CASE REPORT

Cervical diffuse idiopathic skeletal hyperostosis (DISH) causing oropharyngeal dysphagia

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SUMMARY

Diffuse idiopathic skeletal hyperostosis (DISH) is a non-inflammatory condition characterised by calcification and ossification of the vertebral ligaments. It is most commonly seen to affect the thoracic and lumbar vertebrae and is usually seen among elderly men. The cause of this condition is unknown. Risk factors include male gender, obesity, diabetes and advancing age. The majority of these cases are found incidentally on imaging and patients are generally asymptomatic. Cervical DISH is less common than its thoracic and lumbar counterparts. When symptomatic, it can cause dysphagia or sometimes airway compromise. If this happens, surgical intervention should be performed. Although a rare cause of dysphagia, DISH is easily diagnosed with imaging. When identified, surgical decompression produces very good clinical outcomes.

BACKGROUND

Swallowing is a complex process whereby chyme is transported from the mouth to the stomach. Dysphagia is a term that pertains to any abnormality that hinders the process of swallowing. Problems in swallowing can lead to complications like respiratory failure or respiration infection from aspiration. Malnutrition due to various nutritional deficiencies can also be a result of chronic dysphagia. Our patient had severe swallowing abnormality leading to acute respiratory failure. Diffuse idiopathic skeletal hyperostosis (DISH) is a disorder characterised by calcification of the entheses regions and spinal ligaments. This case features a rare case of dysphagia caused by cervical DISH. This case highlights a cause of dysphagia that could be reversible with a surgical intervention.

CASE PRESENTATION

A man aged 76 years with a history of chronic alcohol abuse was brought to the hospital for altered mental status likely due to alcohol intoxication. Once awake, the patient was evaluated by the speech and swallow therapist and noted that the patient had dysphagia and was at high risk of aspiration. Patient's hospital course was complicated by hypoxic respiratory failure requiring intubation due to aspiration. Owing to a concern for a cerebrovascular accident, a CT scan and MRI of the brain was performed. Both studies did not show any signs of acute stroke. An electromyogram was likewise performed for the patient which did not show any neuromuscular abnormalities. The patient was eventually extubated and video fluoroscopy was performed which showed that the epiglottis was hitting

the posterior pharyngeal wall. Imaging of the cervical spine showed bridging osteophytes consistent with DISH that is obstructing the upper third of the oesophagus and hypopharynx ([figure 1](#)). This mechanical cause of dysphagia was identified; hence, a surgical consultation was performed. The patient was evaluated by orthopaedic and neurosurgery who did osteophyte resection at C4–C6 using a left-sided anterior approach with cervical microdiscectomy and anterior cervical plate fixation. The patient had a swallowing study performed 2 weeks post-operatively which still showed aspiration of barium ([figure 2](#)). A temporary percutaneous gastrostomy tube was placed for an access for feeding. The patient underwent speech and swallowing rehabilitation. Six months after the surgical intervention, a repeat swallowing study showed normal swallowing study with no signs of aspiration ([figure 3](#)). Removal of the percutaneous gastrostomy tube was planned.

INVESTIGATIONS

- ▶ MRI of the cervical spine
- ▶ CT scan of the brain
- ▶ MRI of the brain
- ▶ Electromyogram

DIFFERENTIAL DIAGNOSIS

- ▶ Acute cerebrovascular accident
- ▶ Neoplastic process

TREATMENT

Surgery: Cervical osteophyte resection with microdiscectomy and anterior cervical plate fixation.

OUTCOME AND FOLLOW-UP

The patient underwent speech and swallowing rehabilitation. He had complete recovery of swallowing function.

DISCUSSION

DISH also known as Forestier disease is a disease that was originally characterised by flowing ossification along the anterolateral aspect of at least four adjacent vertebral bodies.¹ The understanding of the clinical entity has significantly evolved, since its first description by J Forestier and J Rotes-Querol as a form of senile ankylosing hyperostosis of the spine, since the disease is predominantly seen in the older population.² It is now recognised that extraspinal involvement of the disease exists, hence the term DISH. Ossification of the ligaments can happen in the peripheral joints, pelvis, calcaneus, tarsals, olecranon and patella with distinctive radiological findings.³

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Figure 1 MRI of the cervical spine showing multilevel osteophyte complex of the cervical spine showing narrowing of the spinal canal. There is anterior protrusion of the osteophyte complex at the C3 and C4 level that is leading to compression of the posterior wall of the oropharyngeal area.

Figure 2 Fluoroscopic swallowing study performed 2 weeks postcervical osteophyte removal that is showing barium spillage in the vallecular and piriform sinuses. There is impaired normal inversion of the epiglottis and probable aspiration.

DISH is generally an incidental finding on imaging. Most cases have an indolent course that do not entail any intervention and the exact prevalence of DISH is largely unknown due to a wide variation in the clinical definition of the disease. A series of clinical criteria has been formulated to diagnose the disease. The most widely used definition of the disease was described by Resnick and Niwayama which involves the following: (1) flowing ossification along the anterolateral aspect of at least four contiguous vertebral bodies; (2) relative preservation of intervertebral disc height in the involved segment and (3) absence of apophyseal joint bony ankyloses and sacroiliac joint erosions. In a study performed by Weinfeld *et al*, the incidence of DISH among male patients above 50 years old was 25% and 15% in the female population. Among patients older than 65 years, prevalence could be as high as 42%.⁴⁻⁷ Aside from age, male gender, obesity, diabetes mellitus and hypertension have been associated with development of DISH.^{6 8}

Figure 3 Fluoroscopic swallowing study performed 6 months postcervical osteophyte removal that is showing normal oral barium processing and lingual delivery. The epiglottis with normal inversion. There is no noted aspiration in this study.

