

## Introduction

Diffuse idiopathic skeletal hyperostosis (DISH) is a non-inflammatory systemic condition characterized by calcification and ossification of tendons, ligaments, and entheses of the axial and peripheral skeleton<sup>1,2</sup>. The disorder predominantly affects males and increases in prevalence with aging<sup>3</sup>. The anterior spine is commonly affected, often involving the anterior longitudinal ligament and sparing the intervertebral disk space. Ossification and the eventual formation of osteophytes can lead to multiple clinical manifestations including decreased range of motion, radiculopathy, myelopathy, and dysphonia<sup>4</sup>. Dysphagia caused by mechanical compression is a rare manifestation of the disease presenting in approximately 0.1% to 4% of cases<sup>5</sup>. Operative intervention is most commonly accomplished via osteophyctomy through an anterior approach to the cervical spine<sup>4</sup>.

We present a case of a patient who failed extensive medical modalities and underwent surgical intervention for dysphagia with successful symptom resolution.

## Case Report

An 84-year-old male presented to the medical service with shortness of breath and dysphagia to solid and liquids. He had been previously hospitalized for dysphagia, with less severity. He had undergone multiple EGD's and dilations with decreased symptom resolution from each procedure. A barium swallow study demonstrated esophageal impingement by anterior osteophytes, most significantly at C4-5. Computed tomography obtained during prior hospitalizations demonstrated increasing anterior osteophytes (Figure 1 and 2). Due to the chronicity of his symptoms, failure to significantly improve from GI interventions, and a swallow study demonstrating esophageal impingement it was determined that he would likely benefit from surgical intervention.

### Past Medical Hx:

- Non-smoker, GERD, Hiatal Hernia
- Candida Esophagitis, HIV negative
- Chronic Kidney Disease, Hypertension, Hyperlipidemia

### Past Surgical Hx:

- EGD with Esophageal Dilatation x 3, Appendectomy

### Management:

- C3–C6 Osteophyctomy with partial C3–C6 corpectomies  
**\*Intraoperative findings:** Extensive midline and lateral bone spurs, resection required burring approximately 2.5cm to 3cm of bone to reach vertebral bodies
- Cervical collar and a French drain were placed postop. Drain removed POD #2.
- Secondary to mildly increased stridor he received 4 doses of IV dexamethasone.
- Discharged on a pureed food diet with speech therapy and dietary follow up
- At 3-week postop visit noted marked improvement of swallowing and PO intake
- Repeat fluoroscopic swallow study demonstrated mild pyriform retention without aspiration. Improved compared to previous study preoperatively.
- Approximately at 6 months postop he was tolerating a regular diet with complete resolution of dysphagia symptoms. A repeat barium swallow obtained at that time demonstrated no signs of aspiration or dysphagia.

