

CASE REPORT

Anterior cervical osteotomy of diffuse idiopathic skeletal hyperostosis lesions with computer-assisted navigation surgery: A case report

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Key Clinical Message

Diffuse idiopathic skeletal hyperostosis (DISH) involves spine ligament ossification. Computer-assisted navigation (CAN) effectively aids complex surgeries, such as anterior cervical osteotomy, to alleviate progressive DISH-related dysphagia.

Abstract

We describe a 68-year-old man with sudden onset dysphagia to both solids and liquids. Radiographic Imaging revealed DISH lesions from C2 down to the thoracic spine. The patient was successfully treated with CAN anterior osteotomy and resection of DISH lesions from C3–C6 and had complete symptom relief within 2 weeks post-operatively.

KEYWORDS

computer-assisted navigation, DISH, dysphagia, orthopedics, osteotomy

1 | INTRODUCTION

Diffuse Idiopathic Skeletal Hyperostosis (DISH), also known as Forestier disease, is a skeletal condition characterized by ossification of ligaments, tendons, and entheses near the cervical and thoracic vertebrae, with a prevalence ranging from 4% to 42% depending on the study population.^{1–4} DISH in the cervical spine is frequently asymptomatic and until recently had unclear diagnostic criteria. Consequently, the disease has been underdiagnosed and understudied.^{1–6} When DISH lesions are present in the cervical spine, symptoms typically manifest in males over 50 and commonly include dysphagia, dysphonia, dyspnea, cervical spine pain, and neurological signs.^{1,2,5–9}

Dysphagia is indicated as the most common symptom in DISH patients, affecting between 17% and 25% of patients.^{2,5} Although dysphagia is regularly treated conservatively, DISH-related dysphagia may require surgical intervention due to its advancing character.^{5,6}

Lofrese et al.⁷ adopted a multi-center approach to determine DISH surgical indications, and suggested targeted bone resections for elderly DISH patients, whereas younger patients with DISH may necessitate broader decompressions. However, existing literature on surgical treatment for DISH primarily consists of case reports.^{3,8,9}

Computer-assisted navigation (CAN) is an established tool in spine surgery, enabling pre- and intraoperative spinal imaging registration to provide immediate visual

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feedback to surgeons.¹⁰ Numerous studies have demonstrated that CAN enhances cervical spine surgery, leading to superior pedicle screw accuracy and fewer complications compared to non-navigated procedures.^{10–13} Precision in cervical spine pedicle screw placement is crucial due to small pedicles and potential catastrophic consequences from minor deviations, such as vertebral artery or spinal cord injury.^{10,11,13} CAN is beneficial for anatomically complex procedures, such as spine tumor resection, facilitating optimized resection while minimizing trauma.¹⁴ We present the first case of DISH-related dysphagia treated via anterior cervical osteotomy and resection surgery with CAN.

The patient was informed that data from this case would be submitted for publication, and he provided consent.

2 | DETAILED CASE HISTORY AND REPORT

A 68-year-old man, with a medical history of hypertension and hypercholesterolemia, presented with chief complaints of dysphagia to solids and liquids. Prior to his visit, he was evaluated in a separate institution where he was recommended to undergo a percutaneous endoscopic gastrostomy (PEG) procedure, where a feeding tube would allow him to receive nutrition directly through the stomach. During presentation day, the patient reported recent-onset neck pain, lower back pain from a previous right-sided hernia repair in 2017, and mild right hip discomfort despite having a total hip arthroplasty in 2019. The patient denied daily medication use aside from occasional aspirin. On physical exam, it was noted that the patient adjusted his head laterally to facilitate swallowing.

Cervical spine radiographs revealed extensive structural abnormalities, including diffuse DISH with non-marginal syndesmophytes anteriorly. Syndesmophytes occupied approximately 75% of C3's vertebral body diameter, with accompanying C2–C3 facet joint ankylosis, and ankylosis from C3 to the thoracic spine (Figure 1). Proliferative DISH-type lesions compressed the pharyngeal and esophageal structures, causing dysphagia. Large osteophytes were found at C3–C4, C4–C5, and C5–C6 in pre-operative axial (Figure 1C,D,F,G) and sagittal CT (Figure 1B,E) scans and lateral X-ray scan (Figure 1A). Due to the worsening dysphagia, pain, and complex cervical deformity, the patient was recommended for anterior osteotomy and resection of C3–C6 DISH lesions with CAN.