The axial skeleton is the most commonly affected site of hyperostosis with the thoracic spine being the most common site involved, while the cervical spine is usually minimally affected.^{9 10} The majority of the cases of DISH are asymptomatic but occasionally may present with pain, limited range of spinal movement and increased susceptibility to fractures. Cervical DISH is generally asymptomatic. However, when severe, it can manifest as dysphagia, stridor, dysphonia, aspiration pneumonia and weight loss. Cervical osteophytes can cause swallowing disorders through a variety of mechanisms, including: direct osteophyte compression of the pharynx and oesophagus, disturbances of normal epiglottis tilt, tissue inflammation and oedema around the oesophagus, and pharyngeal muscle spasm.^{4 11} Our case had diffuse levels of hyperostosis, particularly in the C3–C6 which led to dysphagia and aspiration.

The mainstay of treatment for cervical DISH is surgical removal of the osteophytes. Small single-centre cohort studies report good symptom relief and short-term functional outcomes with cervical osteophyte resection, especially in the first 5 years.¹²⁻¹⁴ However, there is some evidence concerning recurrence of osteophytes and dysphagia symptoms during the long-term follow-up period of 10 years or more.¹³ Another important aspect of recovery among patients is speech and swallowing rehabilitation postsurgical removal of osteophytes. Recovery is generally expected in 2 weeks postsurgery. Delayed recovery of swallowing function has also been described which could take months after the operation.^{12 13} Our patient had osteophyte resection from C4 to C6 with C3 and C4 microdiscectomy and cervical plate fixation. He was able to have complete recovery of swallowing function 6 months post-operation after undergoing extensive speech and swallow rehabilitation. Although a rare cause of dysphagia, DISH should be considered especially in elderly male patients with risk factors such as diabetes and obesity. DISH is easily diagnosed with imaging. Surgical decompression produces very good symptom relief and functional outcomes; however, these patients need long-term follow-up surveillance for possible recurrence.

Learning points

- ▶ Cervical diffuse idiopathic skeletal hyperostosis (DISH) is a rare cause of dysphagia.
- ▶ Surgery with osteophyte resection and discectomy is the treatment of choice for cervical DISH causing dysphagia. Patients generally have a good outcome but will need long-term follow-up.
- ▶ When presented with a case of dysphagia, neurological and anatomic causes should be considered in the differential. Anatomic causes like DISH is generally reversible.
- ▶ Given the increasing incidence of obesity and diabetes, DISH could be seen more often; hence, clinicians should be aware of the manifestation and complication of this condition.

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Competing interests None declared.

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REFERENCES

- 1 Mizuno J, Nakagawa H, Isobe M. [Dysphagia caused by ossification of the anterior longitudinal ligament associated with diffuse idiopathic skeletal hyperostosis: report of 2 cases]. *No Shinkei Geka* 1998;26:67–72.
- 2 Forestier J, Rotes-Querol J. Senile ankylosing hyperostosis of the spine. *Ann Rheum Dis* 1950;9:321–30.
- 3 Resnick D, Shaul SR, Robins JM. Diffuse idiopathic skeletal hyperostosis (DISH): Forestier's disease with extraspinal manifestations. *Radiology* 1975;115:513–24.
- 4 Yunoki M, Suzuki K, Uneda A, et al. The importance of recognizing diffuse idiopathic skeletal hyperostosis for neurosurgeons: a review. *Neurol Med Chir (Tokyo)* 2016;56:510–15.
- 5 Resnick D, Niwayama G. Radiographic and pathologic features of spinal involvement in diffuse idiopathic skeletal hyperostosis (DISH). *Radiology* 1976;119:559–68.
- 6 Holton KF, Denard PJ, Yoo JU, et al. Osteoporotic Fractures in Men (MrOS) Study Group. Diffuse idiopathic skeletal hyperostosis and its relation to back pain among older men: the MrOS Study. *Semin Arthritis Rheum* 2011;41:131–8.
- 7 Weinfeld RM, Olson PN, Maki DD, et al. The prevalence of diffuse idiopathic skeletal hyperostosis (DISH) in two large American Midwest metropolitan hospital populations. *Skeletal Radiol* 1997;26:222–5.
- 8 Zicarelli C, Iervolino S, Di Minno MN, et al. Diffuse idiopathic skeletal hyperostosis prevalence in subjects with severe atherosclerotic cardiovascular diseases. *Arthritis Care Res (Hoboken)* 2012;64:1765–9.
- 9 Utsinger PD. Diffuse idiopathic skeletal hyperostosis. *Clin Rheum Dis* 1985;11:325–51.
- 10 Cammisa M, De Serio A, Guglielmi G. Diffuse idiopathic skeletal hyperostosis. *Eur J Radiol* 1998;27(Suppl 1):S7–11.
- 11 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):379–84.
- 12 Urrutia J, Bono CM. Long-term results of surgical treatment of dysphagia secondary to cervical diffuse idiopathic skeletal hyperostosis. *Spine J* 2009;9:e13–17.
- 13 Miyamoto K, Sugiyama S, Hosoe H, et al. Postsurgical recurrence of osteophytes causing dysphagia in patients with diffuse idiopathic skeletal hyperostosis. *Eur Spine J* 2009;18:1652–8.
- 14 Presutti L, Alicandri-Ciufelli M, Piccinini A, et al. Forestier disease: single-center surgical experience and brief literature review. *Ann Otol Rhinol Laryngol* 2010;119:602–8.

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