



## Case Report

## Upper Dysphagia Revealing Diffuse Idiopathic Skeletal Hyperostosis (Forestier's Disease): A Case Report and Review of the Literature

### *Dysphagie Haute Révélant une Hyperostose Squelettique Idiopathique Diffuse (Maladie de Forestier) : À Propos d'un Cas et Revue de la Littérature*

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#### ABSTRACT

Diffuse idiopathic skeletal hyperostosis (DISH) or Forestier's disease is a benign condition characterized by ossification of the ligaments which mainly involves the anterior longitudinal ligament and less frequently the posterior longitudinal part of the spines. We report a case of Forestier's disease revealed by dysphagia to solids in a 62-year-old patient with no particular pathological history, received in our department for progressively worsening solids dysphagia evolving for approximately more than 10 years, with no other associated signs, with a tendency to become complete for one month. The cervical computed tomography (CT) performed revealed an anterior exostosis at the level of C4-C5, anterior marginal osteophytosis, evoking Forestier's disease. The patient underwent complete excision of the osteophytes located at the level of C4-C5 by way of anterolateral cervicotomy. After a follow-up of 09 months postoperatively, the patient was doing well and the cervical CT performed after 03 months postoperatively showed a complete disappearance of the osteophytes. In conclusion, Forestier's disease, although rare, can present with symptoms such as dysphagia and should be considered in the differential diagnosis of patients with progressive difficulty swallowing. Early diagnosis and appropriate management, such as surgical excision of the osteophytes, can lead to a favorable outcome and improvement in symptoms. Regular follow-up is essential to monitor the progression of the disease and ensure optimal patient care.

#### RÉSUMÉ

La maladie diffuse de l'hyperostose squelettique idiopathique (DISH) ou maladie de Forestier est une affection bénigne caractérisée par l'ossification des ligaments qui touche principalement le ligament longitudinal antérieur et plus rarement la partie postérieure des vertèbres. Nous rapportons un cas de maladie de Forestier révélé par une dysphagie aux solides chez un patient de 62 ans sans antécédents pathologiques particuliers, admis dans notre service pour une dysphagie aux solides progressivement aggravée évoluant depuis plus de 10 ans, sans autres signes associés, tendant à devenir complète depuis un mois. La tomodensitométrie (TDM) cervicale réalisée a révélé une exostose antérieure au niveau de C4-C5, une ostéophytose marginale antérieure, évoquant la maladie de Forestier. Le patient a subi une exérèse complète des ostéophytes situés au niveau de C4-C5 par cervicotomie antérolatérale. Après un suivi de 09 mois postopératoire, le patient se portait bien et la TDM cervicale réalisée après 03 mois postopératoire a montré une disparition complète des ostéophytes. En conclusion, la maladie de Forestier, bien que rare, peut se manifester par des symptômes tels que la dysphagie et doit être envisagée dans le diagnostic différentiel des patients présentant une difficulté progressive à avaler. Un diagnostic précoce et une prise en charge appropriée, telle que l'exérèse chirurgicale des ostéophytes, peuvent entraîner un pronostic favorable et une amélioration des symptômes. Un suivi régulier est essentiel pour surveiller l'évolution de la maladie et assurer des soins optimaux au patient.

#### INTRODUCTION

Diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier's disease, is a condition first

described in 1950 by Forestier and Rotes-Querol. It is an ossifying and ankylosing skeletal condition characterized by ossification of the ligaments which mainly concerns the anterior longitudinal ligament and less frequently the

posterior longitudinal part [1]. It is a pathology that not only affects the spine but can be present in all the peripheral skeletons of the human body. Its physiopathology and etiology are not yet well elucidated, hence its diffuse and idiopathic name [2]. The prevalence of DISH varies between 2.9% and 40% in the general population (3% of men at 40 years old, 18.8% of men at 60 years old, and 32.1% of men at 80 years old) but it depends on the criteria diagnostics used and the presence of risk factors. It frequently affects males [1,3]. It is a pathology that is asymptomatic in the majority of cases and it is incidentally discovered on standard radiography of the spine. However, when the involvement is at the level of the cervical spine, the manifestations are of an ENT nature, in particular dysphagia, dysphonia, and dyspnea. More rarely, it manifests late with isolated spinal pain, and spinal stiffness [4]. The involvement is classic at the level of the thoracic or lumbar spine, rarer at the cervical level. Cervical involvement is estimated at approximately 20 to 30% of the general population but most often remain asymptomatic and approximately more than 75% of people aged 65 and over undergo a modification of the spine with the presence of osteophytes. These pre-vertebral osteophytes frequently extend from C2 to C7 which can be responsible for pharyngeal disorders and laryngeal disorders by compression and repression of the structures. The diagnosis is often confirmed by simple complementary examinations, in particular nasofibroscope, oesophagogastroduodenoscopy fibroscopy (OGD), standard spine radiography as well as CT and MRI. Management is often symptomatic and medical. Surgery is required in case of complications [3, 5,6]. We report a case of Forestier's disease revealed by dysphagia to solids and review the recent literature concerning this pathology which, although rare, could concern more and more patients.

