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Angiomyoma in the submandibular gland: a rare location for a ubiquitous tumour

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Abstract
Angiomyoma is a common soft tissue tumour of the head and neck that sometimes presents to the otolaryngologist; however, it seldom occurs in the major salivary glands. We present a case of angiomyoma arising in the submandibular gland, a tumour not described previously in the English literature.

Key words: Angiomyoma; Submandibular Gland

Introduction

Angiomyoma, also known as angioleiomyoma or vascular leiomyoma, is a ubiquitous mesenchymal tumour composed of convoluted thick-walled vessels and smooth muscle fascicles.[1,2] Although it typically involves the superficial soft tissues of the head and neck,[3–5] the occurrence in the major salivary gland is extremely rare.[6] To date, only five cases of parotid angiomyoma have appeared in the English literature.[6–8] We present the hitherto undescribed location of a common angiomyoma which, to the best of our knowledge, is the first example to be documented in the submandibular gland.

Case report

A 40-year-old woman presented with a firm swelling in her left submandibular region. The lesion had been present for one year and was described as increasing and decreasing in size periodically. The patient stated that there had been intermittent pain during mealtimes. Clinical examination revealed a solitary non-tender 2.0 cm mass in the left submandibular gland. Under the pre-operative diagnosis of pleomorphic adenoma, the tumour was removed *en bloc* with the gland. The post-operative course was uneventful.

Gross examination of the resected specimen revealed a well-circumscribed, firm, $2.0 \times 1.8 \times 1.5$ cm mass in the submandibular gland. The cut surface was homogenous and white-yellow in colour. Histologically, the multilobulated tumour was not encapsulated (Figure 1(a)) and composed of tortuous thick-walled vessels with lumina of varying sizes in addition to an intervascular proliferation of smooth muscle (Figure 1(b)). There was no entrapment of glandular elements within the lesion. Most tumour cells were shown to express α-smooth muscle actin and desmin with no reactivity to CD31, CD34 and S-100 protein (Figures 1(c) and 1(d)).

Discussion

Benign mesenchymal tumours of the major salivary glands are rare. In the files of Armed Forces Institute of Pathology, 220 cases, representing 1.4 per cent of all salivary gland tumours accessioned, have been registered.[6] Of these, 87.5 per cent of cases occurred in the parotid gland, whereas the submandibular gland accounted for 12 per cent. More than 60 per cent were vascular and neural tumours, and only 1.5 per cent were smooth muscle tumours.[6]

Angiomyoma of the major salivary gland is an exceptionally rare finding.[6–8] Natiella *et al.*[3] reviewed 67 cases of oral leiomyoma from the English literature and uncovered only one angiomyoma in the parotid gland. Subsequent survey of the worldwide literature from 1884 to 1992 disclosed four leiomyomas in the major salivary gland; two each in the parotid gland and submandibular gland.[4] However, they included the non-vascular solid type of leiomyoma. Another analysis of 109 cases of oral angioleiomyoma revealed no published accounts of major salivary gland lesions.[5] McDaniel[6] briefly mentioned the existence of three previously reported examples of angiomyoma in the major salivary gland: one each in the parotid gland,[8] submandibular gland and sublingual gland. Unfortunately, we were unable to find the latter two published reports by our MEDLINE- or PUBMED-based search. Recently, Wong *et al.*[9] reported an interesting case of angioleiomyoma attached to the submandibular gland. Unlike our tumour, their lesion was completely separated from the gland proper by the fibrofatty tissue.

The aetiopathogenesis of angiomyoma in the major salivary gland is unclear. Its predilection for the parotid gland may be attributed to the absence of a well-defined capsule and the presence of thick-walled vascular structures. It is well-known that the parotid gland is in close contact or embraces branches of the external carotid arteries and the internal jugular vein.[10] On the other hand, the submandibular gland is enveloped in a definite capsule and no larger blood vessels course through the parenchyma.

In summary, we report the first convincing case of angiomyoma occurring in the submandibular gland. Angiomyoma should be considered by the otolaryngologist as part of the differential diagnosis of salivary gland tumours.

