



Forestier's syndrome : a rare cause of dysphagia. A case report and review of the literature

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We report a 72-year-old male with Forestier's syndrome suffering of dysphagia due to an anterior cervical calcification, unusually great in both volume and extent. Its resection by anterior approach allowed the immediate restoration of a normal swallowing. A bony resection is sufficient in case of Forestier's syndrome, but it must be associated with fixation in case of degenerative osteophyte with disc instability. Long-term follow-up is necessary because the recurrence of the calcification is slow but frequent.

Keywords : diffuse idiopathic skeletal hyperostosis (DISH) ; Forestier's syndrome ; dysphagia ; cervical osteophyte ; osteophyte resection.

INTRODUCTION

Diffuse idiopathic skeletal hyperostosis (DISH) or Forestier's disease is characterized by the ossification of the para-vertebral ligaments and muscles joining the ventral aspect of the vertebral bodies. It involves at least four contiguous vertebral bodies with preservation of inter-vertebral disc height. It is most often an accidental discovery on standard radiographs. Sometimes it is manifested by cervical pain or postural spinal stiffness but is not a surgical indication. We report here a case of DISH affecting of an especially large anterior cervical ossification inducing dysphagia and so requiring surgery. The involvement extended from

C2 to C7 was rarely reported so extensive and so voluminous.

CASE REPORT

A 72-year-old male was referred by an "ear, nose and throat (ENT) specialist" with complaints of difficulty in swallowing associated with cervical stiffness. Previously, the patient managed to swallow by chewing for a long time. Gradually the dysphagia became more pronounced and affected even the liquids. Endoscopic examination revealed a bulge of the posterior pharyngeal wall. A cervical X-ray showed voluminous pre-vertebral flowing ossification extending from C2 to C7. (Figure 1A) This hypertrophic bone reaches an antero-posterior thickness of 20 mm. A cervical CT scan confirmed the bony origin of the bulging mucosa at the level of the oropharynx. (Figure 2) Esophagography with barium revealed the compression of the esophagus due to the cervical anterior ossification. (Figure 3 A.)

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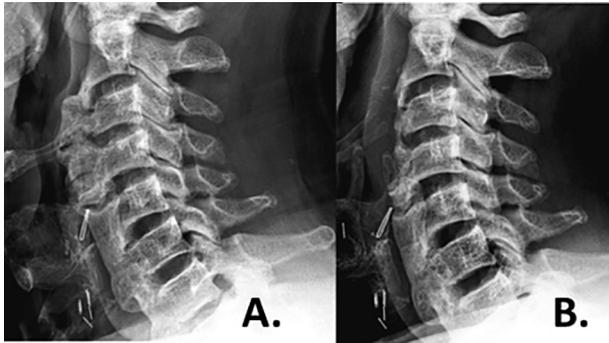


Figure 1. — Lateral X-rays of the cervical spine.
A. Pre-operative view with extended anterior ossifications.
B. Post-operative view after resection.

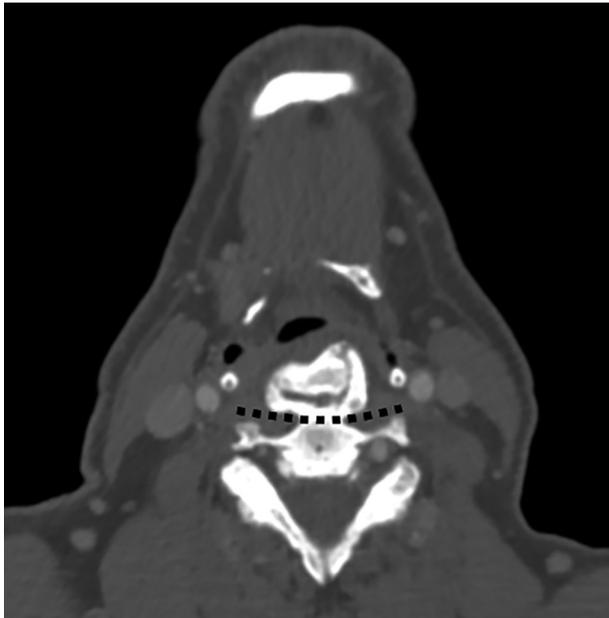


Figure 2. — Cervical CT scan : prominent osteophyte delimited by the dotted line inducing an esophageal compression.

