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ANGIOMYOMA LEIOMYOMA OF TONGUE – A CASE REPORT AND REVIEW OF LITERATURE

Dental Science

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KEYWORDS

Introduction:

Angiomyoma in the oral cavity is an unusual presentation of a tumour that is more persistently found in the stomach and uterus **[Farman AG. Benign smooth muscle tumours. S A Med J 1975, 49, 1333-1340]**[1]**.** Blanc first reported a case of angiomyoma presenting in the oral cavity in as early as 1884[2]. **[Blanc E. Contribution à l'ude des tumeurs fibreuses de la langue. Gaz Hebdomad *de* Med et *de Chir* 1884, 21,611-613.]** This rare tumour makes up for only 0.42% of the total benign tumours of the oral cavity[3]. The rarity of this lesion in the oral cavity is attributed to the lack of smooth muscle in the oral cavity, as this is the site of origin of the tumour. The only places where smooth muscles are found is in the tunica media of the blood vessels and in the circumvallate papillae of the tongue[4].

Although the lesion is known by many names such as angiomyoma, vascular angiomyoma and vascular myoma reported incidences are very infrequently reported in contemporary literature. This case report serves to update the practitioners about this rare entity and gives a review of the available literature on the same.

Case Report:

An 83 year old male patient reported to the department of oral and maxillofacial surgery at our institute in Pune, India with a chief complaint of a recurring swelling along the right side of the tongue for the last 25 years. The swelling was not associated with any pain or discomfort, enlarged to about 1cm in diameter, regressed on its own and recurred in an erratic pattern subsequently. He patient first noticed the swelling on the right lateral border of the tongue around 25 years back. Patient gave no history of any trauma to the region and said that the growth was spontaneous in nature and not associated with any triggering factor for its recurrence. The swelling appeared to be the same colour as that of the tongue, was soft on palpation, showed no fluctuation but was compressible and reducible in nature. The patient had no contributory medical history and regional lymph nodes were also unremarkable. A provisional diagnosis of a mucocele was made and an excisional biopsy was planned under local anaesthesia. The excised lesion appeared to be of a thicker consistency than a mucocele and measured around 2x3cm in size (FIG 1). No complications of the excision procedure were reported and no evidence of recurrence has been seen in the two year follow up period since.

Histopathological Examination:

Histopathological examination of Haematoxyilin and Eosin stained section demonstrated a tumour made up of numerous vascular spaces lined with endothelial cells and interlacing bundles of smooth muscle fibres(FIG 2). Connective tissue stroma was composed of middle-sized, thick-walled blood vessels separated by fibrous stroma whereas some areas showed vascular spaces surrounded by concentric layers of interlacing smooth-muscle(FIG 3).

Masson trichrome (MT) staining showed muscle fibres of the lesion stained a purple colour indicating the presence of myofibrils in the tumour cells(FIG 4). The immunohistochemical stained section showed a strong positivity for the marker α-smooth muscle actin (αSMA). There was a strong and diffuse immunostaining observed for myogenic protein like α-SMA, confirming the muscular origin of the neoplastic cells in this tumour (FIG 5).

Discussion:

According to Brooks *et al.* the overall incidence rate of angiomyoma in the oral cavity is around 0.016%[5]

There are some theories that discuss about the origin of this lesion. It has been suggested the lesion might originate from tunica media of blood vessels, circumvallate papillae and heterotopic smooth muscle.

Another theory suggests vascular leiomyoma as a stage in a process of smooth muscle proliferation[6-8]. It has also been proposed that angiomyoma can develop as a result of infection, trauma, hormones and arteriovenous malformation[8-9].

Pericytes are the progenitors for vascular smooth cells (vSMC) in angiomyoma. These are the mesenchymal-like cell, associated with the walls of capillaries and show a potential to differentiate into other mesenchymal cell types e.g. smooth muscle cell, fibroblast and osteoblast[10]. Similarity of vSMC and pericytes with the help of markers like α-SMA and desmin show that vSMC and pericytes are phenotypic variants of a mural cell type. Pericytes have an intermediate phenotype between vSMC and fibroblasts. Irritants can make pericytes to give rise to vSMC[11].

The diagnosis of angiomyoma is only based on its histological features since it does not present with any special clinical characteristic[6].

Histologically, the proliferation of smooth muscle cells may resemble other benign spindle-shaped lesions giving us the differential diagnosis such as neurofibroma, schwannoma, fibromatosis, fasciitis, fibrous histiocytoma, solitary myofibroma, spindle cell lipoma, peripheral nerve sheath tumor, hemangiopericytoma and leiomyosarcoma[5,6]. Special stains like MT or phosphotungstic acid haematoxylin (PTAH) and immunohistochemical markers such as αSMA, desmin and vimentin can be helpful in the diagnosis. In our case, tumor cells showed strong positive reactivity to SMA and MT stain. These characteristics revealed the muscular origin of the tumor cells. The final diagnosis was confirmed as angiomyoma.