Using CAN in the cervical spine provided real-time anatomical feedback, aiding targeted osteophyte removal and decompression of the C3–C6 region, while safeguarding critical neural and vascular structures. Surgery

utilizing CAN proceeded without complication and was well-tolerated by the patient with adequate resection of bony lesions (Figure 2A–D).

3 | SURGICAL PROCEDURE

The patient was positioned supine, and the anterior cervical region was prepared using standard sterile techniques (Figure 3). Prior to the surgery, a cervical spine CT scan was obtained and uploaded onto the CAN software. Subsequently, co-registration of pre-operative anatomy and instrumentation was defined for the area of interest after placement of a reference array (Figure 4). Real-time feedback of patient anatomy and trajectory was observed on the navigation monitor, facilitated by a direct “line-of-sight” from the array to the navigation camera (Figure 4).

A left-sided Smith-Robinson anterior approach was employed from C3–C6. Retractors were placed, and a Caspar pin was inserted around the C4 level. CAN was used to define points on lateral and oblique fluoroscopy, which were verified intraoperatively. The operative level was confirmed using fluoroscopy and a navigated probe.

Large anterior osteophytic bones were resected, and several lesions were found to be deeper on the anterior and posterior dimensions than the 14 mm Caspar pin insertion. In some regions, the lesions were displaced to the right, and in others, they were paramedian on the left, up to C3 cranially. The remaining bone formation from C3 to C6 was removed using a 4 mm steel bur to complete the primary resection.

An adequate bony resection was confirmed with a 3D fluoroscopic image (Figure 5). A small osteophyte on the right side was still visible on the 3D scan and was subsequently resected. Hemostasis was meticulously confirmed, and the pharyngeal structures of the swallowing mechanism were visualized to ensure no direct injury. A deep surgical drain was inserted, and the incision was closed. The duration of surgery was recorded for a total for 1 h and 46 min. The patient was then transferred to the hospital bed, extubated, and in stable condition. Estimated blood loss was recorded as 100 milliliters and no surgical complications were indicated.

4 | PATIENT-REPORTED OUTCOME MEASURES (PROM)

PROM questionnaires included the Stanford Swallowing Disturbance (SSD), Eating Assessment Tool (EAT-10), SSQ, Numeric Rating Scale (NRS), Scoliosis Research Society-22r (SRS-22r), Neck Disability Index (NDI), and Oswestry Disability Index (ODI) (Table 1). These