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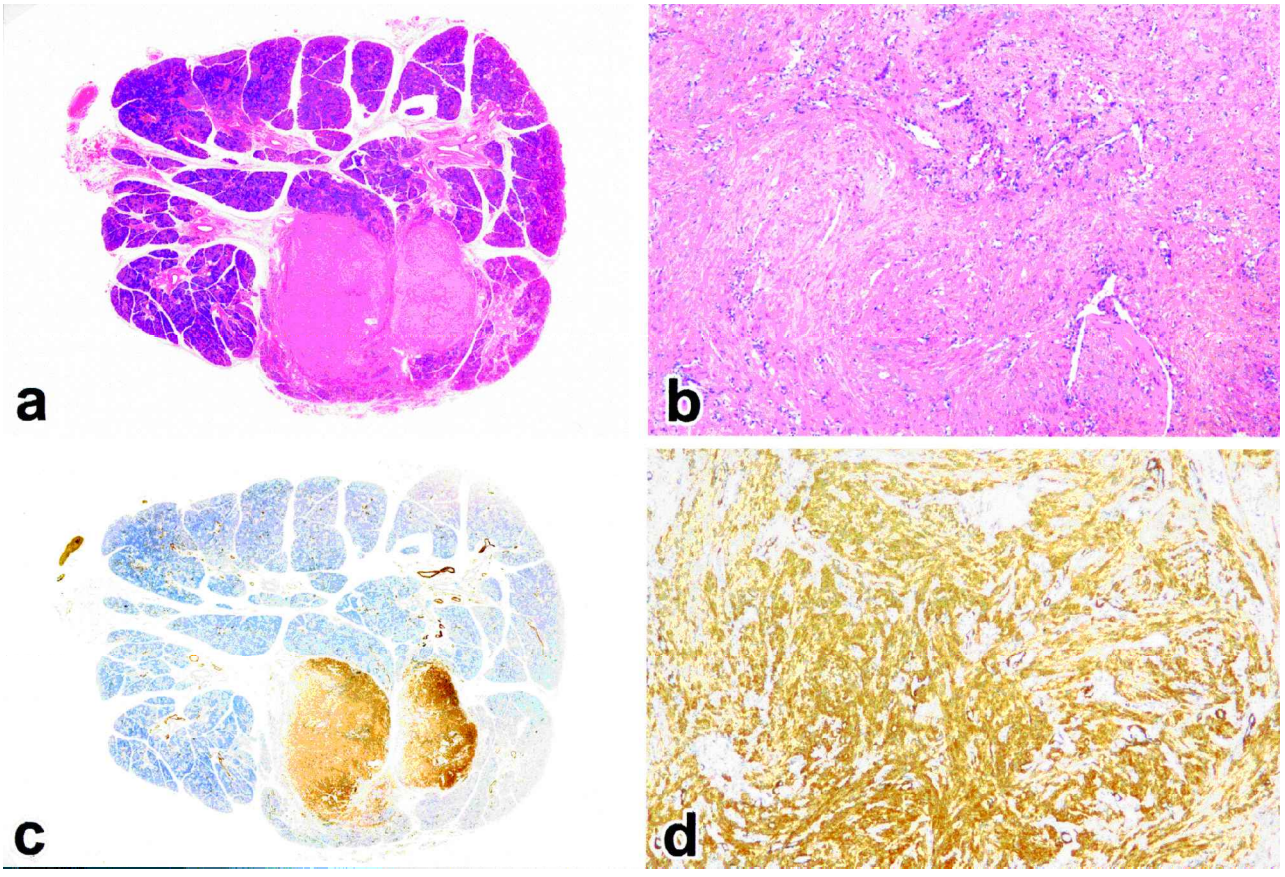

FIG. 1

(a) A well-demarcated multilobular tumour embedded in the submandibular gland (H & E; ×2.5). (b) Bundles of smooth muscle among convoluted thick-walled vessels (H & E; ×100). (c and d) Tumour cells are strongly positive for α-smooth muscle actin (Immunohistochemistry, (c), ×3; (d), ×100).

- **An angiomyoma in the submandibular gland is reported**
- **Such lesions in the major salivary glands are rare and have only been reported previously in the parotid**
- **This is the first reported case of angiomyoma in the submandibular gland**

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