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Clinical Records

Smooth muscle tumour of the pharynx: a rare tumour presenting with globus pharyngeus symptoms

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Abstract
A rare case of a smooth muscle tumour in the pharynx is reported, together with histopathological findings. The patient's psychiatric background and recurrent complaint of a sensation of a lump in the throat all pointed to a psychogenic aetiology, and diagnosis was delayed. The importance of mirror or endoscopic examination of the pharynx is stressed in patients with globus pharyngeus symptoms.

Key words: Muscle, Smooth; Neoplasms; Pharynx; Globus Pharyngeus

Introduction

Smooth muscle tumours may arise in any structure of the body, most commonly in the uterus, skin, subcutaneous tissues and gastrointestinal tract. However, smooth muscle tumours of the pharynx are rare.[1] An extensive review of the world literature showed only three reported cases of leiomyoma (LM) and three reported cases of leiomyosarcoma (LMS) occurring within the pharynx. A further case is presented in a patient who had a long history of intermittent globus pharyngeus symptoms associated with a psychiatric illness.

Case report

A 47-year-old man presented with a four-month history of a sensation of a lump in the throat, in the midline at the level of the cricoid cartilage. He gave a history of mild to moderate depression over the preceding thirty years, and the depressive episodes had often been associated with a sensation of a lump in the throat and fear of swallowing, lasting from a few days to a few weeks at a time. The throat symptoms were especially noticeable when he felt under stress, and his general practitioner had made a diagnosis of globus pharyngeus, secondary to the psychiatric complaint. The depression had worsened recently and consequently his general practitioner had prescribed fluoxetine.

On this occasion, the sensation of a lump in the throat became slowly progressive in nature, and latterly the patient had developed dysphagia to the extent that in the last few days before presentation he had been unable to swallow anything apart from sips of water. He had no history of pain, hoarseness or swellings in the head and neck area but had lost about 6 kg of weight. He smoked 35 cigarettes per day and drank 42 units of alcohol per week. Video fluoroscopy had already been arranged at the suggestion of the speech and language therapists, who had become involved at an early stage, and was performed with some difficulty due to patient anxiety. The investigation showed easy swallowing of the contrast medium with no obvious anatomical abnormality, supporting the view that the problem was functional.

On assessment by the ENT team, the patient was thin, very anxious and moderately dehydrated. No neurological deficit was found and examination of the ears, nose, mouth, oropharynx and neck was unremarkable. Flexible nasopharyngoscopy, however, revealed a 1.5 cm mass in the hypopharynx. This was situated on the posterior pharyngeal wall slightly to the left of the midline, just above the arytenoids. Direct pharyngoscopy under general anaesthetic confirmed a 1.5 cm, firm mass, just proximal to the left arytenoid on the posterior pharyngeal wall. The mass was covered by an intact mucosal layer. Excision biopsy was performed: the mass was well demarcated from the underlying pharyngeal musculature and was easily removed *in toto*. Macroscopically, the resected mass measured 1.5 cm in diameter and on sectioning showed a uniform tan cut surface. Microscopically, sections showed a spindle cell tumour with discrete margins arranged in whorls and fascicles, containing some pleomorphic large nuclei as well as a few mitotic figures (amounting to 3 per 10 high power fields). On immunohistochemistry, the tumour was strongly positive for smooth muscle actin and desmin but was negative for S100 (marker for neural tumours), CD34 and CD117 (markers for gastrointestinal stromal tumours). The appearances were those of a smooth muscle neoplasm of the pharyngeal wall (Figures 1 and 2). Even though the overall features were suggestive of a benign nature, due to the presence of nuclear pleomorphism and some mitotic activity, samples were sent to a pathologist who specialized in smooth muscle tumours. The consensus opinion was that the lesion was a smooth muscle tumour of uncertain

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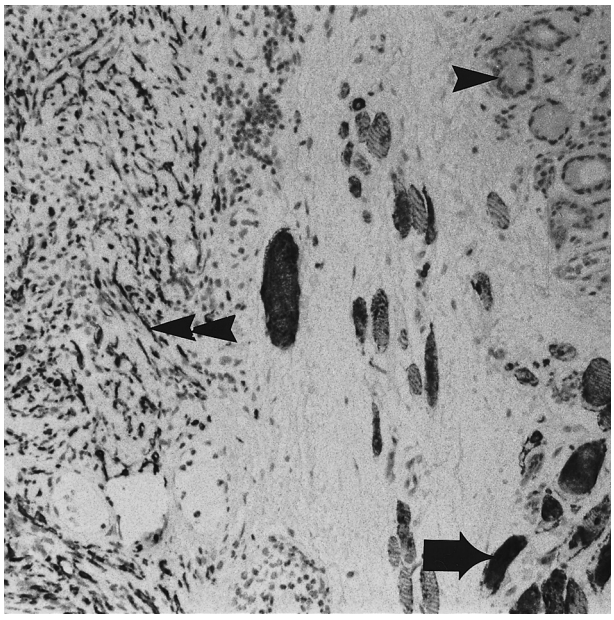

FIG. 1

Immunohistochemistry stain for desmin: spindle-shaped smooth muscle fibres of tumour (double arrowheads) and striated muscle fibres of pharyngeal wall (arrow) stain dark (positive) for desmin, whereas pharyngeal mucous glands (arrowheads) are negative. Magnification × 100.

