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Case Report

Angiomyxolipoma: Unusual swelling of oral cavity

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Introduction

Angiomyxolipoma (AML) is a rare variant of lipoma which presents as well circumscribed tumor characterized by proliferation of adipose tissue with a myxoid stroma and multiple thick and thin walled blood vessels. Review of literature reveals 12 cases of AML with only one case of AML of oral cavity being reported till date. We present a rare case of AML of floor of mouth with relevant review of literature.

Case report

A 70-year-old male patient presented with gradually progressing swelling of floor of mouth of one-year duration. On examination there was a 3 × 3 cm well circumscribed, partly

mobile, firm, non-tender swelling in the floor of mouth anteriorly displacing tongue posterosuperiorly with no attachment to the tongue (Fig. 1). There was mild restriction of movement of tongue due to mass effect but patient had no difficulty in mouth opening or swallowing. Indirect laryngoscopy showed a normal base of tongue, vallecula and endolarynx. CECT neck revealed a well-defined enhancing soft tissue density lesion measuring 32 × 29 × 36 mm in the floor of mouth anteriorly with abutment and displacement of tongue posterosuperiorly (Fig. 2). A punch biopsy of the lesion from the oral cavity was taken. Based on clinical behavior and histologic and immunohistochemical reports, a diagnosis of angiomyxolipoma was reached.

The patient underwent CO₂ laser assisted peroral wide local excision of the lesion. CO₂ laser was used at 5 W, continuous, superpulse mode with use of bipolar coagulation of large vessels. Intraoperatively the lesion was well circumscribed mass with smooth surface and rubbery consistency with no involvement of tongue muscles or submandibular gland. The mucosal defect in the floor of mouth was closed primarily with absorbable sutures (Fig. 3). Cut surface of the lesion was yellowish brown with interspersed gelatinous material. Post op. was uneventful and the patient started oral feeds by third postoperative day.

Final histopathology report revealed adipocytes with background showing extensive myxoid change and numerous proliferating capillaries (Fig. 4). Immunohistochemistry studies of the spindle cells were positive for Vimentin, CD 34 and negative for Desmin, S100 whereas adipocytes expressed S100 and vascular smooth muscle cells expressed Vimentin. These histologic and immunohistochemistry findings

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Fig. 1 – Smooth rounded swelling in floor of mouth pushing the tongue posterosuperiorly.

resulted in a final diagnosis of MPNST. Presently patient is on followup for past two years without recurrence.

Discussion

Lipoma is a common, benign soft tissue neoplasm consisting of mature adipocytes usually found in the subcutaneous tissue of the trunk, proximal limbs, thigh and neck. 20% of lipomas are found in the head and neck region with 1–5% found in the oral cavity. Based on histology, location and presence of other tissue elements they can be classified as classic, spindle cell/pleomorphic, myxoid, vascular, smooth

Fig. 2 – CECT scan reveals a well circumscribed lesion in the floor of mouth.

Fig. 3 – Primary closure of mucosal defect of floor of mouth.

muscles, osteo, fibroblastic tissue and fibro/chondrolipoma. One of the rare variants of lipoma is AML (vascular myolipoma) which was first described by Mai K et al, who identified spindle cells with fat vacuoles (preadipocytes) in transitional areas between myxoid and lipomatous components in electron microscopy.¹

AML is a very rare variant with only 12 cases reported so far in current literature (Table 1). There is a clear male predilection with only one case so far reported in a 60-year-old female in the thigh region. The patients age ranged from 9 to 69 years (mean = 44.58 yrs) with the cases being more common in 5th and 6th decade.^{1–12} There is no predilection to a particular race or ethnicity. The most common sites of AML is subcutaneous tissue of scalp, torso and extremity with one case reported in the spermatic cord, neck and oral cavity.^{1–12} Our case is the second recorded AML of oral cavity.

The lesion is usually asymptomatic and present as slow growing, painless well demarcated solitary subcutaneous

Fig. 4 – Histopathology slide showing benign adipocytes with extensive myxoid change and numerous proliferating capillaries.

Table 1 – Review of cases of angiomyxolipoma in the literature.

Sl no	Sex/ Age	Location	Author (ref)	Year
1	M/32	Spermatic cord	Mai KT et al (1)	1996
2	M/57	Scalp	Zamecnik M et al (2)	1999
3	M/50	Upper back	Okafor O et al (3)	2000
4	F/60	Thigh	Sciot R et al (4)	2001
5	M/66	Scalp	Tardio JC et al (5)	2004
6	M/44	Arm	Lee HW et al (6)	2005
7	M/57	Wrist	Sanchez Sambucety P et al (7)	2007
8	M/43	Subungual area	Kang YS et al (8)	2008
9	M/36	Neck	Usta U et al (9)	2009
10	M/69	Hip	Song M et al (10)	2009
11	M/09	Knee	Kim HJ et al (11)	2010
12	M/12	Oral cavity	Martínez-Mata G et al (12)	2011

mass. The tumor usually shows no signs of local invasiveness or malignant transformation. In our case due to the large lesion in the anterior part of floor of mouth the patient had slight difficulty in tongue movements and swallowing.

The diagnosis of AML rests on characteristic histopathologic and immunohistochemical features. Though the presence of mature fat tissue without lipoblast with paucicellular myxoid areas and numerous thin and thick walled vascular channels is diagnostic, the lesion needs to be differentiated from other variants of lipomas. Immunohistochemistry plays an important role in identifying the various elements in the tumor and differentiate it from other tumors. The spindle cells of the myxoid areas usually express CD 34 and Vimentin and not Desmin, S100 or SMA. The vascular smooth muscle cells express SMA and Vimentin whereas the mature adipocytes are S100 positive.¹⁰ Important differential diagnosis includes myxolipoma, angiolipoma, myolipoma, myxoid/vascular/pseudoangiomatous spindle cell lipomas, angiomylipomas and angiomyxomas. The histopathological characteristics of AML is the presence of vascular, myxoid and lipomatous elements, one of which is usually absent in angio, myo or myxolipoma. The tumors with smooth muscle like myolipoma and angiomylipoma can be differentiated with HMB 45 expression. Myxoid spindle cell lipoma can be differentiated with the characteristic 'ropy' collagen bundles, sparse vascular features with mild positivity to CD 34, whereas absence of myxoid areas and vascular structures help in differentiating vascular and pseudoangiomatous spindle cell lipomas respectively.^{5,9}

CT scan and MRI are important radiological investigations which help in defining the extent of lesion and involvement of

adjacent structures. Treatment is wide local excision and in all the cases reported so far, no recurrence has been reported.

Conclusion

Angiomyxolipoma is a rare benign tumor with characteristic histopathological and immunohistochemical features which can be managed surgically with complete cure.

Conflicts of interest

All authors have none to declare.

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