Transpharyngeal approach for the treatment of dysphagia due to Forestier's disease

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Abstract

Forestier's disease (diffuse idiopathic skeletal hyperostosis) is characterized by extensive spinal osteophyte formation and endo-chondral ossification of paravertebral ligaments and muscles. Dysphagia in the setting of Forestier's disease is a rare and hence often unrecognized entity. The dysphagia is due to mechanical obstruction in the initial stages and later due to inflammation and fibrosis. Most of these patients are treated conservatively in the initial stages and later by excision of osteophytes through a lateral cervical approach. We present a case of dysphagia due to cervical osteophytes in the setting of Forestier's disease causing narrowing of the pharynx. The patient was treated surgically via a peroral-transpharyngeal route with excellent results.

Key words: Cervical vertebrae; Deglutition disorders; Spinal osteophytosis; Hyperostosis, diffuse idiopathic skeletal

Case report

A 55-year-old man presented with a two-year history of neck stiffness and two months history of dysphagia for solids. There was no history of weight loss. He was otherwise well and was not on any medication. On examination a mass was seen displacing the posterior pharyngeal wall anteriorly and causing narrowing of the oropharynx and hypopharynx.

Lateral radiographs of the cervical spine showed large cervical osteophytes arising from the anterior aspect of the cervical spine and bridging the vertebral bodies anteriorly from C2 to C7. Pseudo-arthrosis was seen at C2/3 and C4/5 (Figure 1). Radiograph of the lumbar spine also showed changes of diffuse idiopathic skeletal hyperostosis (DISH). A computed tomography (CT) scan of the neck showed a bony bar arising from the anterior aspect of the cervical spine and displacing the posterior pharyngeal wall anteriorly (Figure 2).

The mass was approached through a mid-line vertical incision in the posterior pharyngeal wall (per-oral transpharyngeal route). The tissues and periosteum were elevated and pushed laterally exposing the mass. The mass was partly chiselled and then drilled to decrease the mechanical burden and increase the oropharyngeal lumen. However as suggested previously (Suzuki *et al.*, 1991) no attempt was made to remove the mass completely. The mucosal flaps were approximated and sutured with 4–0 chromic catgut. The patient was fed via a narogastric tube for three days. Prophlylactic amoxycillin and clavulanic acid was given for seven days post-operatively. The patient made an uneventful recovery and showed a remarkable improvement in deglutition. Histologically the lesion contained fibro-cartilage without any evidence of malignancy. Post-operative lateral cervical radiographs showed only marginal improvement in the pharyngeal lumen (Figure 3).
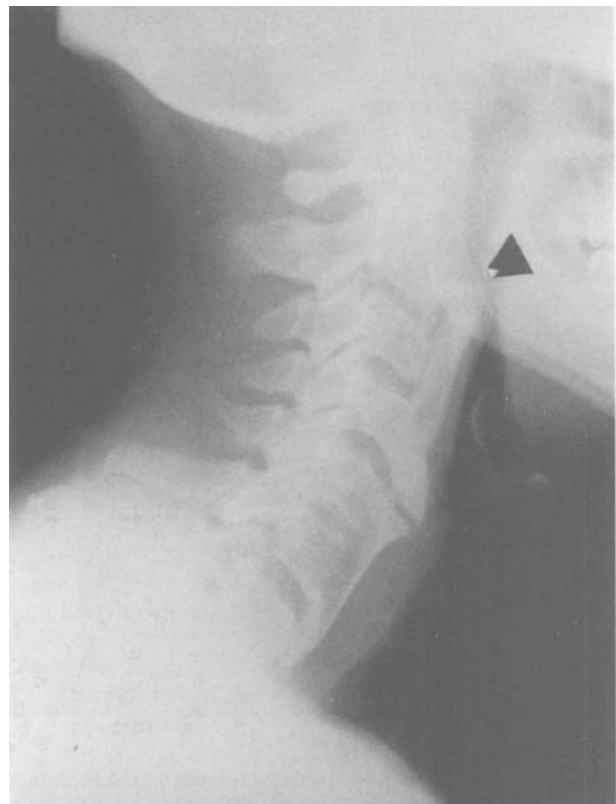

Fig. 1

Lateral cervical radiograph showing a large mass of ossification in the prevertebral space extending from C2 to C7 and showing pseudo-arthrosis at C2/3 and C4/5.

FIG. 2
CT scan showing a bony mass pushing the posterior pharyngeal wall anteriorly.

FIG. 3
Post-operative lateral cervical radiograph. The osteophyte at C2–C3 level has been partially excised leading to a small increase in the oropharyngeal lumen.

Discussion

Diffuse idiopathic skeletal hyperostosis (DISH) is characterized by extensive hyperostotic changes of anterior and posterior aspect of the spine. The condition was originally described by Forestier and Rotes-Querol (Forestier and Rotes-Querol, 1950) and the term DISH was coined by Resnick (Resnick *et al.*, 1975). Resnick established specific criteria for the diagnosis of this condition. These are: (1) flowing calcification and ossification along the antero-lateral aspect of at least four contiguous vertebral bodies with, or without, associated localized pointed excrescences at the intervening vertebral body-disc junctions; (2) relative preservation of intervertebral disc height in affected areas; (3) absence of apophyseal joint ankylosis and sacro-iliac joint erosion, sclerosis or bony fusion (Rusnick and Niwayama, 1976).