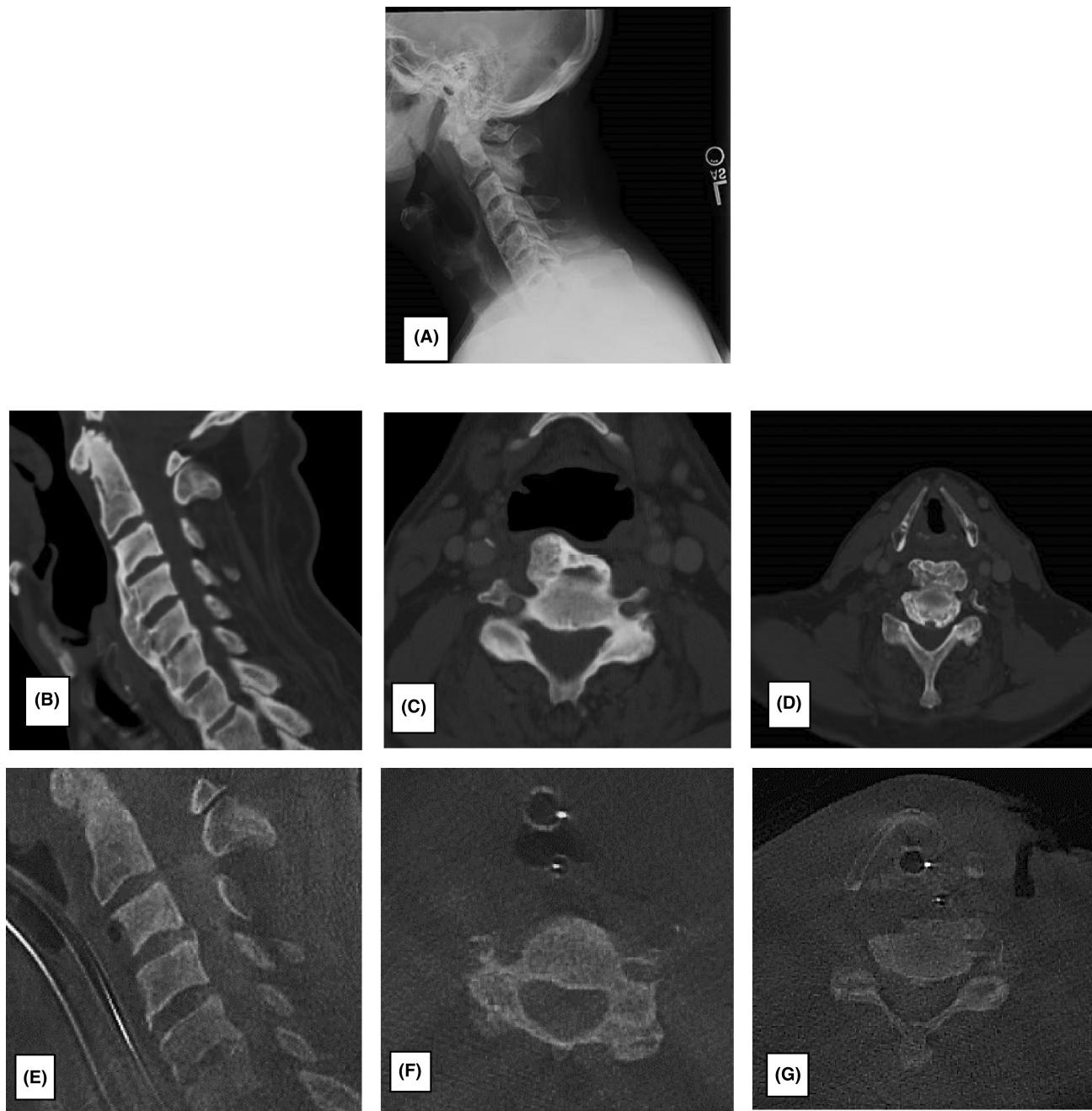


FIGURE 1 (A) Pre-operative lateral X-ray demonstrating large osteophytes formed from C3 to C6. (B) Sagittal: Pre-operative CT scan depicting severe DISH from C3 to C6 obstructing the esophagus. (C) Axial: Pre-operative CT scan depicting hyperostosis at C3–C4. (D) Axial: Pre-operative CT scan depicting hyperostosis at C4–C5. (E) Sagittal: Intraoperative 3D fluoroscopy depicting DISH from C3 to C6 obstructing the esophagus. (F) Axial: Intraoperative 3D fluoroscopy depicting hyperostosis at C3–C4. (G) Axial: Intraoperative 3D fluoroscopy depicting hyperostosis at C4–C5.

questionnaires were employed to evaluate swallowing, neck pain, overall functionality, and related disability.

EAT-10=26 (65%), NRS neck=8/10, NDI=32 (32%), and ODI=48 (48%).

4.1 | Pre-operatively

PROMs were administered pre-operatively and scores were recorded; SSQ=45 (45%), SSD=14 (31%),

4.2 | Post-operatively

PROMs were administered to the patient at follow-up post-operative visits (POVs) of 2 weeks (POV 1), 3 months

