Annals of Medicine and Surgery 72 (2021) 103120

Contents lists available at ScienceDirect

Annals of Medicine and Surgery

journal homepage: www.elsevier.com/locate/amsu

Case Report

Anterior cervical spine surgery for treatment of secondary dysphagia associated with cervical myelopathy in patient with Forestier's disease

Wongthawat Liawrungrueang, Peem Sarasombath, Titinat Maihom, Waroon Tantivorawit, Nantawit Sugandhavesa, Torphong Bunmaprasert *

Department of Orthopaedics, Faculty of Medicine, Chiang Mai University, Chiang Mai, 50200, Thailand

A R T I C L E I N F O

Keywords:
ACDF
Dysphagia
Diffuse idiopathic skeletal hyperostosis
DISH
Forestier's disease

A B S T R A C T

Introduction: and importance: Forestier's disease, also known as a vertebral ankylosing hyperostosis or Diffuse Idiopathic Skeletal Hyperostosis (DISH), is a non-inflammatory enthesopathy that affects primarily elderly males and ossifies the anterolateral spine while sparing the intervertebral discs and joint spaces, especially at the cervical spine. Forestier's disease has resulted in the growth of large anterior cervical osteophytes that may compress the pharyngoesophageal region, producing dysphagia. However, symptomatic Forestier's disease presenting with dysphagia and cervical myelopathy is rarely observed.

Case presentation: A 48-year-old male presented with progressive dysphagia and cervical myelopathy. Based on the presence of radiographic study, Forestier's disease was suspected. Large anterior cervical osteophytes at C4–C6 levels compressed the pharyngoesophageal structure posteriorly. Multilevel degenerative discs compressing the C4 to C6 spinal cord were also seen on sagittal MRI T2-weighted images. Anterior cervical osteophytectomy with anterior cervical discectomy and fusion (ACDF) were performed. The patient made a complete neurological recovery and had no recurrent symptoms at the 5-year follow-up. The patient was extremely satisfied with this treatment and can improved his quality of life (QOL).

Clinical discussion: Treatment of symptomatic Forestier's disease with secondary dysphagia and cervical myelopathy is rare evidenced by the dearth of reports on surgical treatment. Surgical intervention appears to be safe, effective, and able to halt disease progression.

Conclusion: Anterior cervical osteophytectomy combined with ACDF with plate fixation is a preferred technique in both neural decompression and swallowing improvement. Surgical intervention, we consider, provides superior results than prolonged non-surgical treatments.

1. Introduction

Skeletal-related dysphagia is uncommon. Forestier's disease, also known as vertebral ankylosing hyperostosis or Diffuse Idiopathic Skeletal Hyperostosis (DISH), can result in anterior cervical osteophytes. A large anterior cervical osteophyte can cause otolaryngological symptoms including dysphagia, dysphonia, and dyspnea by compressing the pharyngoesophageal region [1]. Secondary dysphagia is a complication of anterior cervical hyperosteophytosis, which requires surgery. Previous literature has demonstrated a less instance of osteophytes compressing the pharyngoesophageal region anterior to the C4 and C6 vertebral spine. The occurrence of symptomatic Forestier's disease with subsequent dysphagia and cervical myelopathy is even less frequent [1, 2]. We describe the results of anterior cervical spine surgery on a middle-aged man who had dysphagia due to large anterior osteophytes on the C4–C6 spine and concurrent cervical myelopathy caused by multilevel disc degeneration and compression of the C4–C7 spinal cord. This work has been reported in accordance with SCARE criteria [3].

2. Case presentation

A 48-year-old Buddhist monk has been suffering from worsening dysphagia and upper extremity weakness for 6 months. His underlying disease was hypertension. He had no previous surgery or history of cervical spine injury. The patient did not smoke, consume alcohol, or use recreational drugs. Physical examination found his initial vital signs,

* Corresponding author.
E-mail addresses: mint11871@hotmail.com (W. Liawrungrueang), peems13063@gmail.com (P. Sarasombath), yournine@gmail.com (T. Maihom), maing131@yahoo.com (W. Tantivorawit), nantawitsu@gmail.com (N. Sugandhavesa), torphong.b@cmu.ac.th (T. Bunmaprasert).

