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Short communication

Angiomyolipoma of the tongue

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Abstract

We present a rare case of angiomyolipoma of the tongue in a 23-year-old man. The clinical appearance was of a small, solitary, well-demarcated, painless mass. The oral angiomyolipoma followed a benign course with no recurrence after excision.

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Introduction

Angiomyolipoma is a benign tumour, which is histologically composed of groups of mature adipose tissue intermixed with convoluted thick-walled blood vessels, interlacing bundles, and irregularly arranged sheets of smooth muscle. It usually occurs in the kidney associated with tuberous sclerosis in the brain. Extra-renal angiomyolipoma can occur in organs such as the liver, lung, uterus, and skin. It is rare in the mouth. We know of only five cases of intraoral angiomyolipoma that have been reported in English; ^{4–7} three were on the hard palate and two on the lower lip.

Case report

A 23-year-old man was referred complaining of a small mass in the centre of his tongue (Fig. 1), which was painless, but had enlarged slowly for 2 years. Oral examination showed a firm mass $6 \text{ mm} \times 8 \text{ mm}$ with normal-coloured mucosa. The lesion was excised with a provisional diagnosis of fibroma. At operation the mass was well-demarcated and easily dissected. Histopathological examination showed an encapsulated lesion composed of proliferation of an intricate mixture of



Fig. 1. Photograph of a tumour on the tongue; it is well demarcated and covered with normal-coloured mucosa.

mature adipose tissue, blood vessels, and smooth muscle (Fig. 2). The vascular components varied in size, number, and type from capillaries to tortuous thick-walled vessels. The interlacing bands of smooth muscle were arranged around vascular spaces. No mitotic figures, nuclear pleomorphism, epithelioid cells, or immature adipocytes were evident. A diagnosis of angiomyolipoma was made. There were no signs, symptoms, or family history of tuberous sclerosis. After 4 years follow-up, there has been no recurrence.

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Table 1 Clinical characteristics of oral angiomyolipomas

First author	Reference no.	Age (years)	Sex	Site	Size (mm)
Redman	4	71	M	Lower lip	20 × 10
Piattelli	5	43	M	Hard palate	10
Yamamoto	6	62	F	Hard palate	10
Yamamoto	6	69	F	Lower lip	10×7
Guttmann	7	39	M	Hard palate	10
Our case		23	M	Tongue	8×6

They were all encapsulated. There were no signs of recurrence in a median of 2.5 years (range 1–7).

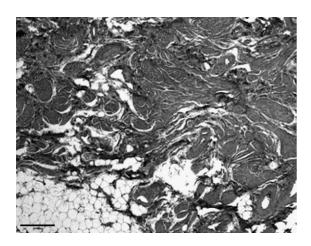


Fig. 2. Photomicrograph of an angiomyolipoma of the tongue (haematoxylin and eosin, original magnification $\times 60$). The tumour is composed of an intricate mixture of mature adipose tissue, blood vessels, and sheets and interlacing bands of smooth muscle. Vascular components vary from capillaries to tortuous thick-walled vessels (bar: $100 \, \mu m$).

Discussion

The five cases that we have found of oral angiomyolipoma^{4–7} (Table 1) occurred in patients between their third and seventh decades. There was no clear sex difference. All the tumours were encapsulated and small, ranging from 7 to 20 mm, and none recurred. Our patient had almost the same features as the others, except that he was younger.

Oral angiomyolipomas, despite the histological similarities, differ in several ways from renal ones, being solitary, small, well-demarcated masses. No patients had had tuberous sclerosis, and none developed any recurrence after excision. ^{4–7} However, renal angiomyolipomas are solitary, or multiple with relatively large masses, and sometimes invade locally. Such patients have usually been associated with tuberous sclerosis, ² and recurrence after resection, and malignant transformation, have been reported. ^{1,2,8}

It is rarely the initial preoperative diagnosis, ¹⁻³ and a final diagnosis requires histopathological examination. The

differential diagnosis includes lipomatous or myolipomatous tumours, angiomyoma, angiolipoma, haemangioma, fibroma, and fibrolipomatous hyperplasia.

Angiomyolipoma could be considered a hamartoma, but there is no consensus that these lesions are a single entity. 1–3 Only a few cases of myomatous hamartoma of the tongue have been documented as small, poorly-circumscribed masses, and these have been in infants or children. 9,10 We diagnosed our case as an angiomyolipoma because the mass had enlarged gradually with active proliferation, and was well-encapsulated. However, it could have been a hamartoma as the patient was younger than the other patients reported. It seems important to increase the documentation of both tumours in the mouth to better clarify the aetiology of oral angiomyolipomas.

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