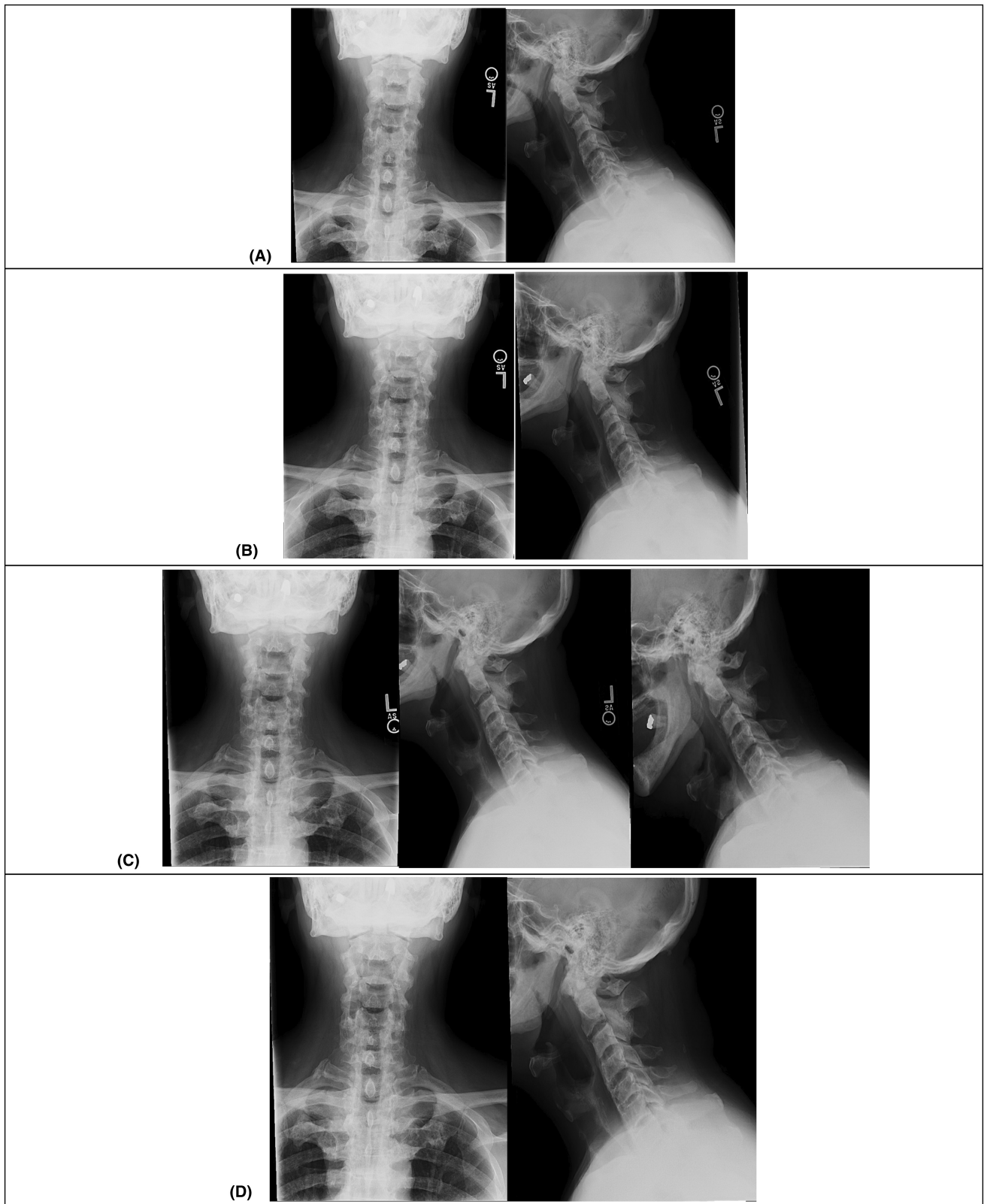


FIGURE 2 Post-operative visit (POV) radiographs. (A) POV 1 (2 weeks) (B) POV2 (3 months) (C) POV 3 (6 months) (D) POV 4 (1 year).

(POV 2), 2 months (POV 3), and 1 year (POV 4) Two weeks post-operatively (POV-1), the patient reported complete resolution of dysphagia and significant reductions in neck pain and disability, with scores of SSQ=0 (0%), SSD=0

(0%), EAT-10=1 (2.5%), NRS neck=0/10, NDI=2 (2%), and ODI=2 (2%) (Table 1). The showed continued improvement, with the ability to consume a greater variety of foods and liquids.



FIGURE 3 (A) Patient placed in a supine position on a Maquet table. (B) Anterior cervical region was prepped and draped in a routine sterile fashion.

