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A case of submandibular teratoma

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Abstract
A rare case of benign teratoma arising in the submandibular gland is presented.

Key words: Submandibular, gland neoplasms; Teratoma

Introduction

Teratomas occur in one in 4000 births with a predilection for ovarian, sacrococcygeal, retroperitoneal and anterior mediastinal sites (Hadju *et al.*, 1966; Willis, 1967; Carneg *et al.*, 1972; Hawkins and Park, 1972). Less than five per cent of reported teratomas arise in the head and neck (Berry and Keeling, 1969; Mahour and Woolley, 1974) and these are usually large (over 5 cm), benign (95 per cent) and arise in, or near, the midline (Batsakis *et al.*, 1964; Rose and Howard, 1982). The present case is interesting because the teratoma was small and arose laterally. In our review of the world literature we have only been able to find one other case of submandibular teratoma.

Case report

A three-year-old boy presented with a six-month history of a gradually enlarging left-sided neck swelling (Figure 1). On examination he had a smooth, firm, mobile, 2 cm mass in the region of his submandibular gland which was bimanually palpable. The rest of his examination was unremarkable. Fine-needle aspiration was nondiagnostic and an excision biopsy was performed. At operation a 3.5 × 2.5 × 2 cm hard, encapsulated mass was found in the deep portion of the submandibular gland, traversed by the hypoglossal nerve (Figure 2). Macroscopically the appearance was of a hypoglossal nerve neuroma. The nerve was divided and the lesion excised. The child made an uneventful recovery from surgery and at follow-up six months later was untroubled by his hypoglossal nerve palsy, and had no evidence of recurrence.

Pathology

The lesion comprised a partially circumscribed fibrous nodule (approximately 3.5 × 2.5 × 2 cm) together with salivary gland at one end of the specimen. A large nerve could be recognized arising at one margin of the lesion. Microscopically (Figure 3), the lesion was poorly defined with striated muscle around its margin. The tumour comprised an admixture of tissues (Figure 4) including fat, some of which was immature in appearance, mature

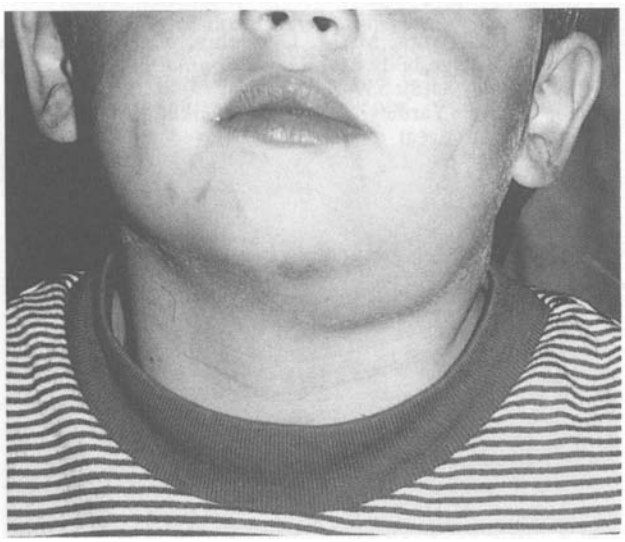

FIG. 1
Photograph of patient at presentation. Note swelling in left submandibular region.