### CASE PRESENTATION

Mr. A.K. is a 62-year-old patient with no particular pathological history, received in our department for progressively worsening solids dysphagia evolving for approximately more than 10 years, with no other associated signs, with a tendency to become complete for one month. The patient reports that dysphagia involves only solid foods and is improved by neck flexion during swallowing. There is no associated dysphonia or dyspnea.

#### • Clinical outcomes:

- The patient was in good general condition. Well-colored integuments and conjunctivae. The general parameters were: temperature: 37.2°C; pulse: 80 bpm; BP: 07/14; SAO2: 99% in ambient air;
- The cervico-facial skin examination was normal.
- Examination of the oral cavity and oropharynx was normal.
- The nasofibroscope had objectified a slight bulge at the level of the posterior wall of the hypopharynx, without mucosal lesion. The larynx was normal in appearance with good bilateral cordoarytenoid mobility.
- Otoscopy and anterior rhinoscopy were normal.
- Examination of lymph node areas was also normal.

- Elsewhere the rest of the patient's clinical examination was featureless.

• **Diagnostic approach:** At the end of the clinical examination, an esophageal pathology was suspected and we completed an esophagogastroduodenal fibroscopy under sedation to explore the esophagus which revealed a tight and complete stricture at the level of Killian's mouth preventing the progression of the fibroscope, with normal-looking mucosa. The cervical computed tomography (CT) performed revealed an anterior exostosis at the level of C4-C5, anterior marginal osteophytosis, evoking Forestier's disease. (**Figure 1,2**)

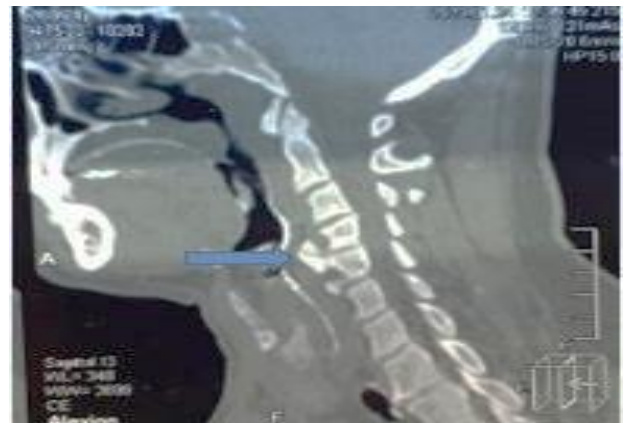


Fig 1. Cervical CT in sagittal section showing ossification of the anterior longitudinal ligament (osteophytes) at the level of C4-C5 (blue arrow).

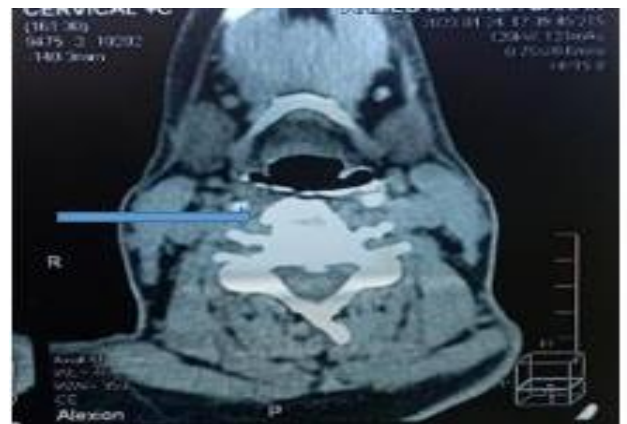


Fig 2. Cervical CT in axial section showing ossification of the anterior longitudinal ligament (osteophytes).

We put the patient under medical treatment initially based on anti-inflammatories, analgesics, and muscle relaxants for three weeks. We also recommended a pasty diet to him, in the absence of the presence of a dietician in our structure.

The patient was seen again three weeks after the start of medical treatment with no improvement in symptoms. After an opinion from an orthopedic surgeon, the indication for surgery is set and the patient is scheduled for surgery.