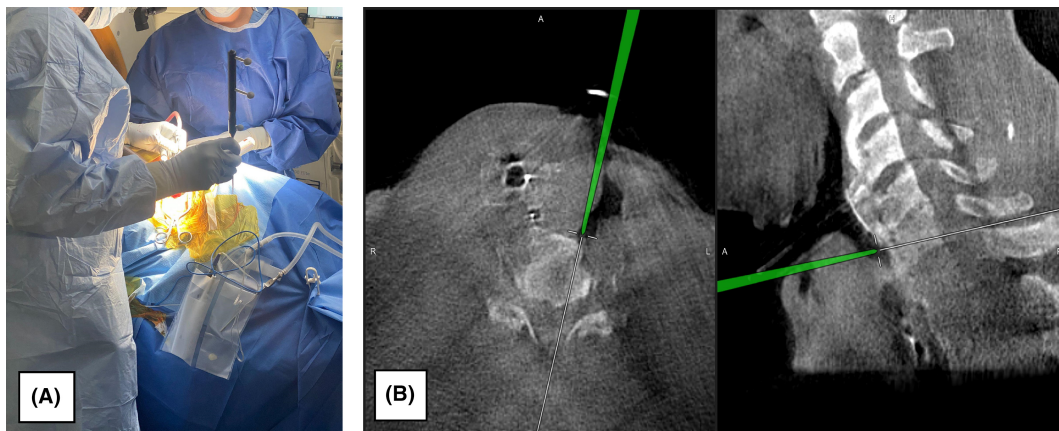


FIGURE 4 (A) CAN navigated probe (green), is a direct “line-of-sight” from the array to the navigational axial view (B) CT image from CAN software in real-time feedback of patient anatomy and trajectory seen from a lateral view on the navigation monitor.

5 | DISCUSSION

DISH predominantly affects middle-aged and older individuals with chronic back pain and spinal stiffness. Its prevalence among those over 50 years old is reported up to 25% in various studies.¹⁵ Often an incidental finding on imaging, DISH diagnosis relies on specific clinical criteria.¹⁶ In the cervical spine, DISH-related osteophytes can compress pharyngeal and esophageal structures, leading to dysphagia for which clear surgical guidelines remain limited.^{1–9} Moreover, individuals with DISH are at an increased risk of fractures through the disc space or vertebral body.¹⁷ This case study supports anterior cervical

osteotomy with CAN as a potential treatment approach for DISH-related dysphagia.

The complex and unique anatomy of DISH patients may necessitate the utilization of CAN during surgical intervention. Investigations have demonstrated high efficacy of CAN technology in complex anatomical surgeries such as thoracic pedicle screw insertion for adult spinal deformity surgery and cervical decompression in a patient with Klippel-Feil syndrome.^{18,19} Recent research has showcased significant advancements in CAN technology, leading to improved accuracy during intraoperative scanning and reduced registration time.^{18,20} These developments are driven by modeling software that

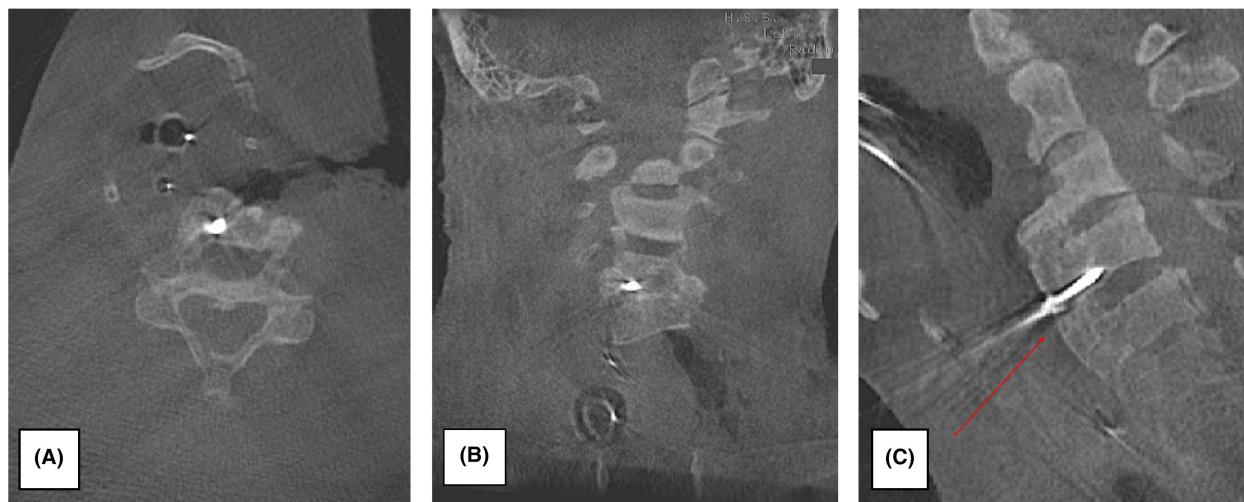


FIGURE 5 Intraoperative 3D fluoroscopic scans were obtained to confirm the bony resection. (A) axial, (B) coronal, (C) sagittal. A small bony region proximally on the right side of sagittal view was still visualized on the 3D scan which was subsequently resected (depicted by red arrow).