https://doi.org/10.1016/j.amsu.2021.103120
Received 29 October 2021; Received in revised form 21 November 2021; Accepted 21 November 2021
Available online 23 November 2021

mental state, gait and ambulatory function to be normal. He had decreased hand function and impaired bilateral upper extremity and grip strength (motor power grade III). Hoffmann's sign, inverted radial reflex, and Trömner sign were all positive for cervical myelopathy. Large anterior osteophytes at the C4–C6 level of the cervical spine appeared to be anterior cervical hyperosteophytosis on a plain x-ray (Fig. 1A,B). The MRI diagnosis was based on ossifications along the anterolateral side of the C4–C6 vertebrae. The most probable diagnosis was Diffused Idiopathic Skeletal Hyperostosis (DISH) (Fig. 1C,D) with bulging discs at the C4–C6 levels, resulting in cervical spinal stenosis and significant spinal cord compression (Fig. 2). Dysphagia was most likely caused by a massive bulging ossification at the C4–C6 level, which caused mass effect on the posterior pharyngeal wall. The cause of dysphagia was investigated and confirmed by ENT specialists. Anterior Cervical Discectomy and Fusion (ACDF) with plate fixation was performed simultaneously with surgical osteophytectomy by experienced spine surgeon (TB). The anterior cervical spine from C4 to C6 was exposed using Smith–Robinson approach. Intraoperatively, large protruding anterior cervical osteophytes were identified. We performed an anterior osteophytectomy at C4 to C6 levels, discectomies from C4–C5 to C6–C7, and spinal fusion using iliac autogenous bone graft and a locking plate from C4 to C7. Dysphagia improved substantially after surgery. Complete solid bony union was achieved at six months. At 5-year follow-up, the patient's radiographic imaging (Fig. 3) and clinical condition (Fig. 4) had significantly improved without return of dysphagia or myelopathy symptoms. The patient was extremely satisfied with treatment and could improve his quality of life (QOL).

3. Discussion

Large anterior cervical osteophytes caused by Forestier's disease or DISH can compress the pharyngoesophageal region, causing the symptoms including dysphagia, dysphonia, and dyspnea [1]. In the elderly Asian cultures, the prevalence of cervical anterior osteophytes ranges from 10% to 25% [1,4,5]. DISH patients, on the other hand, are usually asymptomatic. Anterior osteophyte of the cervical spine was identified in 10% of individuals over 65 years old, while DISH symptoms of dysphagia were observed in more than 17% [4,5]. Osteophytes at the C4-6 of the cervical spine can contribute to dysphagia by causing anomalies in the epiglottis tilt mechanism. Sagittal or lateral radiographs frequently show anterior cervical hyperosteophytosis, which is the cause of the dysphagia and may necessitate surgical treatment [1]. The surgical treatment for symptomatic anterior cervical osteophytes caused by Forestier's disease should be an anterior cervical osteophytectomy without fusion [6]. Moreover, the surgical indications for ACDF include persistent or recurrent arm discomfort or numbness that has not responded to conservative therapy and presence of neurologic deficit (myelopathy) [1]. In the patients with cervical myelopathy caused by disc compression, appropriate discectomy and fusion are also required [7]. In this case, the main indication for surgical intervention was disabling cervical myelopathy and dysphagia.

Spinal cord decompression is required for patients with anterior cervical osteophytes or DISH coupled with cervical myelopathy. Posterior cervical decompressive surgery is an alternative treatment for multilevel cervical myelopathy. In contrast, anterior surgery provides direct neural decompression concurrent with osteophyte removal. Treatment solutions for cervical spondylotic myelopathy are suggested by the AO Spine recommendations and the Cervical Spine Research Society (CSRS). For individuals with dysphagia but no cervical myelopathy, they recommend anterior cervical osteophytectomy without fusion [7,8]. Anterior cervical discectomy and fusion (ACDF) with plating is recommended for individuals with DISH and cervical myelopathy [7].

A review of the literature and case reports showed very few cases of secondary dysphagia from Forestier's disease following anterior cervical osteophytectomy with ACDF plus plate (Table 1). Following anterior cervical osteophytectomy and ACDF with PEEK cage plus plate, three studies [2,9,10] showed secondary dysphagia caused by Forestier's disease. All patients who had surgical treatment improved significantly, with no recurrence of dysphagia. Only one report, Ruetten et al. [2] performed anterior cervical osteophytectomy and ACDF plus plate which was similar to the current study. To avoid segmental instability and osteophyte recurrence, an osteophytectomy with fusion and a plate system may be necessary. Postoperative administration of oral indomethacin (50 mg twice day for 10 days) and radiotherapy are occasionally recommended as a prophylaxis against recurrent DISH or anterior cervical osteophytes, although this treatment is controversial [2]. In summary, our review of current research on surgical treatment of massive bridging osteophytes of the anterior cervical spine caused by Forestier's disease. We found that surgical management appears to be safe and successful in halting disease progression.