Medical history included chronic global back pain without neurological involvement. However, the patient reported progressive cervical stiffness. Indeed, the clinical examination showed a limitation of cervical mobility at 60° of rotation to the left and 45° to the right, as well as a contracture on the right of the cervical para-vertebral musculature from C2 to C4.

There was also a dyslipemia and a hypo-parathyroidism treated by calcium and calcitriol. Other laboratory tests including HLA-B27 were within normal limits.

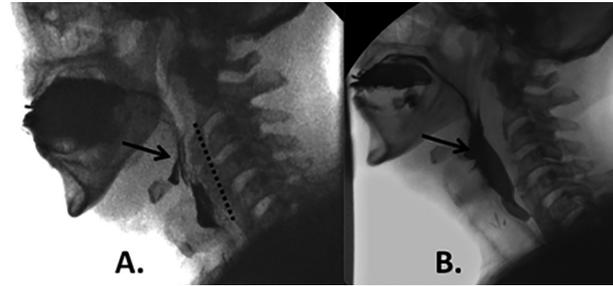


Figure 3. — Barium esophago-graphy.
A. pre-operative : narrowing of the esophagus compressed by the ossification (delimited by the dotted line).
B. postoperative re-expansion of the esophagus allowing normal swallowing (arrow).

The intensity of the dysphagia led to perform a surgical release. Cervical osteophyte resection was performed through an antero-lateral approach using a high-speed tour between C2 and C5. Because of the preservation of the disc heights, no fusion or instrumentation was performed. No preoperative or postoperative complication was encountered.

Disappearance of dysphagia for liquids and even solids was reported by the patient the day after the surgery and persists. The radiography confirmed the quality of bone resection (Figure 1 B) The post-operative esophago-graphy objectified the restoration of normal swallowing. (Figure 3 B)

DISCUSSION

Cervical area is less affected by DISH compared to thoracic and lumbar vertebral segments (4). In these cases, the most commonly affected levels are from C4 to C7, the relatively more mobile regions of the cervical vertebra (3,4,5,6,8). In lateral X-rays, hyperostosis is observed only on the anterior surface of the vertebra in the beginning of the disease, while osteophytes are firmly adherent to the cortex in the latter stages of the disease.

Dysphagia was reported in 0.6 to 1 % of DISH cases (2,3) although some consider it more common, up to 28% (9). It appears typically as a result of esophageal compression in the lower cervical segments, mostly at C4 and C5 level (4). In our case, the extensive ossification of the anterior longitudinal ligament was starting from C2 extending to anterior C7 vertebra and encroaching the esophagus.

Dysphagia secondary to DISH affects mature males, often more than 60-year old. Its frequency increases after the 5th decade and may be associated with endocrines and metabolic syndromes (4). The dysphagia results from several mechanisms: direct compression of the pharynx or esophagus, inflammatory reaction in para-esophageal tissues, disorder of the epiglottis at the laryngeal inlet or crico-pharyngeal spasm (4). Barium swallowing study is a useful method in determining the degree and the localization of the esophageal compression (7,9,11,12). Conservative therapies (like diet modifications and swallowing therapy, non steroidal anti-inflammatory drugs, steroids, muscle relaxants and anti-reflux drugs) can be used before proposing surgery. So, early diagnosis is important for conservative therapy response (4,10,12).

They are few reported cases of dysphagia due to DISH. Most of them involved only 1 or 2 cervical levels, always in the upper part of the cervical spine (C3/C4, C4/C5, exceptionally C5/C6) (4,7,8,11). Indeed, in the lower part of the cervical spine, the vertebrae are more posterior and no longer in contact with the esophagus. The mean thickness of the ossification was reported to be 15.9 millimeters (6). The present reported case is exceptional because involving 6 levels, with a bony thickness of 20 mm, although esophageal compression was located mostly in the upper levels (C3 to C5). This was defined by the oesophago-graphy (figure 3A), allowing removing the bone at these levels only to improve the swallowing.

