analysis

All studies were reviewed and analyzed including by study design, age, nationality and clinical findings associated with DISH that were shown in Tables 1.

Study design. Ten studies were retrospective because DISH was asymptomatic; there were only a small number of patients with DISH-related dysphagia. There were no randomized controlled trial studies for this condition. Only the study of Miyamoto et al. had a prospective study design.

Age. All the studies found that elderly patients were associated with DISH with dysphagia and 3 reported this combined with respiratory compromise. The study by Chacko et al. showed the lowest average age (55 ± 1 year), while Miyamoto et al. and Oppenlander et al. found that age >65 years was a strong factor in the development of DISH.

Nationality. All of the studies showed the nationality of the study for DISH with dysphagia and combined with respiratory compromise. The idiopathic cause of DISH was not related with nationality due to the small sample size.
Clinical findings associated with DISH. The most common clinical presentation was dysphagia. Respiratory compromise associated with moderate to severe dysphagia was reported by Carlson et al.,11 Von der Hoeh et al.,12 and Scholz et al.. Other studies have shown an association between anterior cervical osteophyte causing secondary dysphagia with hoarseness, notably Scholz et al.13 and Ruetten et al.14

Surgical treatment and outcomes

The surgical treatment and clinical outcomes of patients were shown in Tables 2. There were different surgical interventions for DISH with dysphagia or respiratory compromise. The most common level of bridging osteophytes of the cervical spine was C3–C5. The most common surgical treatment was anterior cervical osteophytectomy without fusion: 1 article for multilevel cervical oblique corpectomy, 1 article for anterior cervical discectomy with ACDF plus plate, and 1 article for anterior cervical osteophytectomy with PEEK cage alone without plating or PEEK plus plate. Most of the articles reported significantly improved clinical results without recurrence. However, Miyamoto Kei et al. reported 1 recurrence case; 1 study reported post-operative complications of stroke and Horner’s syndrome, which improved at 6 months. The follow-up duration was 3–70.3 months. Overall, anterior cervical osteophytectomy with or without fusion had significantly good to excellent outcomes with no serious complications of dysphonia or progressive dysphagia.

Discussion

The incidence of cervical anterior osteophytes in the elderly population is about 10–20%.1,2 Symptomatic dysphagia or dysphonia from large anterior cervical osteophyte or DISH causing secondary dysphagia is rare, with a prevalence of 5%.1–4 However, dysphagia permanently impaired the quality of life for eating.4 Radiologic evaluation based on cervical lateral radiographs can quickly identify the likely diagnosis. Computerized tomography (CT) scan and magnetic resonance imaging (MRI) can assess the esophagus, along with a barium swallow test.19 However, patients with dysphagia from other causes, such as neurological dysphagia from stroke, Parkinson’s disease, multiple sclerosis, or dementia, need to be excluded.5–7 In addition, malignancies such as laryngeal or esophageal cancer may be the cause of dysphagia.20–23 In all cases, we recommend consultation with ENT to confirm the cause of dysphagia. Patients diagnosed with anterior cervical osteophyte or DISH causing secondary dysphagia who received surgical intervention had improved clinical outcomes when compared with conservative treatment.5–15 However, observation was recommended when there were no signs or symptoms of dysphagia or dysphonia. When surgery is indicated, the most common operation is anterior osteophyte excision. A standard Smith–Robinson anterior cervical approach is utilized.24–27 Large anterior cervical osteophytes are usually the main cause of compression of the esophagus and other anterior structures. Studies on some controversial surgical protocols found that patients who had operative treatment using these protocols had good improvement.5–15 A study by Ruetten et al.14 recommended the use of prophylactic medications, such as indomethacin (50 mg twice daily for 10 days) or radiation (5 doses of 2 Gray) to prevent recurrence. However, there is no definitive proof that this is necessary. The summary of the advantages and disadvantages of the surgical protocol with anterior cervical osteophytectomy with and without fusion is shown in Table 3. It appears that anterior cervical osteophytectomy alone, without fusion, achieved good relief and significantly improved dysphagia. A relatively simple removal of anterior osteophytes resulted in shorter operative

