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Leiomyoma of the Gingiva

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ABSTRACT

Leiomyomas are benign tumors of smooth muscle origin which occur more commonly in the uterus and the gastrointestinaltract. Oral leiomyomas have been reported in the literatures which are commonly present in the lip, tongue, buccal mucosa, palate and mandible. Most of the reported cases are in the 4th and 5th decades of life with very few cases reported in pediatric age group. Most of the lesions are small, asymptomatic and present as a smooth elevated area with pigmentation. Intraosseousleiomyoma's have also been reported in the mandible where it mimics a cyst. We present an 8 year old Indian (Asian) boy who presented with a large exophytic mass with a granulomatous surface on the lingual aspect of the mandible. The size, the site and the age of presentation makes this a rare case worth to be recorded.

Key words: Leiomyoma, mandible, immunohistochemistry.

INTRODUCTION

Myoma is a non-malignant neoplasm arising from the muscle tissue. Histologically they are of two types – Rhabdomyoma, arising from striated muscle tissue and Leiomyoma being of smooth muscle origin.[1]Leiomyomas generally appear in the location where the smooth muscle is present. The most common area of occurrence is the uterus (95%) followed by the gastrointestinal tract and skin. The incidence is quite rare in the oro-facial region accounting tojust about0.065%[2,3,4,5,]. Oral leiomyoma shows a male predilection of 1.43 : 1[1,2,6,7,8] and usually presents itself at around 40 – 49 years of age.[1,2,3,7,8]Clinically oral leiomyomas present as an asymptomatic, smooth elevated area[7,8,9,10]and occur mostly on the tongue, lip, palate and cheek[2,3,5,11]. Hereby we present a case report of leiomyoma arising from the gingiva on the lingual aspectof the right side of the mandible in a young male patient.

Case report

A 10 year old Indian (Asian) boy reported to the out-patient clinic of the Department of Oral & Maxillofacial Surgery with the complaint of a large mass on the lingual aspect of his right mandible in relation to the molar region. The lesion has been present (noticed) for 6 months and has been growing gradually. Apart from getting traumatized frequently during mastication it was largely asymptomatic. The patient experienced minor bleeding episodes which stopped spontaneously. No paraesthesia of the lower lip or tongue was reported.

Clinical examination revealed an exophytic mass with a granular surface, measuring about 3 x 4 cm in size. The lesion extended anteriorly from the deciduous first molar to the first permanent mandibular molar on the right side. Superiorly the lesion extended to the line of occlusion and inferiorly up to the floor of mouth. Medially the lesion was adherent to the lingual

aspect of mandible with a broad base. The lesion was firm in consistency with a pale pink color (Fig 1,2,3). No regional lymphadenopathy was evident.

Radiographic evaluation was unremarkable showing no signs of bony involvement. Blood investigations were performed, including the calcium profile and serum alkaline phosphatase. All the parameters were within normal limits.

The differential diagnoses considered wereEpulis or Granular cell tumor. Langerhan's cell Histiocytosis was largely ruled out with no bony involvement noticeable on radiography and normal serum alkaline phosphatase levels. Brown's tumor of hyperparathyroidism again was ruled out on similar grounds and normal calcium profile. However a Giant cell lesion or malignancy like sarcoma could not be ruled out.

Routine pre-anesthetic work up was carried out and the patient was admitted in the hospital. Considering the age of the patient, an excision biopsy under general anesthesia was planned and performed. Intra-operatively the lesion peeled away from the adjacent lingual aspect of the mandible uneventfully. The pedicle was broad extending from the deciduous second molar to the distal aspect of permanent first molar(Fig 4,5,6). The area was thoroughly curetted and haemostasis achieved. Coe-pack dressing was applied. Post operative recovery was uneventful. The soft tissue healing was complete in 2 weeks time. Patient remains free of any recurrence in the last 12 months follow up. The excised specimen was send for histopathological examination and immunohistochemical studies.