## Imaging

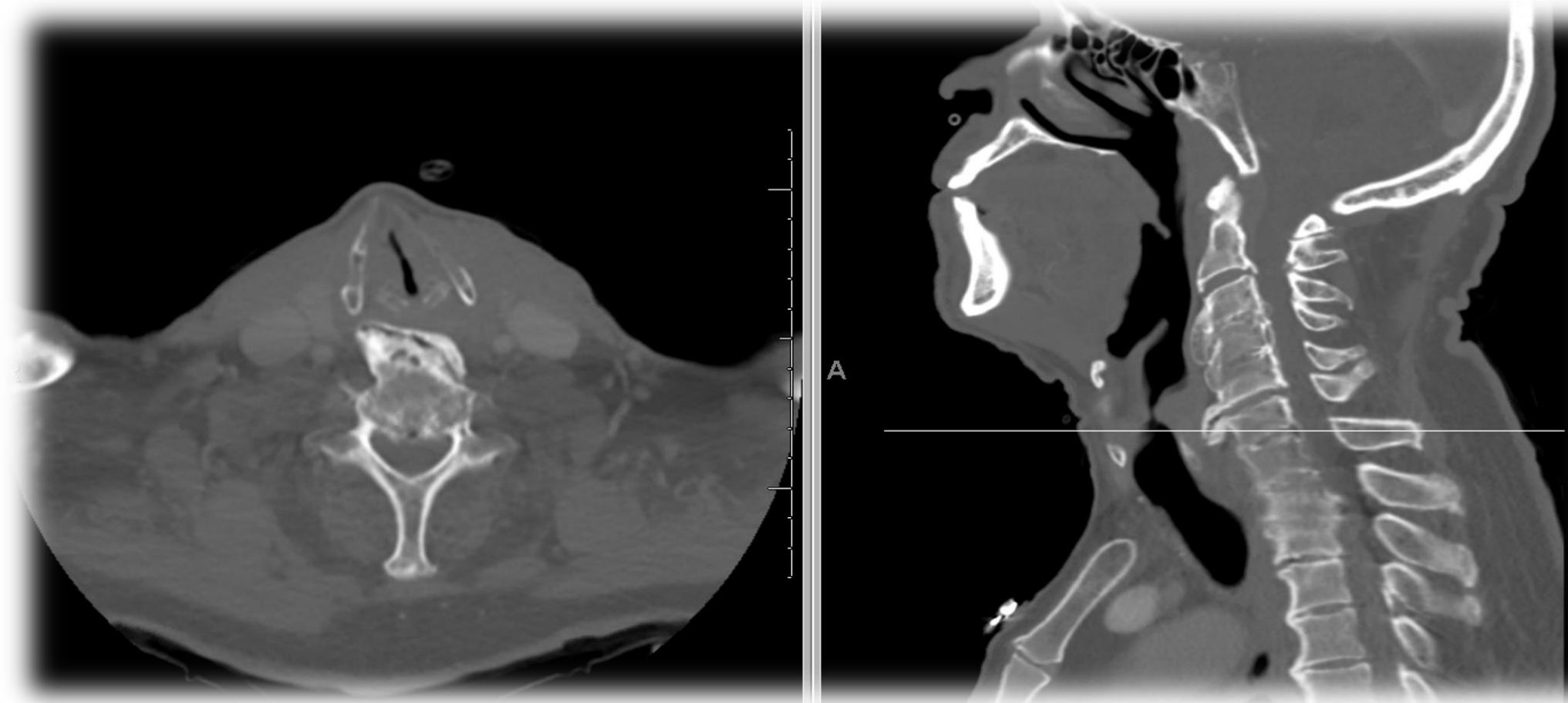


Fig. 1: CT axial and sagittal, respectively, of cervical spine two years prior to surgical intervention



Fig. 2: CT axial and sagittal, respectively, of cervical spine approximately a year prior to intervention demonstrating large anterior osteophytes from C3 – C6