| Author(s)         | Year | Study design | Sample size | Average age (Mean ± SD.) (years) | Nationality | Clinical findings (diagnosis: DISH)                  |
|-------------------|------|--------------|-------------|----------------------------------|-------------|----------------------------------------------------|
| Chacko et al.9     | 2005 | Retrospective| 3           | 55 ± 1                           | India       | Dysphagia or dysphonia                             |
| Urrutia et al.15   | 2009 | Retrospective| 5           | 71 ± 8                           | USA         | Dysphagia                                          |
| Miyamoto et al.8   | 2009 | Prospective  | 7           | 65 ± 8                           | Japan       | Odynophagia to severe dysphagia                    |
| Oppenlander et al.10 | 2009 | Retrospective| 9           | 65 ± 12                          | USA         | Dysphagia                                          |
| Carlson et al.11   | 2011 | Retrospective| 6           | 68 ± 6                           | USA         | Dysphagia or respiratory compromise                |
| Von der Hoeh et al.12 | 2014 | Retrospective| 6           | 67 ± 5 years                     | Germany     | Dysphagia or respiratory compromise                |
| Yoshioka et al.16  | 2018 | Retrospective| 4           | 67 ± 5 years                     | Japan       | Dysphagia                                          |
| Lui Jonathan et al.17 | 2018 | Retrospective| 6           | 59 ± 16 years                    | UK          | Moderate to severe dysphagia                       |
| Scholz et al.13    | 2019 | Retrospective| 5           | 62 ± 15 years                    | Germany     | Dysphagia, hoarseness or breathing difficulties    |
| Ruetten et al.14   | 2019 | Retrospective| 14          | 57 ± 16 years                    | Germany     | Dysphagia, hoarseness, dyspnea                     |
| Mattioli et al.18  | 2020 | Retrospective| 21          | 70 ± 11 years                    | Italy       | Dysphagia or dysphonia                            |
| Author          | Year   | Study design | Sample size | Radiologic Findings | Surgical treatment | Outcome                                                                 | Mean follow-up |
|-----------------|--------|--------------|-------------|---------------------|--------------------|--------------------------------------------------------------------------|----------------|
| Chacko et al.9   | 2005   | Retrospective| 3           | C2–T1 (1)           | Multilevel cervical oblique corpectomy (3) | Significant improvement (2) Horner’s syndrome (1) improved at 6 months | 12 months      |
| Urrutia et al.15 | 2009   | Retrospective| 5           | C3–C4(3)            | Anterior cervical osteophytectomy without fusion (5) | Significant improvement without recurrence (5) | 59.8 months    |
| Miyamoto Kei et al.8 | 2009 | Prospective | 7           | C2–C7(1)           | Anterior cervical osteophytectomy without fusion (7) | Significant improved (6) recurrence (1) | 9 months       |
| Oppenlander et al.10 | 2009 | Retrospective| 9           | C2–C3(1)           | Anterior cervical osteophytectomy without fusion (9) | Significant improved with full resolution (7) and delayed resolution (2) | 9.8 months     |
| Carlson et al.11 | 2011   | Retrospective| 6           | C2–C7(1)           | 1. Anterior cervical osteophytectomy without plating (4) | Significant improved without recurrence (5) | 3 months       |
|                 |        |              |             | C2–T1 (1)          | 2. Anterior cervical osteophytectomy + tracheostomy (2) | Stroke (1) |                       |
|                 |        |              |             | C3–C6(1)           |                                 |                                                                 |                |
|                 |        |              |             | C3–C7(3)           |                                 |                                                                 |                |
| Von der Hoeh et al.12 | 2014 | Retrospective| 6           | C3–C4 (3)          | 1. Anterior cervical osteophytectomy + PEEK cage stand-alone without plating (4) | Significant improved without recurrence (6) | 23 months      |
|                 |        |              |             | C3–C6 (1)          | 2. Anterior cervical osteophytectomy + PEEK cage plus plate (2) |                                                                 |                |
|                 |        |              |             | C4–C6 (1)          |                                 |                                                                 |                |
|                 |        |              |             | C5–C6 (1)          |                                 |                                                                 |                |
| Yoshioka et al.16 | 2018 | Retrospective| 4           | C3–C4(2)           | 1. Partial anterior cervical osteophytectomy without fusion (3) | Significant improved with full resolution (4) | 12 months      |
|                 |        |              |             | C4–C5(2)           | 2. Extensive anterior cervical osteophytectomy without fusion (1) |                                                                 |                |
| Lui Jonathan et al.17 | 2018 | Retrospective| 6           | C2–T1(1)           | Anterior cervical osteophytectomy without fusion(6) | Significant improved without recurrence (6) | 42.3 months    |
|                 |        |              |             | C3–T1(1)           |                                                                 |                                                                 |                |
|                 |        |              |             | C3–C6(1)           |                                                                 |                                                                 |                |
|                 |        |              |             | C4–C6(2)           |                                                                 |                                                                 |                |
|                 |        |              |             | C4–C7(1)           |                                                                 |                                                                 |                |
| Scholz et al.13  | 2019   | Retrospective| 5           | C3–C5(2)           | 1. Anterior cervical osteophytectomy without plating (2) | Significant improved without recurrence (5) | 70.3 months (one case dies at 5 years after surgery without associated with the treatment) |
|                 |        |              |             | C3–C7(2)           | 2. Anterior cervical osteophytectomy + anterior cervical discectomy and fusion (ACDF) plus plate (3) |                                                                 |                |
|                 |        |              |             | C4–C5(1)           |                                                                 |                                                                 |                |
times when compared with anterior cervical osteophytectomy with fusion (PEEK cage alone/PEEK plus plate/ACDF with plate system).