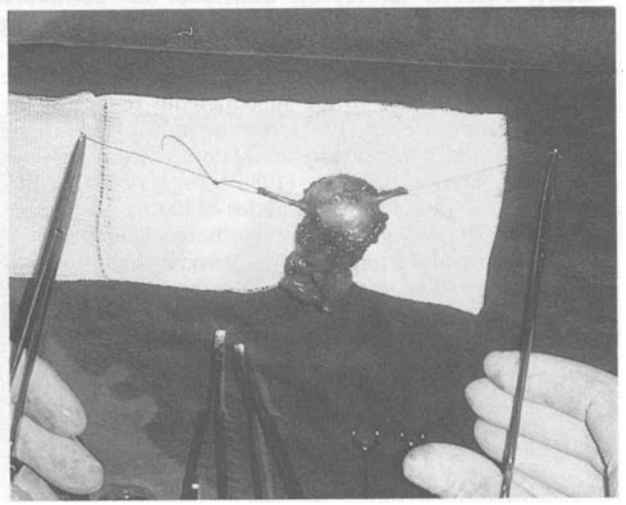

FIG. 2
Teratoma : note that it is encapsulated and is traversed by the hypoglossal nerve.

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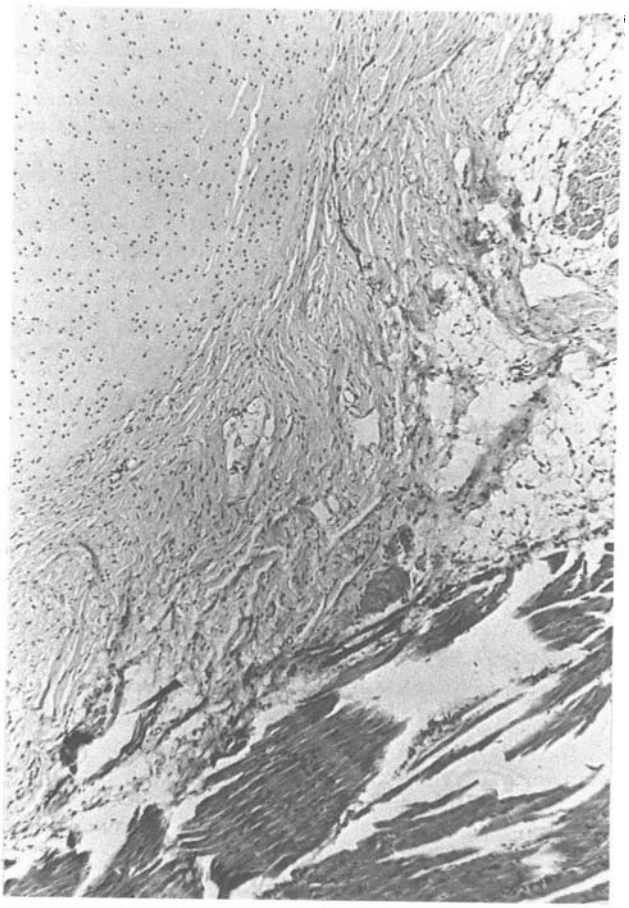

FIG. 3

Margin of tumour showing some mature cartilage and an ill-defined border. Normal striated muscle is present at the bottom of the photomicrograph.

cartilage and irregular spaces lined by simple cuboidal epithelium, often thrown up into papillary folds; this was regarded as choroid plexus differentiation. In addition there was mature neuroglial tissue that was both S100 and GFAP-positive immunocytochemically, distributed irregularly throughout the lesion. The hypoglossal nerve was not involved with the tumour.

Discussion

Weaver *et al.* (1976) defined a teratoma as a neoplasm, composed of multiple tissue types foreign to the site of origin, that displays progressive uncoordinated growth as opposed to a quiescent malformation. Certainly this last point is highlighted in the present case by its rapid increase in size in the six months prior to presentation and makes this unlikely to be a heterotopic mass. Further we have no evidence that it is a primary neural tumour. The lesion compresses rather than arises from the hypoglossal nerve and the mixture of tissues have not been reported in any benign peripheral nerve neoplasm. The tumour in our case is best regarded as a teratoma with elements from two germ layers. Our review of the literature suggests that oronasopharyngeal and basicranial teratomas are characteristically midline. Even in the case of cervical teratoma it is likely that the lump which presents clinically is in the majority of cases a lateral extension of the tumour. We found only two other cases of teratoma arising laterally in salivary glands. Shahid and Engeron (1975) reported the case of a benign teratoma arising in the parotid gland of a 24-year-old lady and Rose and Howard (1982) the case of congenital teratoma in the submandibular gland of a female infant. This latter case, quite apart from the anatomical site of the tumour, was similar to our case in that the teratoma was unusually small (2 cm in diameter). Also, as in our case histologically there was the normal preponderance of central nervous tissue.