generates detailed anatomical maps, and tracking software that localizes instruments in real time across multiple displays.^{18,21} Consequently, stereotactic navigation has emerged as a valuable supplement for spine surgery, notably improving instrumentation accuracy in cases with atypical anatomy.¹⁸

Accurate instrumentation of the cervical spine is critical due to its proximity to vital structures such as the vertebral artery, nerve root, and spinal cord.^{22,23} CAN technology can aid in achieving reproducible anatomical precision by reducing error and enhancing the surgeon's skill.^{18,22} However, despite several CAN technologies being granted clearance by the US Food and Drug Administration for use in spinal surgery, their application in cervical spine surgery remains in its nascent stages compared to the thoracic and lumbar spine.^{23,24} Efforts are being made to overcome software limitations, allowing CAN to be used not only for bony anatomy in instrumented cases but also for decompression portions of procedures.²² Minor innovative challenges such as adapting commonly used surgical instruments to accommodate navigable optical arrays need to be addressed.^{21,22} Comprehensive surgical systems with the ability to navigate decompressive procedures and overlay augmented reality onto the operative field are on the horizon,²² and have the potential to aid in complex anatomical cases such as DISH.

In this case of DISH-related dysphagia, the patient presented with significant morbidity, including impaired swallowing, compromised neck functionality, and significant pain. Radiographs of the cervical spine demonstrated compression of esophageal and pharyngeal structures due to vertebral osteophytes. Despite a previous visit to another institution with similar symptoms a month prior,

conservative treatment provided no relief. Conservative treatment for DISH commonly involves dietary restrictions, physical and swallowing therapy, and medications such as corticosteroids, antireflux agents, and muscle relaxants. No conservative treatment has proven to be effective, as surgery is employed in 66% of symptomatic DISH cases.²⁵ Consequently, given his complex anatomy, the patient was recommended for curative osteotomy with CAN assistance.

Post-operative improvement was remarkable, with the patient experiencing immediate relief from his symptoms during his recovery in the post-anesthesia care unit. Subsequently, PROMs were collected at 2 weeks, 3 months, 6 months, and 1 year post-operatively, revealing statistically significant improvement compared to pre-operative measurements. The patient expressed satisfaction following the first post-operative visit, and at the final follow-up, he remained asymptomatic and expressed gratitude for the improvement in his quality of life. A systematic review by Harlianto et al.²⁵ demonstrated a post-operative complication rate of 22.4% following surgery for DISH in the cervical spine, with recurrent dysphagia as the most common complication. Therefore, the patient was closely monitored following the surgery to ensure no symptoms recurred. Achieving such a result may have proven challenging without the added accuracy provided by CAN during this complex surgical procedure.

6 | CONCLUSION

This case provides the first evidence in support of the utilization of CAN for addressing DISH-related dysphagia, underscoring its potential benefits in cases of complex

TABLE 1 Pre-operative and post-operative PROM values were compared to assess the patient's surgical outcomes.

PROMs	Pre-operative	POV 1 (2 weeks) (N, %)	POV 2 (3 months) (N, %)	POV 3 (6 months) (N, %)	POV 4 (1 year) (N, %)
Stanford Swallowing Disturbance (SSD)	14 (31%)	0 (0.0%)*	1 (7.1%)**	0 (0.0%)**	0 (0.0%)**
Sydney Swallow Questionnaire (SSQ)	45 (45%)	0 (0.0%)*	0 (0.0%)*	0 (0.0%)*	0 (0.0%)*
Eating Assessment Tool (EAT-10)	26 (65%)	1 (2.5%)**	1 (2.5%)**	2 (5.0%)**	0 (0.0%)**
Numeric Rating Scale (NRS)	24/30 (80%)	7/30 (23%)**	2/30 (6.7%)**	10/30 (33%)**	0/30 (0.0%)**
Neck	8	0	0	0	0
Back	8	2	1	0	0
Leg	8	5	1	100	0
Scoliosis Research Society-22r (SRS-22r)	2.6 (52%)	4.0 (80%)**	4.0 (80%)**	4.2 (84%)**	4.5 (90%)**
Neck Disability Index (NDI)	32 (32%)	2 (2.0%)**	0 (0.0%)**	0 (0.0%)**	0 (0.0%)**
Oswestry Disability Questionnaire (ODI)	48 (48%)	2 (2.0%)**	0 (0.0%)**	0 (0.0%)**	0 (0.0%)**

Note: * $p < 0.05$, ** $p < 0.001$.

anatomy. Subsequent investigations should further assess the efficacy of CAN in managing surgically complex cases in the cervical spine.