AUTHOR	SITE
Marden F[12] 2004	Tongue
Srinath VS[13] 2004	Soft Palate
SH Al-Amad[14] 2006	Hard Palate
Manor E[15] 2007	Buccal Vestibule
R Luaces Rey[16] 2007	Retromolar Trigone
Y-H Kim[17] 2010	Buccal Space
Y-H Kim[18] 2010	Tongue
Gueiros LA[19] 2011	Upper and Lower Lips
Mendetti[20] 2012	Gingiva
Eley KA[21] 2012	Hard Palate
M Veeresh[22] 2013	Hard Palate
Tsuji T[23] 2014	Hard Palate
Ranjan S[24] 2015	Gingiva
Osano H[25] 2015	Buccal Mucosa
Inaba T[26] 2015	Buccal Mucosa
da Silva LAB[27] 2017	Upper Lip

FIG 1

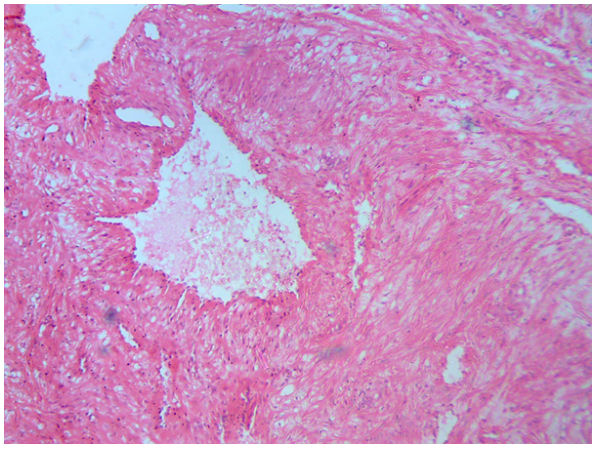

FIG 2

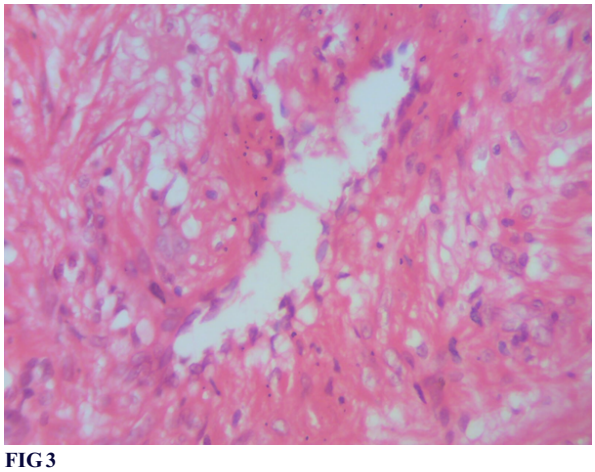

FIG 3

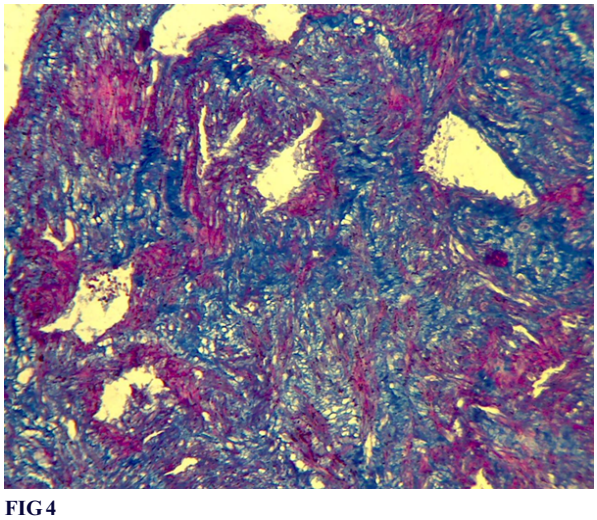

FIG 4

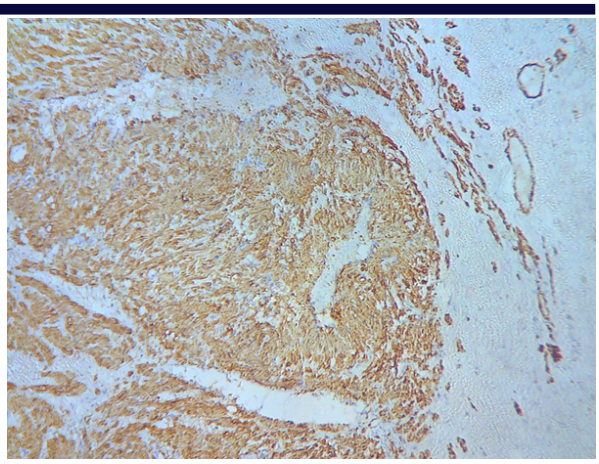

FIG 5

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