Recurrent cervical osteophytes were rare and it developed slowly in long-term follow-up over 10 years. Routine additional fusion was therefore discussed not only due to possible operation induced instability, but also because segmental mobility can promote the further development of osteophytes. Surgical resection of cervical osteophytes was a sufficient method for treating spondylogenic dysphagia but prophylaxis of recurrent anterior cervical ossifications was limited experience and no precise guidelines. However, prophylaxis treatment using indomethacin or radiation also appears to be an option.

DISH-related respiratory problems are rare. When present, it was believed to be due to compression of the airway by large hypertrophic anterior cervical osteophytes. Reported clinical symptoms include hoarseness, stridor, and dyspnea to acute respiratory distress. Large osteophytes can potentially compress the posterior pharyngeal wall of the esophagus and trachea. Hypertrophic anterior cervical osteophytes at C4–C5 level was the narrowest region and felt to be the cause of the airway obstruction. The esophagus was the first affected structure. It was under direct compression and entrapment, resulting in dysphagia. Esophageal compromise can then be followed by compression of the trachea that can result in respiratory compromise due to the excessively large osteophytes. Large hypertrophic anterior cervical osteophytes can cause direct compression of the trachea, as well as entrapment and compromise of both laryngeal nerves with bilateral vocal cord immobility. The result was dysphonia and stridor. Patients who have severe aspiration with acute respiratory distress may require urgent tracheotomy for airway management. The surgical excision of the osteophytes in the reported case was successful and effective with improvement in the dysphagia and respiratory discomfort.

| Author et al.14 | 2019 | Retrospective | 14 | C2–C7(2) | 1. Anterior cervical osteophytectomy (12) | Significant improved with full resolution (14) | 50 months |
|-----------------|------|--------------|----|---------|-------------------------------------------|-----------------------------------------------|--------|
| Mattioli et al.18 | 2020 | Retrospective | 21 | Most common at C3–C5 (not identify the level of anterior osteophyte) | Anterior cervical osteophytectomy without fusion (21) | Significant improved with full resolution (16) and delayed resolution (5) | 66 months |

Table 2. Comparison of surgical techniques for DISH anterior cervical osteophytes causing secondary dysphagia.

| Surgical protocol | Advantages | Disadvantages |
|-------------------|------------|---------------|
| Anterior cervical osteophytectomy alone without fusion | 1. Good relief and significant improvement of dysphagia without recurrence 2. Easy to remove anterior osteophyte fragments 3. Shorter operative time and less blood loss 4. Reduce the risk of spinal cord injury, dural tear, or spinal fluid leakage | 1. May require revision surgery in the future for myelopathy or post-operative instability |
| Anterior cervical osteophytectomy with fusion (PEEK cage alone/PEEK plus plate/ACDF with plate system) | 1. Good relief and significant improvement of dysphagia without recurrence 2. Maintain disc space height 3. Reduced post-operative instability | 1. Loss of movement at the fused segment 2. Adjacent segment degeneration 3. Prolonged operative time in multilevel fusions |
Anterior cervical osteophytes or DISH combined with cervical myelopathy requires spinal cord decompression.\(^3\) The AO Spine guidelines (AO Spine North America) and the Cervical Spine Research Society (CSRS) suggest treatment options for CSM.\(^3\),\(^6\) They suggest anterior cervical osteophytectomy without fusion for patients with dysphagia without cervical myelopathy. For patients with DISH and cervical myelopathy, they recommend anterior cervical discectomy and fusion (ACDF) plus plating. The authors propose the treatment algorithm shown in Figure 1.