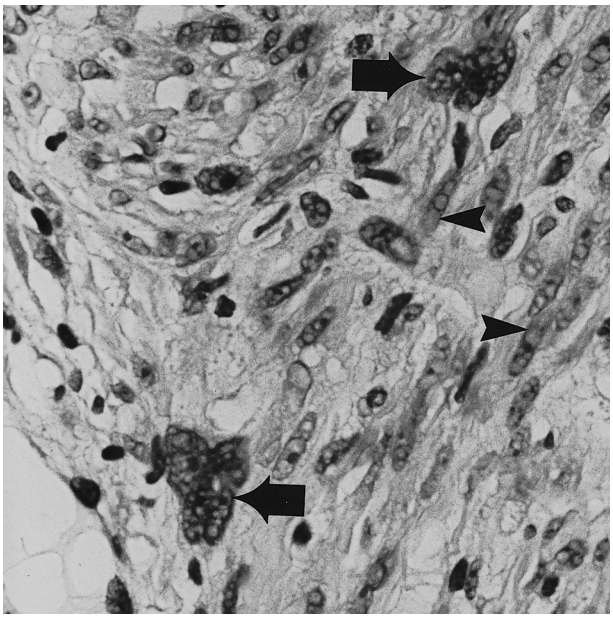

FIG. 2

H&E stain of tumour at high magnification: spindle-shaped smooth muscle fibres with cigar-shaped nuclei (arrowheads) and a few large, atypical cells with large, bizarre nuclei and prominent nucleoli (arrows). Magnification × 400.

malignant potential, but the small size of the tumour and the relatively low mitotic count pointed to benign rather than malignant behaviour. It was therefore decided that further management should comprise close follow-up rather than more extensive surgery or radiotherapy. Two years after the excision the patient remained well with no clinical evidence of recurrence. He had regained his original weight and his symptoms of dysphagia and globus pharyngeus had disappeared.

- **Globus-like symptoms may mask organic pathology**
- **In this case endoscopic examination of a patient with apparent globus revealed a mass in the pharynx which was a smooth muscle tumour**
- **The importance of a full examination is stressed**

Discussion

Most cases of smooth muscle tumour in the pharynx present with difficulty in swallowing.[1] In this case, the patient had a long history of depression associated with recurrent episodes of a sensation of a lump in the throat. A diagnosis of globus pharyngeus had been made previously and it was erroneously assumed that the current symptoms also had a non-organic basis. Radiology proved misleading – the investigation was difficult due to patient anxiety and movement. With hindsight, the examination was inadequate to definitely exclude pathology. However, even in ideal circumstances the hypopharynx is not always easy to visualize on contrast swallow examinations, reflecting the importance of visualizing the pharynx by indirect laryngoscopy, flexible endoscopy and / or rigid endoscopy to avoid missing such tumours. Alerting symptoms in this case were the progressive dysphagia and weight loss, neither of which are typical features of globus pharyngeus.

The occurrence of smooth muscle tumours in the oral cavity and pharynx is rare, probably because of the paucity of smooth muscle tissue in these sites. In 1975, Farman's extensive review of smooth muscle tumours throughout the body showed that only 0.064 per cent had an intra-oral location.[2] Most LMs of the oral cavity arise from the lips, tongue, hard and soft palate, and cheek. Leiomyosarcomas, in contrast, tend to arise around the maxilla and the mandible.[1] In 2000, a review of world literature from 1884 to 1996 showed only two cases of LM and two of LMS which had arisen within the pharynx.[1] A case of angiomyoma of the hypopharynx was reported in 1998.[3] Angiomyoma is a synonym for a vascular variant of LM. A further case of LMS was reported in 2002.[4]

The commonest spindle cell tumours in the pharynx are spindle cell variants of squamous cell carcinoma; immunohistochemical stains have enabled pathologists to make the distinction between spindled carcinoma and other spindle cell tumours more confidently. However, distinguishing between benign and malignant smooth muscle tumours continues to be difficult. Clinical features such as rapid growth, ulceration of the lesion or pain point towards malignancy. Histologically, the overall morphology of the tumour and the degree of cellular pleomorphism are useful indicators of the lesion's behaviour. The number of mitotic figures per high power field is probably the single most important criterion of malignancy.[1] Most investigators agree that if two or fewer mitoses per 10 high power fields are found, the prognosis is good. Greater numbers of mitotic figures are associated with increased malignant potential. It is suggested that in difficult cases the entire specimen must be examined, as the malignant pattern may be present in isolated parts of the tumour.[5] Some authors believe that size is important too: smooth muscle tumours of 2.5 cm or larger are more likely to be malignant.[6]

Because smooth muscle tumours of the pharynx are rare there is little evidence regarding their management. Most have been treated by local excision, taking a wide

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margin of surrounding normal tissue if LMS was suspected. It is generally thought that LMS is relatively radioresistant, but radiotherapy has been used as adjuvant treatment. Chemotherapy has only been used for palliative reasons.

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