Histopathology and Immunocytochemistry

The routine H & E section showed the lesion with parakeratinized squamous epithelium on the surface and underlying mature fibrous connective tissue stroma. The tumor was composed of interlacing fascicles of spindle cells with eosinophilic cytoplasm and elongated nuclei. There was also the presence of few compressed slit like vessels. No mitotic activity was seen.

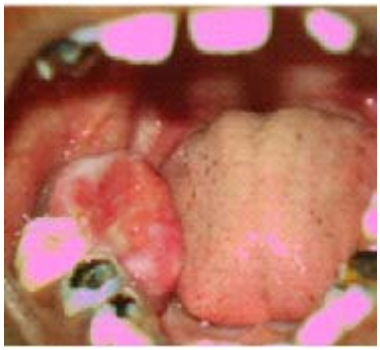
Histological differential diagnosis included Leiomyoma, Nodular fasciitis,Neurilemoma and Myxoma. Immunohistochemistry (IHC) studies were performed for confirmation of diagnosis.

IHC studies were done for tissue markers S-100 and SMA (Smooth Muscle Actin). SMA was strongly positive and S-100 was negative confirming the diagnosis of Leiomyoma.

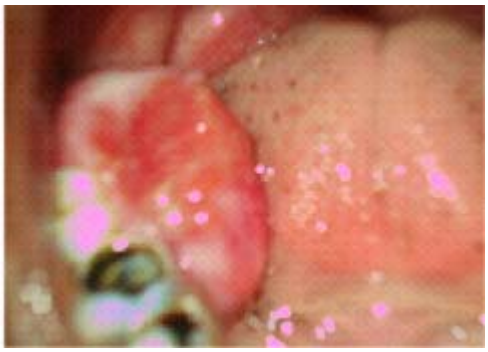
In identifying the sub type, the presence of compressed vessels favors the diagnosis of Solid variant of Leiomyoma. Angio variant which is widely reported in the head and neck region is usually characterized by dilated vessels surrounded by smooth muscle layer.

DISCUSSION

Leiomyoma is a benign tumor of smooth muscle origin occurringcommonly in the uterus and GIT. According to English language medical literature Oral leiomyomas are a rarityaccounting for less than 1% of all leiomyomas[1-5].Of 7,748 identified cases of leiomyoma Farman reported that only 5 were located in the oral cavity[1]Natella etal[1]and Baden etal[12] on the basis of literature



1- Intra-oral view of lesion



2 - Close up view of lesion

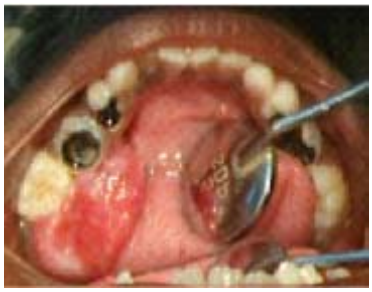
describing cases since 1884 cited only 78 cases of Oral leiomyoma in 1982. Since then only 6 new cases have been reported[1].

Origin of the leiomyomas of the oral cavity is restricted to areas with smooth muscle. These areas are: 1) Tunica Media of blood vessels 2) DuctusLingualis 3) Circumvallate papillae.[2]Most common sites of occurrence of oral leiomyomas are tongue, lips, palate and cheek. Other less frequent locations are the floor of the mouth and gingiva.[2]An intra-osseous presentation of angio-leiomyoma presenting as either a unilocular or multilocular cysts on radiographs have also been reported[10].

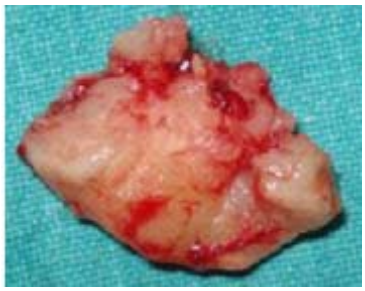
Clinically oral leiomyomas are unspecific masses which are well defined, slow growing andusually asymptomatic. Symptoms induced due to local growth include difficulty in deglutition, toothache, lose teeth or referred pain in TMJ. Pain is suspected to be prompted by local ischemia due to tumoral vessel contraction and due to neural irritation near the tumor. Pain may also be induced due to surface ulceration.[2]Generally these tumors do not undergo malignant transformation.