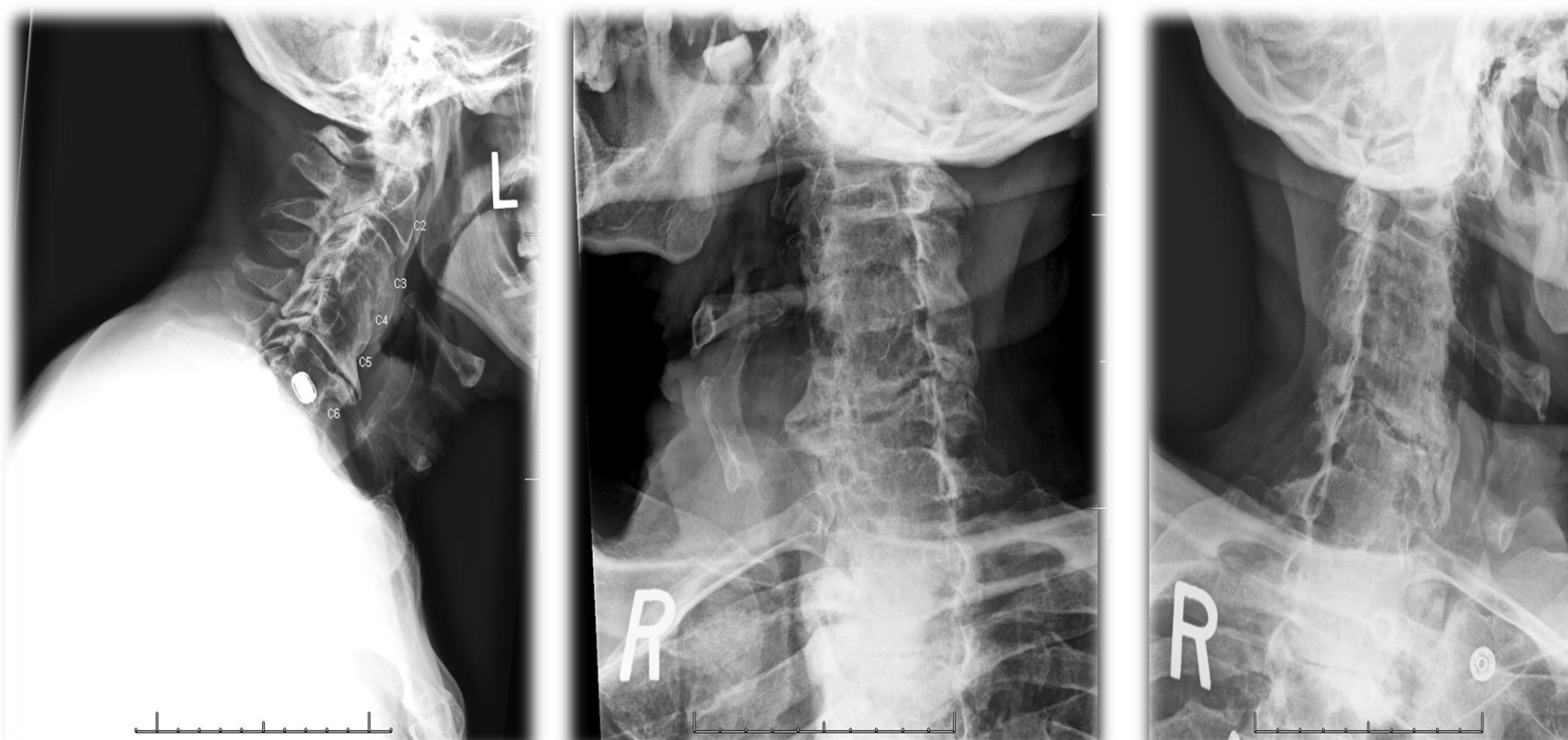


Fig. 3: Lateral and oblique cervical spine radiographs obtained one week preoperatively

## Discussion

The diagnosis of DISH is established through the utilization of radiographic and CT imaging of the spine. Initial diagnostic criteria required the involvement of four contiguous vertebrae with “flowing” osteophytes without involvement of the intervertebral disk.<sup>2</sup> The diagnostic criteria has continued to evolve over time, however the exact pathophysiology remains uncertain. A genetic component is hypothesized as the cause of DISH as multiple metabolic imbalances have been reported in close association. An increase in the cases of DISH is expected due to the enlarging presence of metabolic disorders in the general population.<sup>5,6</sup>

Numerous clinical manifestations as a result of DISH have been reported and are widespread as well as diverse. Dysphagia is a relatively uncommon presenting symptom as it develops insidiously due to the progression of anterior cervical spine osteophytes.<sup>7,8</sup> Osteophytes are commonly encountered at C4-C7, causing mechanical compression of the esophagus. A systematic review of reported cases of DISH with dysphagia showed a predilection for males and the elderly.<sup>9</sup> Approximately 10.5% of dysphagia in adults over 60 may be secondary to DISH.<sup>6</sup>

Currently, an established standard of care for the treatment of DISH does not exist. The literature has shown success with both nonoperative and operative management. It is suggested that the adjunct use of anti-inflammatories and medical management of metabolic disorders are beneficial in overall treatment.<sup>10</sup> Recent retrospective and prospective studies have demonstrated improved symptom resolution with surgical intervention for dysphagia compared to nonoperative management.<sup>11-13</sup> This raises the question if earlier surgical intervention may be more beneficial for these patients. As with any surgical procedure there are associated risk. In particular, all patients with DISH undergoing operative intervention have a risk of osteophyte and dysphagia recurrence postoperatively.

