

Forestier's Disease

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Abstract: Forestier's disease is a musculo-skeletal disorder characterized by diffuse exuberant hyperostosis in anterolateral faces of the vertebral bodies essentially affecting the anterior longitudinal ligament. It generally affects males over 50 years. It is often incidentally discovered on standard spine X rays. Even more rarely, it manifests by neck stiffness, pain or dysphagia. We present a case of a 70 years old patient with difficulty in swallowing solids for several months. Pharyngo-laryngoscopy revealed a bulging of the pharyngeal wall indicating an extrinsic compression. Barium swallow showed paravertebral ossification affecting five consecutive vertebrae from C3 to C7. CT-scan confirmed the osseous proliferation with a split between the vertebral body and the hyperostosis confirming the diagnosis of Forestier's disease. The patient was operated with good post-operative outcomes. In conclusion, Forestier's disease presents with exaggerated hyperostosis can result in complications requiring neurosurgical intervention.

Keywords: Forestier's disease – ligamental ossification – dysphagia

I. Introduction

Forestier's disease or deciduous senile spinal hyperostosis or DISH (Diffuse Idiopathic skeletal hyperostosis) was first described in 1950 by Jacques Forestier (1, 3, 8, 9). It is a non-inflammatory benign disease characterized by a musculoskeletal disorder responsible for exacerbated ossification at sites of ligamentous and tendon insertion (1) with spinal (paravertebral ligaments) and extra spinal (peripheral entheses) manifestations. It is situated mainly on the anterolateral surfaces of the vertebral bodies, affecting essentially the anterior longitudinal vertebral ligament (8). It affects males. Generally asymptomatic, it is incidentally discovered on standard spine x-rays. Rarely, it manifests itself as back pain, spinal stiffness or dysphagia (3, 4, 5, 8, 9). Complications should be suspected in acute symptoms. The most severe are cervical myelopathies, vertebral fractures and compressions of other structures (esophagus, larynx and trachea, inferior vena cava).

II. Clinical Case

This is a 70-year-old patient with a past medical history of high blood pressure, who was operated a year ago for prostate adenoma. He presented with a history of progressive dysphagia for four years, at first involving solid foods, then semi-solid and lastly liquids. Clinical examination including examination of the oral cavity and the oropharynx was strictly normal. The simple X-rays of the cervical spine showed a pre-vertebral ossification bridges extending from C2 to C7. The TOGD showed a prevertebral ossification bridges involving five consecutive cervical vertebrae. The pharyngo-laryngeal fibroscopy revealed a bulging of the posterior pharyngeal wall (cervical esophagus) which directed to an extrinsic compression. The cervical spine ct- scan showed an exuberant ossification of the anterior longitudinal ligament producing bone flows (bridges) extending from C2 to C7 with a cleavage plane between the vertebral body and the ossification bridge asserting the ligament involvement with compression effect on the oropharynx especially on the esophageal opening which was slightly deflected without wall thickening. The patient was operated with good post-operative outcome.

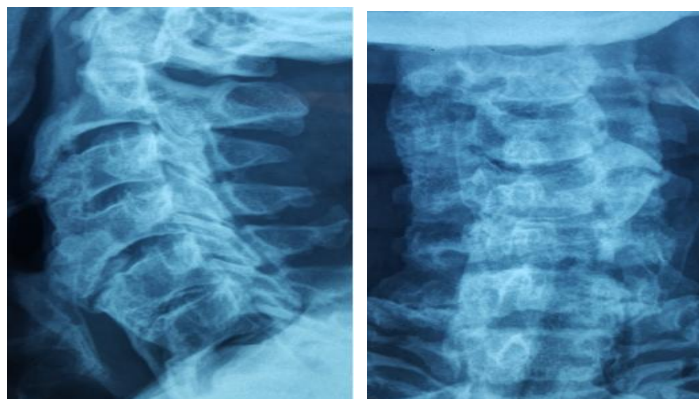


Fig1: Antero-posterior and lateral view cervical spine x-rays showing a prevertebral ossification flow (bridge) extending from C2 to C7

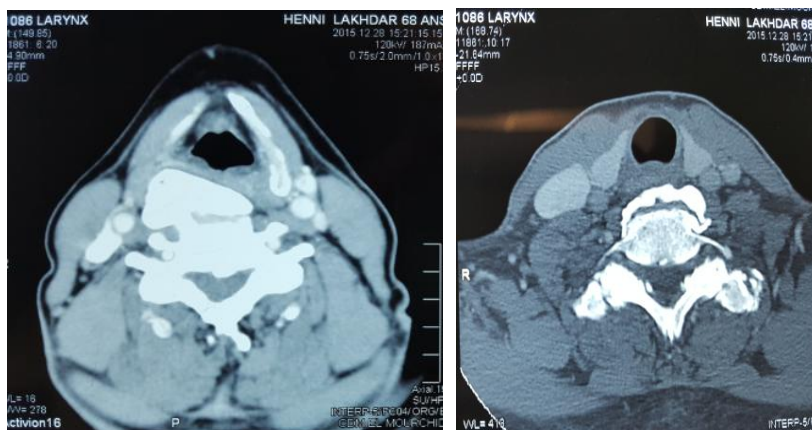


Fig2: Axial and sagittal slices of cervical spine ct-scan showing the ossification of anterior longitudinal ligament