WHO distinguishes three different histological variants of leiomyoma – vascular leiomyoma, solid leiomyoma and the epitheloid leiomyoma.[1,6,13]In the oral cavity, vascular or angio-leiomyoma is the most common variant (75%) followed by the solid variant (24%). Epitheloid variant (Leiomyoblastoma) is said to be extremely rare accounting for less than 1%[3,11,14]

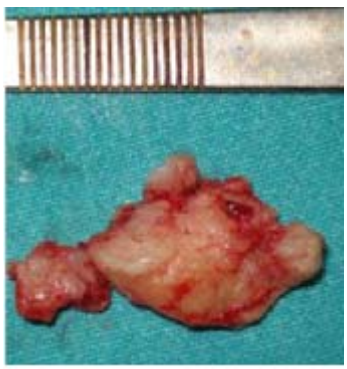
Angioleiomyomas are derived from smooth muscle of the blood vessel. The smooth muscle fibres of the vessels exhibit a circular distribution around the lumen and are associated with collagen fibres.Solidleiomyomas are well defined tumors which consist of a network of fusiform cells with collagen and with varying vascularization.Leiomyoblastoma (epitheloid leiomyoma) is composed of round or polygonal cells with clear areas surrounding the nucleus and an acidophilic cytoplasm. Smooth muscle fibers are rarely found[1,5].Special stains such as Van Gieson's