Forestier's disease affects men twice more frequently than women. It occurs predominantly in the middle to older age group, the mean age being 65 years (Resnick *et al.*, 1978). No racial predilection has been established (Murray *et al.*, 1990). The condition is characterized by large osteophyte formation, the new bone being formed on the antero-lateral aspect of vertebrae across the front of the vertebral discs. This leads to the formation of a bony ridge immobilizing the involved segment of the spine (Doyle, 1986). There may be extensive endochondral ossification involving the paraspinal ligaments and muscles.

Clinically the condition may be entirely asymptomatic and detected only in routine radiographs (Murray *et al.*, 1990). More commonly varying degrees of spinal pain and stiffness are reported. Dysphagia, dyspnoea, sleep apnoea are rare manifestations. Cervical spine alterations in DISH occur most commonly between the fourth and seventh cervical vertebral bodies.

Clinical studies have shown that 17 to 24 per cent of patients with Forestier's disease manifest symptoms of dysphagia (Resnick *et al.*, 1978). Less than 10 per cent require surgical decompression (McCafferty *et al.*, 1995). Most of the patients are approached through the trans-cervical route. Only one previous account of a per-oral transpharyngeal approach is available in the literature (Saffouri and Ward, 1974). Since dysphagia due to Forestier's disease is uncommon, controversy exists regarding the appropriate treatment. Diet modification and anti-inflammatory medication have been suggested as appropriate treatment in the early stages (Ratnesar, 1970). Dysphagia due to cervical osteophytes is a manifestation of mechanical obstruction in the initial stages. Later fibrosis due to parapharyngitis and paraoesophagitis hamper deglutition (Bauer, 1953). Early excision of osteophytes leads to the resolution of dysphagia due to the elimination of mechanical burden. Later, further resolution of dysphagia follows abatement of inflammation and fibrosis (McCafferty *et al.*, 1995).

Re-accumulation of osteophytes has been reported many years after excision and it is believed that the growth of osteophytes and ligamentous ossification progresses until rigidity is re-established (Suzuki *et al.*, 1991). Hence, when complete resection is not necessary for decompression, the continuity of ossification should be preserved by partial resection. It is for this reason that the prevertebral bony bar was only partially excised in this patient. However, despite the small increase in the oropharyngeal lumen (Figure 3) the patient showed marked improvement in deglutition. This is due to the combined effect of increase in the oropharyngeal lumen (although small) and the resolution of inflammation of the

retropharyngeal tissues. When complete resection is required for sufficient decompression, bone grafting should be added for segmental spondylodesis (Suzuki et al., 1991).

The lateral cervical approach is commonly used and provides wide exposure to lower cervical osteophytes (C5–C7). However, this approach provides limited access to C2–C4 vertebrae. The per-oral transpharyngeal route provides wide access to this region and has the advantage of being simple and direct. Orthopaedic surgeons prefer the lateral cervical route because of the risk of osteomyelitis from contamination by pharyngeal flora. However, the maxilla and mandible are often exposed without such complications. It has previously been suggested that the per-oral transpharyngeal route is a credible alternative to the lateral cervical route without the risk of osteomyelitis (Saffouri and Ward, 1974). Hence, we feel that this approach has an unequivocal place in the treatment of dysphagia arising from high cervical osteophytes in cases of Forestier's disease.

References

Bauer, F. (1953) Dysphagia due to cervical spondylosis. *Journal of Laryngology and Otology* **67:** 615–630.

Doyle, D. (1986) Clinical aspects of osteoarthrosis. In *Coperman's Textbook of Rheumatic Diseases*. 6th Edition. (Scott, J. J., ed.) Churchill Livingstone, Edinburgh, pp 846–873.

Forestier, J., Rotes-Querol, J. (1950) Senile ankylosing hyperostosis of the spine. *Annals of the Rheumatic Diseases* **9:** 321–330.

McCafferty, R. R., Harrison, M. J., Tamas, L. B., Larkins, M. V. (1995) Ossification of the anterior longitudinal ligament and Forestier's disease: an analysis of seven cases. *Journal of Neurosurgery* **83:** 13–17.

Murray, R. O., Jacobson, H. G., Sroker, D. J. (1990) Diffuse idiopathic skeletal hyperostosis. In *The Radiology of Skeletal Disorders*. 3rd Edition, Churchill Livingstone, Edinburgh, pp 715–717.

Ratnesar, P. (1970) Dysphagia due to cervical exostosis. *Laryngoscope* **80:** 469–471.

Resnick, D., Niwayama, G. (1976) Radiographic and pathologic features of spinal involvement in diffuse idiopathic skeletal hyperostosis (DISH). *Radiology* **119:** 559–568.

Resnick, D., Shapiro, R. F., Wiesner, K. B., Niwayama, G., Utsinger, P. D., Shaul, S. R. (1978) Diffuse idiopathic skeletal hyperostosis (DISH) [Ankylosing hyperostosis of Forestier and Rotes-Querol]. *Seminars in Arthritis and Rheumatism* **7:** 153–187.

Resnick, D., Shaul, S. R., Robins, J. M. (1975) Diffuse idiopathic skeletal hyperostosis (DISH): Forestier's disease with extraspinal manifestations. *Radiology* **115:** 513–524.

Saffouri, M. H., Ward, P. H. (1974) Surgical correction of dysphagia due to cervical osteophytes. *Annals of Otology – Rhinology and Laryngology* **83:** 65–70.

Suzuki, K., Ishida, Y., Ohmori, K. (1991) Long term follow up of diffuse idiopathic skeletal hyperostosis in the cervical spine. *Neuroradiology* **33:** 427–431.

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