• **Therapeutic intervention and follow-up:** The diagnosis of dysphagia secondary to Forestier's disease is retained and the patient underwent complete excision of

the osteophytes located at the level of C4-C5 by way of anterolateral cervicotomy. (Figure 3,4). The postoperative course was simple. A semi-liquid diet is resumed normally postoperatively without discomfort. We noted a significant improvement in dysphagia.



Fig 3. Image of surgical approach for left anterolateral cervicotomy



Fig 4. Intraoperative image after complete excision of the osteophyte by left anterolateral cervicotomy

After a follow-up of 09 months postoperatively, the patient was doing well and the cervical CT performed after 03 months postoperatively showed a complete disappearance of the osteophytes (Figure 5).



Fig 5. Postoperative cervical CT scan in sagittal section showing the absence of osteophytes present preoperatively at the level of C4-C5 (blue arrow).

The esophagogastroduodenal transit (TOGD) shows a typical passage of the product through the esophageal lumen (Figure 6).



Fig 6. Image of esophagogastroduodenal transit (TOGD) showing the typical passage of the product through the esophageal lumen postoperatively.

From his first day of consultation in our department and throughout the treatment of his pathology, the patient was delighted with the care he received and was optimistic about the evolution of the condition of his state. The patient was informed of the authors' interest in publishing his clinical case. He voluntarily agreed that we could use his photos for this case report.

## DISCUSSION

Diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier's disease, is a condition first described in 1950 by Forestier and Rotes-Querol. Cervical osteophytes are common in elderly patients, occurring in approximately one-third of individuals over the age of 60. However, the prevalence of diffuse idiopathic skeletal hyperostosis (DISH) varies between 2.9% and 40% in the general population [2,3]. It is a benign pathology most often asymptomatic, but the existence of osteophytes in the cervical spine, in particular, the damage located at the level of C2 to C7, can cause ENT disorders. These clinical manifestations are often in the form of dysphagia, dysphonia, odynophagia, or dyspnoea due to compression and repression of structures [1,3,7]. However, dysphagia is the most frequently reported symptom in the manifestation of DISH of the cervical spine. It is present in up to 28% of cases in Forestier's disease and is directly linked to the existence of cervical osteophytes, particularly those ranging from C4 to C6. This dysphagia is generally more marked for solids than for liquids, improves on flexion of the cervical spine, and worsens on extension. Therefore, the existence of progressive and fluctuating dysphagia which improves on flexion of the cervical spine and worsens extension in an elderly person should alert the otorhinolaryngologist to evoke Forestier's disease among the etiologies of the dysphagia [4,6,8]. However, dysphonia due to recurrent involvement, dyspnea due to tracheal compression or swallowing pneumopathy linked to false passages as well as neurological complications due to spinal or radicular compression linked to ossification of the posterior common vertebral ligament can complete the clinical picture of cervical involvement [7,9, 10]. In our case, the symptomatology was made up of chronic dysphagia to

solids which was improved by flexion of the cervical spine and especially by maneuvers adopted by the patient himself to pass food. The diagnosis must always be confirmed by additional examinations. The latter is based on: a pharyngolaryngeal examination thanks to nasofibroscope which will make it possible to objectify a bulging of the posterior wall of the hypopharynx, will also assess the laryngeal mobility and will also make it possible to eliminate other pharyngolaryngeal causes of dysphagia, including tumors [8]. Simple lateral radiography of the cervical spine alone confirms the diagnosis of Forestier's disease by showing at the cervical level ossification of the anterior and/or posterior longitudinal ligament, bony bridges (spondylophytes). But it also shows a pathognomonic radiological sign in favor of Forestier's disease, which is the presence of a clear line separating the anterior cortex from the ossifying cast [6,11,12]. Although a plain X-ray of the spine is sometimes sufficient to identify diagnostic parameters, computed tomography (CT) and magnetic resonance imaging (MRI) of the spine are also useful, especially in case of neurological complications. They provide additional information, such as the evaluation of the affected soft tissues, and also allow us to appreciate and provide images of the intramedullary involvement. They also make it possible to eliminate any other associated pathology [12,13,14]. The radiological diagnosis of DISH is based on the criteria developed by Resnick and Niwayama in 1976 which are still valid and especially which are gaining importance in the elimination of the differential diagnoses of Forestier's disease. These criteria are [11]:

1. Calcification and ossification along the anterolateral aspect of 4 contiguous vertebral bodies.
2. Relative preservation of the height of the intervertebral discs in the affected areas.
3. Absence of ankylosis of the posterior inter-apophyseal joints and absence of sclerosis of the sacroiliac joints.