The prognosis for head and neck teratomas is variable.

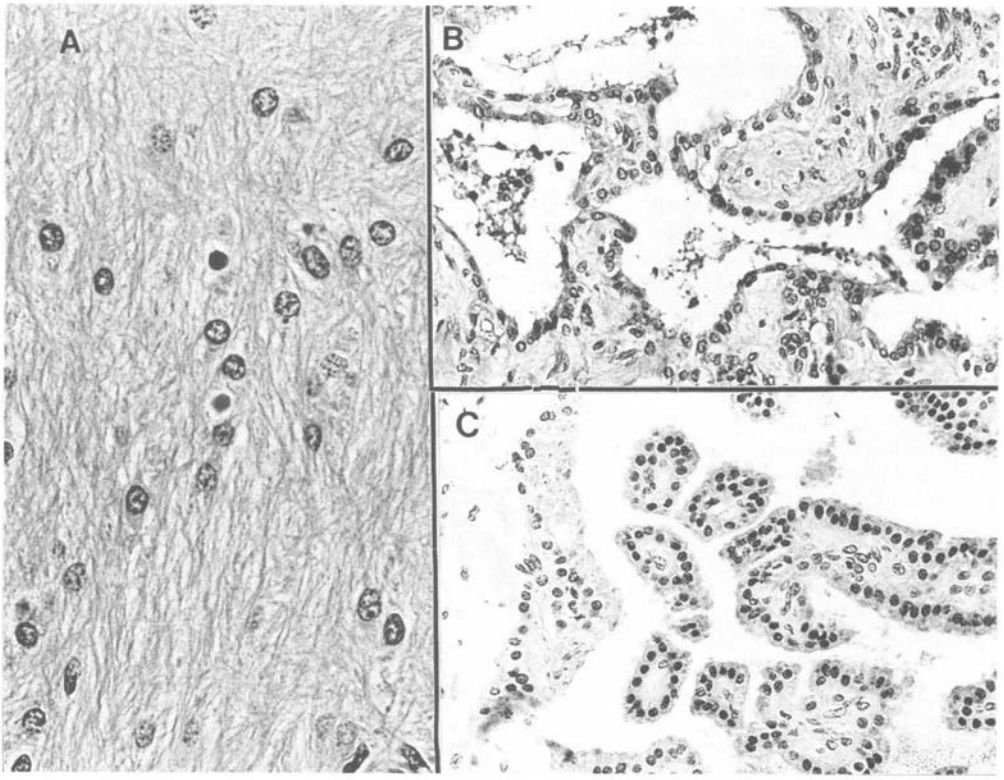

FIG. 4

Composite photomicrograph of (A) well differentiated glial tissue that was shown to be GFAP-positive by immunostaining. (B) and (C) show that in some areas there are slit-like spaces in dense fibrous tissue lined by cuboidal epithelium difficult to attribute to a specific tissue but in others the tumour forms a more delicate papillary pattern resembling the choroid plexus.

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Congenital teratomas pose a considerable risk to the airway and in a series of 18 cases reported by Byard *et al.* (1990) six were either stillborn or died within two days. The prognosis in the five per cent or so of malignant teratomas is also bleak. In the series of 20 malignant sinonasal teratomas reported by Heffner and Hyams (1984) the average survival after treatment was less than two years. However, the majority of head and neck teratomas are benign and excision is usually curative. In our case, there was no suggestion of malignancy histologically and as excision appeared complete, no further treatment was planned. The prognosis appeared excellent and at the one year follow-up there was no evidence of recurrence.

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