Most of such dysphagia cases were treated conservatively in the initial stages, and later by excision of the osteophytes through a classical antero-lateral cervical approach, which is considered safe and effective (4,6,7,8,10). However, it may lead to some complications such as recurrent laryngeal nerve injury, Horner syndrome, fistula formation and instability. Only one case was reported treated through a trans-pharyngeal approach, which the most important complications are the infections occurring as a result of the contamination by the oro-pharyngeal flora (7,10). It is also to highlight that dysphagia is a common complication of anterior surgery of the cervical spine for degenerative or traumatic indications, even for the classical antero-

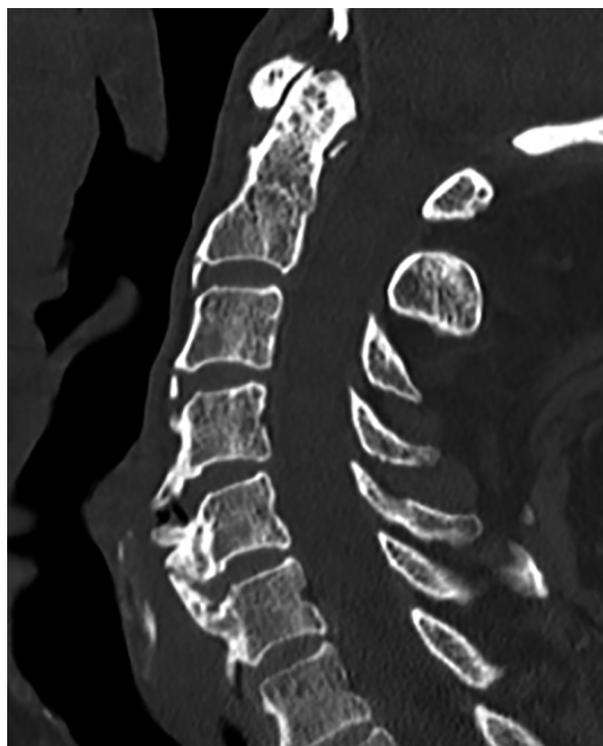


Figure 4. — CT scan of a 71-year-old man whose dysphagia was induced by a C4/C5 C5/C6 degenerative osteophyte whose instability was evidenced by a vacuum image.

lateral approach (3). The incidence of post-operative dysphagia may be as high as 71% within the first two weeks after surgery, but gradually decreases during the following months. However, 12% to 14% of patients may have some persistent dysphagia one year after the procedure (1). It is of multifactorial origin as esophageal retraction, prominent cervical plate or screw, pre-vertebral swelling (1,10). It is proposed to perform pre-operative tracheal exercises and using per-operatively retropharyngeal steroids (1,3).

Surgical results for ossification resection for dysphagia in DISH reported in the literature were most of the time favorable. Only one hematoma requiring drainage was described but no laryngeal nerve palsy nor wound infection (4,6,8,11,12). Most of the time, normal swallowing was restored in the post-operative month (4,6,8). The long term prognosis varies in the literature. Most authors didn't report recurrence of dysphagia at a follow up of 2 to 13 years (4,6). Conversely, Hirano et al. reported a 65%

recurrence rate after a mean postoperative follow-up period of 4.5 years (2). Similarly, Miyamoto reported seven patients with a mean postoperative follow-up period of 9 years and detected 100% recurrence of osteophytes, with a rate of approximately 1 mm/year, especially if disc mobility persists (7). But these recurrent ossifications were symptomatic only in 2 cases, one of them requiring re-intervention after 11 years with favorable result (7).

We didn't perform fusion in our case because of the conservation of the disc height in the upper levels. Effectively, it is important to distinguish DISH hyperosteoic structures from classical osteo-arthritic osteophytes since each has a different surgical treatment. (Figure 4) Removal of hyperosteoic structures is sufficient in surgical treatment of DISH. Nevertheless, additional stabilization of vertebrae is required when the compression originates from an osteophyte induced by degenerative disc instability (1,4,12).

CONCLUSION

Forestier syndrome presenting with dysphagia is very rare. After thorough evaluation of rule of other intrinsic or extrinsic causes of swallowing difficulty and after failure of the conservative management, surgery may be considered and offer good clinical and radiographic outcomes.

Despite the possibility of recurrence in long-term follow-up, surgical resection of osteophytes via a classical anterior cervical approach is reliable and effective in DISH. A follow-up of more than 10 years is essential to detect a possible recurrence, which is rarely symptomatic. Fusion is not indicated in case of Forestier's disease.

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