The pharynx-oesophageal transit is necessary : it demonstrates the stenosis by extrinsic compression and gives the exact level of the attack. It also eliminates a possible pharynx-oesophageal diverticulum that may be responsible for the dysphagia. The oesogastroduodenal fibroscopy (OGD) also plays a vital role in the search for the etiological diagnosis of any dysphagia to solids. It allows endoscopic exploration of the pharyngeal and esophageal lumen to look for other causes of dysphagia, in particular, tumor causes [1 , 5]. The exact causes of Forestier's disease are still unknown but several associations, including metabolic causes (hyperuricemia, hypervitaminosis A), endocrine causes (type 2 diabetes, dyslipidemia, obesity) as well as traumatic causes have been put forward in several studies. However, only age and male sex seem to have a strong correlation with DISH [15-17]. The exact mechanism of dysphagia caused by DISH is not known but several theories have been proposed such as:

- Growth of osteophytes around the C3 and C4 vertebrae corresponding to the anatomical anchorages of the esophagus which may be responsible for the dysphagia,
- Obstruction of the esophageal lumen by external compression due to osteophytes; this is the case of our patient,
- Local inflammation caused by the friction of large osteophytes on the surrounding tissues causing cricopharyngeal muscle spasm, edema of the esophageal mucosa, and the formation of fibrosis which may be responsible for dysphagia,
- Finally, a restriction of movement of the pharyngolaryngeal junction leads to a decrease in the ascent of the epiglottis, hence swallowing disorders [1, 4,5]. Therapeutically, the management of dysphagia caused by Forestier's disease is multidisciplinary involving physiotherapists, nutritionists, otolaryngologists and orthopedic surgeons, neurosurgeons, and neurologists. The treatment is initially symptomatic, based on the prescription of analgesics, nonsteroidal anti-inflammatory drugs (NSAIDs), sometimes a bolus of corticosteroid therapy, muscle relaxants, and anti-reflux treatment. This is associated with rehabilitation by physiotherapists to reduce stiffness and spinal pain [18]. The involvement of the Dietician is also important in the medical management of dysphagia caused by Forestier's disease. The latter helps to improve caloric intake with the introduction of pasty food and to put in place food hygiene measures when surgical management is not indicated [18]. However, according to Sebaaly *et al.* [1] 80% of dysphagia respond to medical treatment but only temporarily. Surgical treatment is reserved for patients in the event of failure of medical treatment but also in the event of neurological type complications and especially for patients with severe dysphagia or in the event of significant prominent cervical hyperostosis causing false routes that may be responsible for respiratory distress. Three surgical approaches have been described in the literature to gain access to the cervical spine: the transoral approach which addresses extended osteophytes from C1 to C3, the anterolateral cervical approach allowing access to the cervical spine from C2 to C7, passing behind the aerodigestive axis and in front of the vascular axis. It has the advantage of only moderately retracting the carotid axis; however, this access remains limited by the thyroid vessels, the laryngeal nerves and the omohyoid muscle, and finally, the posterolateral cervical route which passes behind the aerodigestive and vascular axes, and therefore gives access to the entire height of the prevertebral space of C3 at C7 [19,20]. According to the systematic literature reviews Choi, H.Y. and Jo, D.J (2022) report that at present, surgery is the choice of management for dysphagia caused by Forestier's disease which gives an excellent result by alleviating symptoms and functional results in the immediate postoperative period. However, this surgery is not devoid of complications, the most common of which is recurrent laryngeal nerve damage. The other much rarer complications are the type of esophageal fistula by

perforation, injury of the great vessels, damage to the mixed nerves, and vertebral osteitis when the cervical spine is approached by the anterolateral cervical approach. However, after an average follow-up of 3 to 70.3 months, no patient had a postoperative recurrence, but this period is short and these patients need follow-up monitoring for a possible long-term recurrence [19,20; 21]. In our case, the patient presented with severe dysphagia resistant to symptomatic medical treatment. He underwent surgical excision of the cervical osteophytes with a very good improvement in symptoms with the expected resumption of food immediately postoperatively.

## CONCLUSION

Diffuse idiopathic skeletal hyperostosis (DISH) or Forestier's disease is a common benign pathology in elderly males and dysphagia is one of the telltale symptoms to consider. Its diagnosis is easy thanks to simple complementary examinations made of standard radiography of the spine, upper digestive endoscopy, CT, and MRI to better study and exclude other causes of dysphagia. Its management is multidisciplinary and primarily medical. Surgery is indicated in complicated cases or cases of medical treatment failure.

## Conflict of interest

All authors declare having no conflict of interest in the publication of this work

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