4. Conclusion

Surgical intervention in cases of Forestier's disease or secondary dysphagia associated with cervical myelopathy (DISH), including anterior cervical osteophytectomy combined with ACDF plus plate fixation, provides better outcomes than non-surgical options although fusion can result in limitation of cervical spine motion.

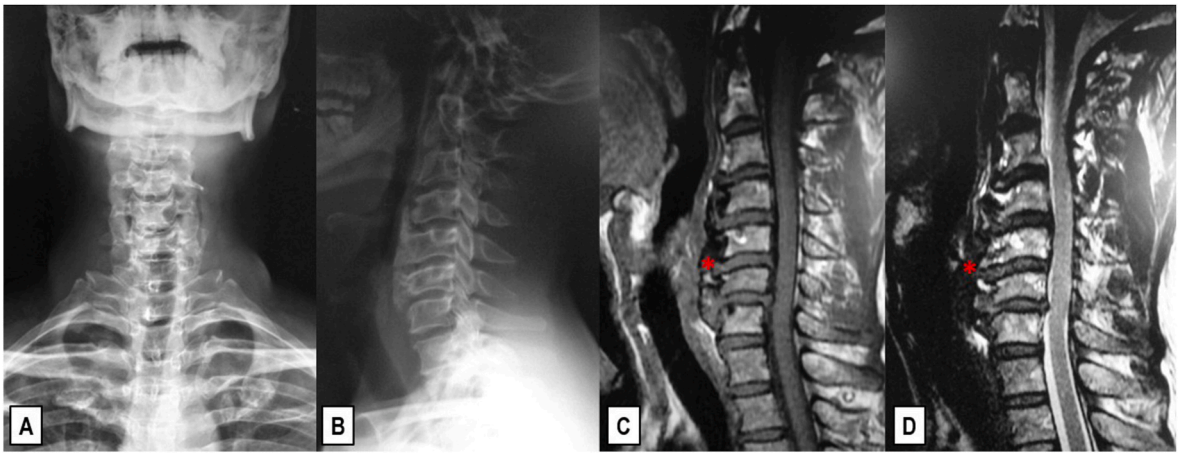

Fig. 1. Radiographic study of large anterior cervical osteophytes caused by Forestier's disease; X-ray in AP-view (A) and Lateral view (B), Sagittal MRI T1-weighted (C) and T2-weighted image of cervical spine (D) demonstrated anterior cervical osteophytes (red asterisk) at C5-6 compressing the pharyngoesophageal structure. (For interpretation of the references to colour in this figure legend, the reader is referred to the Web version of this article.)

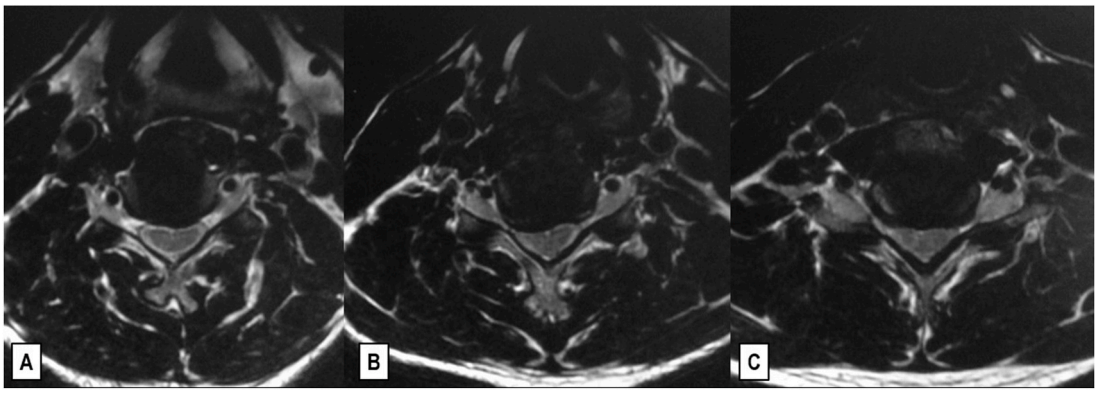

Fig. 2. Radiographic MRI showed cervical spinal cord compression; Axial MRI T2-weighted images of cervical spine at C4/5 (A), C5/6 (B) and C6/7 (C) level.