AUTHOR CONTRIBUTIONS

Fedan Avrumova: Conceptualization; data curation; formal analysis; investigation; methodology; project administration; resources; validation; visualization; writing – original draft; writing – review and editing. **Samuel N. Goldman:** Data curation; formal analysis; investigation; validation; writing – original draft; writing – review and editing. **Franziska Altorfer:** Formal analysis; investigation; methodology; validation; visualization; writing – review and editing. **Gregory K. Paschal:** Data curation; investigation; validation; writing – original draft. **Darren R. Lebl:** Conceptualization; formal analysis; investigation; methodology; project administration; resources; supervision; validation; visualization; writing – original draft; writing – review and editing.

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Not applicable.

CONFLICT OF INTEREST STATEMENT

Fedan Avrumova, Samuel N. Goldman, Franziska Altorfer, and Gregory K. Paschal do not have any conflicts of interest to disclose.

Darren R. Lebl reports royalties from Stryker, royalties from NuVasive, ownership interest from Woven Orthopedic Technologies, ownership interest from Viseon, Inc, ownership interest from Vestia Ventures MiRus Investment LLC, ownership interest from ISPH II, LLC, ownership interest from HS2, LLC, ownership interest from Remedy Logic, on the advisory board for Remedy Logic, on the advisory board for Choice Spine, a consultant for Choice Spine, a consultant for Depuy Synthes and a consultant for Viseon, Inc outside the submitted work.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available from the corresponding author upon request.

ETHICS STATEMENT

Ethical approval was obtained from the Institutional Review Board at the Hospital for Special Surgery. The study was conducted in accordance with the Declaration of Helsinki. Approval was granted by the Ethics Committee at the Hospital for Special Surgery.

CONSENT

Written informed consent was obtained from the patient to publish this report in accordance with the journal's patient consent policy.

