



FORESTIER'S SYNDROME CAUSING DYSPHAGIA: REPORT OF A RARE CASE AND REVIEW OF THE LITERATURE

Radiology

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ABSTRACT

Forestier syndrome, also known as diffuse idiopathic skeletal hyperostosis, is a clinical condition characterized by paravertebral ligament ossification and diffuse spinal osteophytes. In Forestier's syndrome, mechanical obstruction occurs at the pharyngeal level due to compression of osteophyte and ligamentous ossification, and dysphagia is seen in patients. Symptoms are usually progressive and surgical excision of osteophytes is performed in cases that can not be treated conservatively. In this study, a patient who is presented with dysphagia and diagnosed as Forestier's Syndrome with pharyngeal obstruction due to widespread anterior osteophytes, will be presented with preoperative CT and esophagography images and postoperative CT images.

KEYWORDS:

Dysphagia; Forestier's syndrome

CASE PRESENTATION

A 56-year-old male patient who applied to different centers several times with a complaint of increased swallowing strength during the last one year stated that the tonsils and oropharyngeal levels were normal and the symptomatic treatment was suggested. On June 2016, laryngoscopic examination of the patient who applied to the otorhinolaryngology clinic of our hospital revealed that the posterior oropharyngeal wall was spread forward and the passage was narrowed and the patient was sent to our clinic for barium esophagography. The barium esophagograph revealed that the level of hypopharynx and proximal esophagus narrowed significantly (Fig 1). The patient was evaluated by computed tomography examination to exclude a possible lesion which can make an external compression. CT image shown anterior vertebral osteophytes which has a pronounced posterior pressure on the hypopharyngeal level and replaced the posterior pharyngeal wall to anterior side (Fig 2). The patient's rheumatological blood tests were normal. Patient underwent surgical operation and osteophytes were excised. After the first postoperative month, control laryngoscopic examination of the patient who's swallowing difficulty declining, was performed and the posterior pharyngeal wall was seen in the normal position and the passage width was normal. In the control cervical CT examination of the patient, osteophytes are not observed and hypopharyngeal passage is observed to be normal width (Fig 3 A-B). The patient taken to routine follow-up.



Figure 1: Preoperative esophagography image shows a marked narrowing of the hypopharyngeal and proximal esophageal passage.



Figure 2: Preoperative sagittal CT images show the posterior pressure-induced mechanical obstruction of the osteophytes in the hypopharyngeal and proximal esophageal passage.

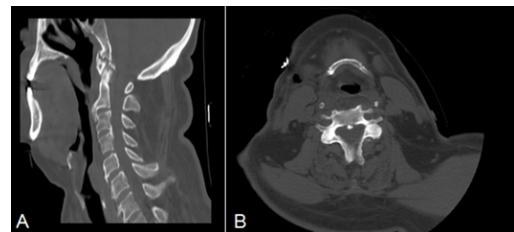


Figure 3 A-B: Post-operative sagittal CT images in Figure 3A and postoperative axial CT images in Figure 3B show no osteophytes and no narrowing in the hypopharyngeal and proximal esophageal passage.

DISCUSSION

Diffuse idiopathic skeletal hyperostosis (DISH), also known as Forestier's syndrome, is characterized by osteophyte formation with the thickening and calcification of tendon and joint capsules (1). Firstly, dysphagia due to cervical osteophyte was described by Mosher in 1926 (2). However, it was first described by Forestier and Rotes-Queral in 1950 as a syndrome (3). In 1975, Resnick et al. described this disease as "Diffuse Idiopathic Skeletal Hyperostosis", and published a case of dysphagia due to diffuse idiopathic skeletal hyperostosis in 1976 (4, 5).

Etiopathogenesis is not known clearly and some theories are suggested. It has been suggested that connective tissue calcifications 5-

10 mm in thickness occur in the vertebrae and ligaments are degenerated with calcification due to the decrease in motion (1). Obesity, hyperlipidemia, hypertension, diabetes, hypervitaminosis A and intake of vitamin A derivatives and osteoblast-stimulating insulin-like growth factor increase are other factors responsible for etiopathogenesis (6, 7).

The most common C4-C7 involvement in the neck region has been shown (8). Dysphagia is the most common symptom in neck involvement, with a frequency of 17-28%. Different mechanisms of dysphagia mentioned such as esophageal protrusion of osteophytes, neuropathy due to recurrent laryngeal nerve injury or esophageal fibrosis and inflammation due to osteophyte irritation. In addition, voice coarseness, stridor, dyspnea and aspiration can also be seen (9).

DISH (Forestier's syndrome) is diagnosed radiologically. Computed tomography (CT) is the gold standard for diagnosis. However, cervical radiographs are useful in demonstrating osteophytes and barium radiographs can demonstrate the passage of pharynx (7, 10).

The diagnostic criterias of diffuse idiopathic skeletal hyperostosis, recommended by Resnick and Niwayama are (5):

1. Paraspinal longitudinal ligament calcification and ossification continuing along the at least four level vertebral body,
2. No disk degeneration, the conservation or slight reduction of disk space,
3. The absence of other signs of degenerative spinal disease.

Common idiopathic skeletal hyperostosis treatment is usually performed conservatively with nonsteroidal anti-inflammatory drugs, steroids, muscle relaxants and anti-reflux therapy, unless there are advanced symptoms (11). Surgery can be planned in cases that can not be treated conservatively (12). Although, in the presence of hyperostotic occurrence in diffuse idiopathic skeletal hyperostosis, it is enough to surgical excision of osteophytes, it is important for different surgery choice to distinguish hyperostosis from osteoarthritic osteophytes because of the need for more complex surgical approaches involving vertebra stabilization in osteoarthrosis (13). Surgical approach may be through the anterior neck, posterior-lateral neck and oral transpharyngeal pathway. A recurrence rate of 65% was reported in the literature (12).

CONCLUSION

Forestier syndrome or diffuse idiopathic skeletal hyperostosis can make an external pressure effect to oropharynx or esophagus and should be kept in mind as a rare cause of dysphagia for the cases without intraluminal pathology.

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