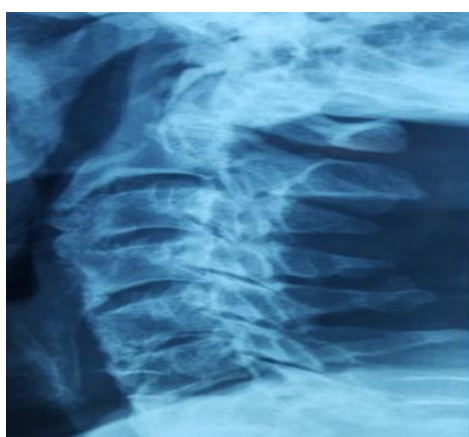


Fig3: Post-operative lateral view cervical spine x-ray showing excision of the prevertebral osseous bridges

III. Discussion

Forestier's disease was first described in the 1950s by Jacques Forestier (1). It all began when he was gathering the observations of ankylosing spondylitis for the writing of a book with Jaume Rotés-Querol. In one of the articles published at a later date, Jacques Forestier said that he remembered two vertebral columns with vertebral ankylosis which he had dissected in the 1940s. They were elderly patients with spinal stiffness, which often involved the dorsal and lumbar spine and in three cases the cervical spine. In particular, the radiological aspect distinguished these patients. The continuous anterior osseous "flow", on the dorsal spine, became discontinuous on the lumbar and cervical spine without altering the height of the disc. Contrary to what is found in the SPA, there was no sacroiliac arthritis. The anatomopathological examination of two vertebral columns showed a cortical bone structure.

In France, Arlet showed that besides asymptomatic forms, Forestier's disease could have severe clinical repercussions. Forestier's disease is considered as a form of degenerative arthritis (1) whose frequency has long been underestimated. It results in bony bridges initially between the dorsal vertebrae, extending progressively to the lumbar and cervical levels. It sometimes affects hip joints and may cause a narrowing of the spinal canal. It affects between 6% and 12% of North Americans, often those over 50 years of age, and about 6% of the world's population. Unlike most other forms of arthritis, it most often affects men (sex ratio of 3/1). However, its aetiological mechanism remains unknown until present. Incriminated risk factors include diabetes, hypertension or hyperuricemia (2). Forestier's disease is manifested by intermittent back pain and spinal stiffness; and the dorsal spine is particularly affected. Digestive disorders can follow the clinical course such as nausea, vomiting or diarrhea but it is mainly dysphagia that becomes the presenting sign of this pathology, reported in 17 to 28% of patients (3), it is of insidious onset, more prominent with solids and accentuated by the extension movements of the neck, progressive development, and may be accompanied by anodynophagia. Dysphonia (hoarse voice) by recurrent laryngeal nerve involvement (4), respiratory distress due to tracheal compression (exceptional) or aspiration pneumonia (due to aspiration) and peristaltic disturbances by osteophytic compression (5) may be encountered. The general condition can be altered with a significant weight loss. The simple spine x-rays are very useful, showing ossification of the anterior and / or posterior longitudinal

ligament, bone bridges (spondylophytes) and a clear line separating the anterior cortex from the ossification flow (pathognomonic sign). CT scans precisely shows the relationship between bone neoproduction and surrounding tissue. The diagnosis of Forestier's disease is based on radiological criteria established by Resnick et al in 1974 (7): calcification and ossification along the anterolateral surface of four consecutive vertebral bodies, anterior predominance of hyperostosis, absence of ankylosis of the interapophyseal joints, relative preservation of the height of the intervertebral discs in the affected areas, absence of erosion or ankylosis of the sacroiliac joints. Naso-fibroscope is used to evaluate the degree of esophageal compression (8). The pharyngo-esophageal barium swallow is necessary because it shows the stenosis due to extrinsic compression and gives the exact level affected (9). The differential diagnosis is mainly with ankylosing spondylitis, psoriatic arthritis and Fiessinger-Leroy Reiter syndrome. The treatment is first symptomatic and medical, combining a semi-liquid diet adapted to the clinical symptoms, rehabilitation to reduce spinal stiffening and analgesics. In case of complications, surgical management is required. Three pathways have been described: the transoral pathway for extensive bone neoproduction from C1 to C3, the anterolateral cervical pathway for approaching the C2 to C7 cervical spine, and finally the posterolateral cervical approach which is behind the aerodigestive and vascular axes and gives access to the entire height of the prevertebral space of C3 to C7 (10). Some publications report excellent results with a follow-up of 30 months to 7 years, others show more than 65% symptomatic recurrences within 4 to 5 years of symptomatic regression despite all surgical treatments.

IV. Conclusion

Forestier's disease is a common condition after the age of 50 years. Its assessment is first by standard x-rays which make it possible to establish the diagnosis, then a ct- scan specifying the level affected. The treatment is first of all medical, surgery is to be considered only in case of persistence or deterioration of the symptomatology, recurrence of the symptoms after successful medical treatment or complications.

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