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REFERENCES

- Mader R, Verlaan JJ, Buskila D. Diffuse idiopathic skeletal hyperostosis: clinical features and pathogenic mechanisms. *Nat Rev Rheumatol*. 2013;9:741-750. doi:10.1038/nrrheum.2013.165
- 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. doi:10.1016/j.wneu.2019.08.124
- Jeyaraman M, Ramesh R, Prajwal S. Diffuse idiopathic skeletal hyperostosis (DISH)—a case report. *IP Int J Orthop Rheumatol*. 2018;4(2):76-79.
- Kim SK, Choi BR, Kim CG, et al. The prevalence of diffuse idiopathic skeletal hyperostosis in Korea. *J Rheumatol*. 2004;31(10):2032-2035.
- Choi HY, Jo DJ. Surgical treatment of dysphagia secondary to anterior cervical osteophytes due to diffuse idiopathic skeletal hyperostosis. *Medicina*. 2022;58(7):928. doi:10.3390/medicina58070928
- Aydin E, Akdogan V, Akkuzu B, Kirbas I, Ozgirgin ON. Six cases of Forestier syndrome, a rare cause of dysphagia. *Acta Otolaryngol*. 2006;126:775-778. doi:10.1080/00016480500504192
- Lofrese G, Scerrati A, Balsano M, et al. Surgical treatment of diffuse idiopathic skeletal hyperostosis (DISH) involving the cervical spine: technical nuances and outcome of a multicenter experience. *Global Spine J*. 2021;12(8):1751-1760. doi:10.1177/2192568220988272
- Fox TP, Desai MK, Cavenagh T, Mew E. Diffuse idiopathic skeletal hyperostosis: a rare cause of dysphagia and dysphonia. *BMJ Case Rep*. 2013;2013:bcr2013008978.
- Epstein NE. Simultaneous cervical diffuse idiopathic skeletal hyperostosis and ossification of the posterior longitudinal ligament resulting in dysphagia or myelopathy in two geriatric north Americans. *Surg Neurol*. 2000;53(5):427-431.
- Lange N, Meyer B, Meyer HS. Navigation for surgical treatment of disorders of the cervical spine—a systematic review. *J Orthop Surg (Hong Kong)*. 2021;29(1_suppl):23094990211012865. doi:10.1177/23094990211012865
- Verma R, Krishan S, Haendlmayer K, Mohsen A. Functional outcome of computer-assisted spinal pedicle screw placement: a systematic review and meta-analysis of 23 studies including 5,992 pedicle screws. *Eur Spine J*. 2010;19(3):370-375.
- Fichtner J, Hofmann N, Rienmüller A, et al. Revision rate of misplaced pedicle screws of the thoracolumbar spine – comparison of three-dimensional fluoroscopy navigation with freehand placement: a systematic analysis and review of the literature. *World Neurosurg*. 2018;109:e24-e32.
- Shin BJ, James AR, Njoku IU, Härtl R. Pedicle screw navigation: a systematic review and meta-analysis of perforation risk for computer-navigated versus freehand insertion. *J Neurosurg Spine*. 2012;17(2):113-122.
- Nasser R, Drazin D, Nakhla J, et al. Resection of spinal column tumors utilizing image-guided navigation: a multicenter analysis. *Neurosurg Focus*. 2016;41(2):E15.
- Riaz S, Kortbeek FB. Images in spine surgery: diffuse idiopathic skeletal hyperostosis (DISH). *J Pak Med Assoc*. 2007;57(3):157-158.
- Candelario N, Lo KB, Naranjo M. Cervical diffuse idiopathic skeletal hyperostosis (DISH) causing oropharyngeal dysphagia. *BMJ Case Rep*. 2017;bcr2016218630. doi:10.1136/bcr-2016-218630
- Taher AW, Page PS, Greenway GP, et al. Spinal fractures in the setting of diffuse idiopathic skeletal hyperostosis conservatively treated via orthosis: illustrative cases. *J Neurosurg Case Lessons*. 2022;3(20):CASE21689. doi:10.3171/CASE21689
- Wallace N, Schaffer NE, Freedman BA, et al. Computer-assisted navigation in complex cervical spine surgery: tips and tricks. *J Spine Surg*. 2020;6(1):136-144. doi:10.21037/jss.2019.11.13
- Rajasekaran S, Vidyadhara S, Ramesh P, Shetty AP. Randomized clinical study to compare the accuracy of navigated and non-navigated thoracic pedicle screws in deformity correction surgeries. *Spine (Phila Pa 1976)*. 2007;32(2):E56-E64. doi:10.1097/01.brs.0000252094.64857.ab
- Jakubovic R, Guha D, Gupta S, et al. High speed, high density intraoperative 3D optical topographical imaging with efficient registration to MRI and CT for craniocervical surgical navigation. *Sci Rep*. 2018;8(1):14894. doi:10.1038/s41598-018-32424-z
- Mezger U, Jendrewski C, Bartels M. Navigation in surgery. *Langenbecks Arch Surg*. 2013;398(4):501-514. doi:10.1007/s00423-013-1059-4
- Lebl DR, Avrumova F, Abjornson C, Cammisa FP. Cervical spine navigation and enabled robotics: a new frontier in minimally invasive surgery. *HSS J*. 2021;17(3):333-343. doi:10.1177/15563316211026652
- Coric D, Rossi V. Percutaneous posterior cervical pedicle instrumentation (C1 to C7) with navigation guidance: early series of 27 cases. *Global Spine J*. 2022;12(2_suppl):27S-33S. doi:10.1177/21925682211029215
- Kelley BV, Hsiue PP, Upfill-Brown AM, et al. Utilization trends and outcomes of computer-assisted navigation in spine fusion in the United States. *Spine J*. 2021;21(8):1246-1255. doi:10.1016/j.spinee.2021.03.029
- Harlianto NI, Kuperus JS, Mohamed Hoesein FAA, et al. Diffuse idiopathic skeletal hyperostosis of the cervical spine causing dysphagia and airway obstruction: an updated systematic review. *Spine J*. 2022;22(9):1490-1503. doi:10.1016/j.spinee.2022.03.002

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