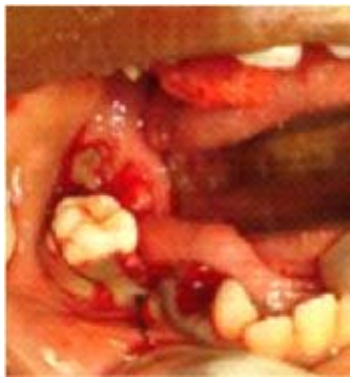
3- Mirror view of lesion



4 - close up of excised specimen



5 - excised specimen

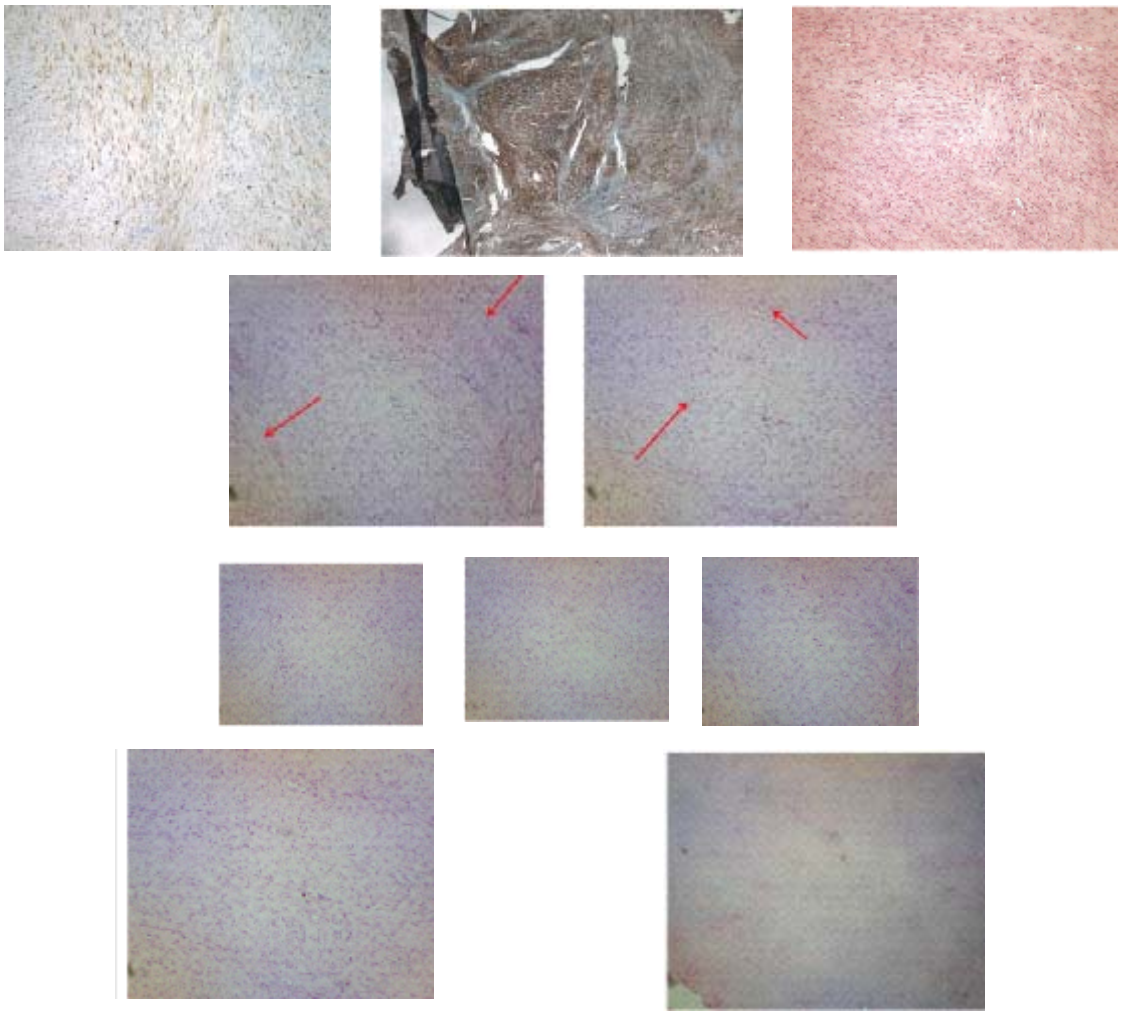


6 - Post operative wound healing

stain and Masson Trichromic stain can be used to stain muscle cells and collagen. Mallory Phosphotungstic acid – Hematoxylin (PTAH) is specific for myofibrils[1,5,6,10].

Immunohistochemistry plays a very important role in the diagnosis of leiomyoma. Immunohistochemical studies reveal a positive expression for actin (smooth muscle actin) and vimentin whereas expression for cytokeratin and S-100 is negative.[6,7,12]SMA(smooth muscle actin) corresponds to the alpha fraction of the actin chain and is specific immunomarker ofsmoothmusclecells.Vimentin is a structural protein of the cytoplasmic filrils of mesenchymal cells[7].

Leiomyoma can be easily confused with other mesenchymal tumors such as fibroma, neurofibroma, low-grade fibrosarcoma or leiomyosacoma.[1,7] Differential Diagnosis should also include vascular tumors such as hemangioma, lymphangioma or pyogenic granuloma; Salivary gland neoplasms (Pleomorphic Adenoma, mucocele etc.); Dermoid cysts and other fusocellular tumors like myopericytoma and myofibroma[7] and other spindle cell neoplasms which include nodular fasciitis, neurilemmoma, schwannoma and Myxoma.[4,7,9.10] Definitive diagnosis of oral leiomyoma can be achieved with histopathological examination and Immunohistochemistry.



Histology & Immunohistochemistry images

Treatment of choice is complete surgical removal. Incidence of recurrence following complete removal is rare. Recurrences reported in the literature have been attributed to incomplete removal[2,3,7].

CONCLUSION

Leiomyoma is a rare oral tumor. Tumors in young children have to be treated with due attention as theycan be locally aggressive. However Leiomyoma responds to simple surgical excision and rarely recurs. Prognosis is very good in cases of complete removal. IHC studies are invaluable in confirming the diagnosis.

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