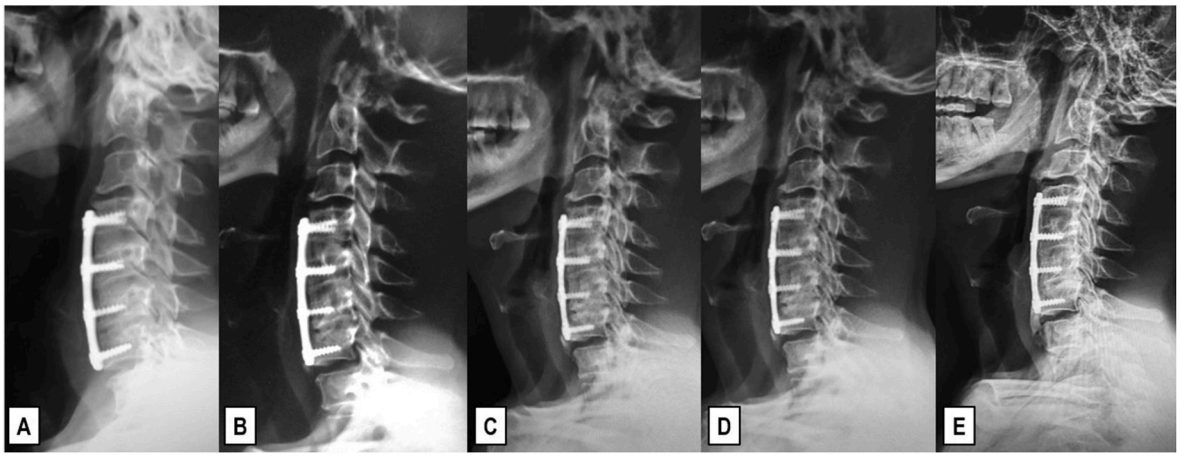

Fig. 3. Radiographic x-ray outcome after anterior osteophytectomy combined with ACDF plus plate fixation at 3 months (A), 6 months (B), 1 year (C), 3 years (D) and 5 years (E) follow up.

Fig. 4. Hand function improved at 6-month after surgery improved and no recurrent myelopathic symptom appeared at 5-year follow-up.

Table 1
Reports of dysphagia from Forestier's disease following anterior cervical osteophytectomy with anterior cervical discectomy and fusion (ACDF) plus plate fixation published in the English language.

Author	Year	Diagnosis	Number of Cases	Preoperative symptoms	Levels of anterior osteophyte	Operative procedure(s)	Final clinical outcome	Mean follow-up
Von der Hoeh et al. [9]	2014	Forestier's disease	2	Dysphagia, weight loss and neck pain	C3–C4 (1) C4–C6 (1)	Anterior cervical osteophytectomy and ACDF (PEEK cage) plus plate	Significant improved without recurrence dysphagia	2 years
Scholz et al. [10]	2019	Forestier's disease	2	Dysphagia, hoarseness and chronic neck pain	C3–C5(1) C4–C5(1)	Anterior cervical osteophytectomy and ACDF (PEEK cage) plus plate	Significant improved without recurrence dysphagia	4.5 years
Ruetten et al. [2]	2019	Forestier's disease	3	Dysphagia and neck pain	C2-T1(1) C3-T1(1) C4–C6(1)	1. Anterior cervical osteophytectomy and ACDF plus plate (2) 2. Anterior cervical osteophytectomy and ACDF (PEEK cage) plus plate (1)	Significant improved without recurrence dysphagia	4 years
This case	2021	Forestier's disease	1	Dysphagia and cervical myelopathy	C4–C6	Anterior cervical osteophytectomy and ACDF plus plate	Significant improvement of hand function without recurrence of dysphagia	5 years

Abbreviations; PEEK: Polyetheretherketone.

Provenance and peer review

Not commissioned, externally peer-reviewed.

Ethical approval

This case report was approved by the Institutional Review Board, Faculty of Medicine, Chiang Mai University.

Source of funding

Funding for this research was provided by the Faculty of Medicine, Chiang Mai University, Chiang Mai, Thailand. The funders had no role in the study design, data collection and analysis, decision to publish or preparation of the manuscript.