## Conclusion

Diffuse idiopathic skeletal hyperostosis (DISH) as a cause of dysphagia remains a rare occurrence. It is important to undergo a thorough workup by a gastroenterologist or otorhinolaryngologist prior to considering surgical intervention. Operative and nonoperative treatments have both demonstrated positive outcomes. The risk and benefits associated with surgical intervention should always be discussed. Patients that are deemed appropriate surgical candidates may benefit from an osteophyctomy; however, there is always a risk of symptom recurrence. The utilization of a multidisciplinary approach that included speech and diet therapy are essential to attain adequate nutrition and symptom relief.

## References

1. Forestier J, Rotes-Querol J. Senile ankylosing hyperostosis of the spine. *Ann Rheum Dis.* 1950;9(4):321–330
2. Resnick D, Shaul SR, Robins JM. Diffuse idiopathic skeletal hyperostosis: Forestier's disease with extraspinal manifestations. *Radiology.* 1975;115(3):513–524
3. Mader R. Clinical manifestations of diffuse idiopathic skeletal hyperostosis of the cervical spine. *Semin Arthritis Rheum.* 2002;32(2):130–135.
4. Verlaan JJ, Boswijk PF, de Ru JA, Dhert WJ, Oner FC. Diffuse idiopathic skeletal hyperostosis of the cervical spine: an underestimated cause of dysphagia and airway obstruction. *Spine J.* 2011;11(11):1058–1067.
5. Chung YS, Zhang HY, Ha Y, Park JY. Surgical Outcomes of Dysphagia Provoked by Diffuse Idiopathic Skeletal Hyperostosis in the Cervical Spine. *Yonsei Med J.* 2020;61(4):341–348.
6. Mader R. Diffuse idiopathic skeletal hyperostosis: a distinct clinical entity. *Isr Med Assoc J.* 2003;5(7):506–508.
7. Ghammam M, Houas J, Bellakhdher M, Abdelkefi M. Dysphagia revealing diffuse idiopathic skeletal hyperostosis: report of two cases and literature review. *Pan Afr Med J.* 2019;32:189. Published 2019 Apr 17
8. Mattioli F, Ghirelli M, Trebbi M, Silvestri M, Presutti L, Fermi M. Improvement of Swallowing Function After Surgical Treatment of Diffuse Idiopathic Skeletal Hyperostosis: Our Experience. *World Neurosurg.* 2020;134:e29–e36.
9. Dutta S, Biswas KD, Mukherjee A, et al. Dysphagia due to forestier disease: three cases and systematic literature review. *Indian J Otolaryngol Head Neck Surg.* 2014;66(Suppl 1):379–384
10. Mader R, Verlaan JJ. Diffuse idiopathic skeletal hyperostosis: clinical features and pathogenic mechanisms. *Nat Rev Rheumatol.* 2013;9(12):741–750
11. Urrutia J, Bono CM. Long-term results of surgical treatment of dysphagia secondary to cervical diffuse idiopathic skeletal hyperostosis. *Spine J.* 2009; 9(9):e13–17
12. Scholz C, Naseri Y, Hohenhaus M, Hubbe U, Klingler JH. Long-term results after surgical treatment of diffuse idiopathic skeletal hyperostosis (DISH) causing dysphagia. *J Clin Neurosci.* 2019;67:151–155.
13. Miyamoto K, Sugiyama S, Hosoe H, Iinuma N, Suzuki Y, Shimizu K. Postsurgical recurrence of osteophytes causing dysphagia in patients with diffuse idiopathic skeletal hyperostosis. *Eur Spine J.* 2009;18(11):1652–1658.