Overall post-operative complications after anterior cervical spine surgery in current systematic review\(^3\) found dysphagia (5.3%), recurrent laryngeal nerve palsy (1.3%), infection (1.2%), hematoma (1%), CSF leak (0.5%), Horner’s syndrome (0.4%), vertebral artery injury (0.4%) and esophageal perforation (0.2%).\(^3\) However, our review articles in DISH causing secondary dysphagia found less reported in the complications after anterior cervical osteophytectomy. In all patients whom diagnosed with DISH presenting by dysphagia or respiratory compromise, only the study by Chacko et al.\(^9\) reported postoperative Horner’s syndrome, which had spontaneous improvement at 6 months. Another study by Carlson et al.\(^1\) showed post-operative stroke. Post-operative persistent dysphagia was improved during the duration of 3–70.3 months in our review.

All patients with DISH causing secondary dysphagia could be evaluated by internal medicine and ENT physicians for the risks to carry on surgery and conducted airway management before operation. Our recommendations for diminishing and preventing the post-operative complications are urging the surgeon to perform adequate surgical approach-incision, using microscope-assisted surgery for better visualization and decreasing the risk of adjacent structure injuries during anterior cervical osteophytectomy or discectomy such as airways, esophagus, vertebral artery, and CSF leakage. Patients with risk factors (DISH, OPLL, long operative time (more than 5 h), multi-level surgery (more than 3 levels), and therapeutic heparin administration) might relate to acute post-operative retropharyngeal hematoma and airway complications.\(^3\) We highly recommended to check and stop the possible occult bleeding before closure. Effective intraoperative hemostasis after anterior cervical osteophytectomy was required to prevent post-operative retropharyngeal hematoma.\(^3\)

**Clinical case**

A 54-year-old female presented with DISH causing dysphagia with cervical radiculopathy. She had progressive dysphagia over 3 years. Her general physical examination was normal without respiratory compromise. She had positive Spurling’s test (bilateral dermatomal C5 and C6 radiculopathy). Plain radiographs demonstrated prominent anterior cervical osteophytes at C3–6 (Figure 2(a)). Sagittal MRI T2-weighted images showed prominent large anterior cervical osteophytes at C3–6 which compressed the pharyngoesophageal structure (Figure 2(b)). The axial images at C5–C6 (Figure 2(c)) and C6–C7 (Figure 2(d)) showed bilateral foraminal stenosis with C6–C7 nerve root compression.
We used the Smith–Robinson approach to expose the anterior cervical spine from C3 to C6. Large prominent anterior cervical osteophytes were found (Figure 2(e)). We performed an osteophytectomy from C3 to C6, discectomy C5–C7 (Figure 2(f)), and fusion with iliac autogenous bone graft (Figure 2(g)) plus plate (Figure 2(h)). There were no iatrogenic neurovascular injuries, and the alignment was acceptable. Complete bony union was achieved at 6 months post-operatively. The patient had significant improvement clinically and radiographically (Figure 2(h)) without recurrence of dysphagia at 3-years follow-up.

A previous study by Liawrungrueang et al. reported on an 81-year-old patient with a “giant beak-like lesion” from DISH causing dysphagia. The osteophytic lesion was anterior to the C3 and C4 vertebrae, causing compression of the pharyngoesophageal segment. Patient refused anterior cervical osteophytectomy. He had progressive dysphagia and recurrent aspiration pneumonias, impacting his quality of life.

In summary, we examined the current evidence on surgical management of large bridging osteophytes of the anterior cervical spine secondary to DISH. We found that surgical intervention appears to be safe, effectiveness and halts the progression of disease. We believe that our algorithm will guide surgeons in their decision-making process. We fully expect that this algorithm will be modified and revised in the future, as more knowledge and novel treatments regarding this condition becomes available.

**Conclusion**

We conclude that large osteophytes due to DISH can cause dysphagia, but it remains unclear why some with such osteophytes remain asymptomatic, whereas others with smaller osteophytes are symptomatic. Surgical intervention with anterior cervical osteophytectomy has the advantage of improved outcomes. Cervical spine motion is likely to be impacted if a fusion is also performed. However, the fusion may prevent a recurrence of the osteophytes. Further
randomized controlled studies and studies with long-term outcomes are required to determine the optimal procedure for anterior cervical osteophytectomy with or without fusion.

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K. Daniel Riew and Torphong Bunmaprasert: Supervision, Writing - Review & editing; Jakkrit Keeratiruangrong and Nantawit Sukantathan: Resources, Data curation; Wongthawat Liawrungrueang: Conceptualization, Methodology, Visualization, Writing-original draft; K. Daniel Riew and Torphong Bunmaprasert: Supervision, Writing - Review & editing. All authors have read and approved the final manuscript.

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