Author contributions

Wongthawat Liawrungrueang: Conceptualization, Methodology, Visualization, Writing -original draft.

Peem Sarasombath: Resources, Data curation.

Titinat Maihom: Resources, Data curation.

Waroon Tantivorawit: Resources, Data curation.

Nantawit Sugandhavesa: Resources, Data curation.

Torphong Bunmaprasert: Data curation, Writing - review & editing, Supervision.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in Chief of this journal upon request.

Registration of research studies

None.

Guarantor

Torphong Bunmaprasert, MD., Associate Professor.

Declaration of competing interest

None.

Acknowledgments

The authors would like to express their sincere thanks to Dr. G. Lamar Robert, Ph.D., and Assoc. Prof. Dr. Chongchit Sripun Robert, Ph. D., for editing the English manuscript. We would also like to express our thanks to the Research Unit, Department of Orthopaedics, Faculty of Medicine, Chiang Mai University for their support.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.amsu.2021.103120.

References

[1] T. Bunmaprasert, J. Keeratiruangrong, N. Sugandhavesa, K.D. Riew, W. Liawrungrueang, Surgical management of diffuse idiopathic skeletal hyperostosis (DISH) causing secondary dysphagia (Narrative review), J. Orthop. Surg. 29 (2021), https://doi.org/10.1177/23094990211041783, 23094990211041784.

[2] S. Ruetten, X. Baraliakos, G. Godolias, M. Komp, Surgical treatment of anterior cervical osteophytes causing dysphagia, J. Orthop. Surg. 27 (2019), https://doi.org/10.1177/2309499019837424, 2309499019837424.

[3] R.A. Agha, T. Franchi, C. Sohrabi, G. Mathew, A. Kerwan, SCARE Group, The SCARE 2020 guideline: updating consensus surgical CAse REport (SCARE) guidelines, Int. J. Surg. 84 (2020) 226–230, https://doi.org/10.1016/j.ijsu.2020.10.034.

[4] M. Uehara, J. Takahashi, S. Ikegami, R. Tokida, H. Nishimura, N. Sakai, et al., Prevalence of diffuse idiopathic skeletal hyperostosis in the general elderly population: a Japanese cohort survey randomly sampled from a basic resident registry, Clin Spine Surg 33 (2020) 123–127, https://doi.org/10.1097/BSD.0000000000000919.

[5] S.-K. Kim, B.-R. Choi, C.-G. Kim, S.-H. Chung, J.-Y. Choe, K.-B. Joo, et al., The prevalence of diffuse idiopathic skeletal hyperostosis in Korea, J. Rheumatol. 31 (2004) 2032–2035.

[6] F. Mattioli, M. Ghirelli, M. Trebbi, M. Silvestri, L. Presutti, M. Fermi, Improvement of swallowing function after surgical treatment of diffuse idiopathic skeletal hyperostosis: our experience, World Neurosurg 134 (2020) e29–36, https://doi.org/10.1016/j.wneu.2019.08.124.

[7] M.G. Fehlings, L.A. Tetreault, K.D. Riew, J.W. Middleton, B. Aarabi, P.M. Arnold, et al., A clinical practice guideline for the management of patients with degenerative cervical myelopathy: recommendations for patients with mild, moderate, and severe disease and Nonmyelopathic patients with evidence of cord compression, Global Spine J. 7 (2017) 70S–83S, https://doi.org/10.1177/2192568217701914.

[8] J.R. McCormick, A.J. Sama, N.C. Schiller, A.J. Butler, C.J. Donnally, Cervical spondylotic myelopathy: a guide to diagnosis and management, J. Am. Board Fam. Med. 33 (2020) 303–313, https://doi.org/10.3122/jabfm.2020.02.190195.

[9] N.H. von der Hoeh, A. Voelker, J.S. Jarvers, J. Gulow, C.E. Heyde, Results after the surgical treatment of anterior cervical hyperostosis causing dysphagia, Eur. Spine J. 24 (Suppl 4) (2015) S489–S493, https://doi.org/10.1007/s00586-014-3507-4.

[10] C. Scholz, Y. Naseri, M. Hohenhaus, U. Hubbe, J.-H. Klingler, Long-term results after surgical treatment of diffuse idiopathic skeletal hyperostosis (DISH) causing dysphagia, J. Clin. Neurosci. 67 (2019) 151–155, https://doi.org/10.1016/j.jocn.2019.05.057.