Angioleiomyoma of cheek: report of two cases

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Abstract Leiomyomas are benign tumors derived from smooth muscle that most commonly arise in the uterus, followed by the gastrointestinal tract and subcutaneous tissues. Deep presentations are typically asymptomatic unless they achieve substantial size, whereas subcutaneous presentations may be associated with considerable local tenderness and pain, in addition to alteration of the overlying skin color. The highest incidence occurs between 40 and 49 years of age in the orofacial region, the angioleiomyoma is the predominant subtype, representing nearly 75% of all reported cases [1]. Oral leiomyomas are considered uncommon neoplasms, accounting for about 0.065% [2]. Despite the rather characteristic histopathologic features of the leiomyoma, a definitive diagnosis often depends on the confirmation of smooth muscle origin by immunohistochemical studies. We report two cases of angioleiomyoma of cheek with their clinical presentation histopathological features and surgical techniques employed. No recurrence has been seen as of yet in both of our cases.

Keywords Angioleiomyoma · Oral cavity · Leiomyosarcoma

Introduction

Leiomyomas are the widely acknowledged as rare lesion in oral cavity. The vascular leiomyoma is a subtype of leiomyoma that is highly vascular. It may arise from the smooth muscle of arteries and veins and frequently is richly innervated. Vascular leiomyomas are composed of blood vessels with thick, smooth muscle walls. Baden et al. [3] reviewed the literature and found only in five cases (0.0062%) of oral leiomyomas in a series of 7,748 leiomyoma from all sites. Leiomyomas has been divided into three types in the world health organization classification: leiomyoma (solid), angioleiomyoma (vascular leiomyoma) and epithelioid leiomyoma (leiomyomablastoma). We report two cases of angioleiomyoma with their clinical presentations, histological features and which were treated with surgical excision. No recurrence has been seen as of yet.

Case report: 1

A 32-year-old healthy male reported to the Department of Oral and Maxillofacial

Surgery with complaint of a painless swelling in the left cheek since past five years. He had noticed a small nodule in that region initially, which disappeared after hot fomentation. Two months later, the nodule re-appeared and gradually increased to the present size, with no significant symptoms.

Clinical examination showed, a solitary well circumscribed unilobular swelling over the left side of face measuring 4.5cms x 4cms, soft to firm in consistency with no involvement of the skin. There was no associated tenderness or regional lymphadenopathy. Intraorally, the overlying mucosa was normal. Routine blood and urine investigations were found to be in normal limits

CT scan was evident of a large non-homogenously enhancing mixed density lesion in the infra-temporal fossa and the buccal space. It showed a moderately enhancing soft tissue component superolaterally, with no calcification or necrosis.

Aspiration biopsy did not yield cystic or hemorrhagic fluid. An incisional biopsy stained with H & E section (Fig. 1) shows multiple blood vessels with proliferation of smooth muscle wall surrounding it and

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nuclei typically elongated with blunt ends. The overall histopathological features were suggestive of angioleiomyoma.

Surgery was performed under general anesthesia. The soft tissue mass was approached through an intraoral horizontal incision in the buccal mucosa. The buccinator muscle was dissected to reach the mass. The mass was then excised in toto, and primary closure done in layers. The postoperative recovery was uneventful. The histopathology of the specimen confirmed the diagnosis of angioleiomyoma. No signs of recurrence observed in the follow up of 6 years.

Case report: 2

A 32-year-old healthy male was referred to the Department of Oral and Maxillofacial Surgery with complaint of a painless swelling in the upper left side of the face since 2 years.

Patient was apparently normal 2yrs back when a swelling appeared on the left side of the face. The swelling was initially small in size and slowly increased and was associated with pain. Patient visited a

H & E stained sections show numerous blood vessels, most of which have a thick muscular coat. Nuclei of smooth muscle cells are elongated with blunt ends.

Fig. 1 H & E section of case (10X)

Fig. 2 Patient photo case II showing the extraoral presentation of lesion

Fig. 3 Surgical procedure, a) Preoperative b) Lesion in toto with capsule c) Residual surgical defect d) Specimen of lesion

Fig. 4 Histopathology of angioleiomyoma a) H & E stain (10x) b) Masson Trichrome stain (10x)

private hospital for which he was treated with an extraoral incision and drainage suspecting a space infection. Patient was not relieved of symptoms for which he was referred to the Department of Oral and Maxillofacial Surgery, V S Dental College and Hospital.

On examination, an ovoid swelling was present over the left side of the middle third

of the face measuring about 3cms x 3cms extending superiorly from the line joining lateral canthus of eye to upper part of tragus 3cm inferiorly to the line joining lateral corner of mouth to posterior border of the ramus. Posteriorly 2cm in front of tragus and anteriorly around 2cm to the left ala of nose. Skin over the swelling appeared normal (Fig. 2). On palpation, swelling was

firm, tender, fixed with distinct borders and with no rise in temperature and skin over the swelling was mobile.

Intraorally, a diffuse swelling present over the upper left buccal vestibule extending from mesial aspect of 26 to the middle of 28 with obliteration of buccal vestibule. Opening of stensons duct appeared normal and mucosa over the swelling appeared normal. On palpation the swelling was firm, tender and fixed, with no rise in temperature. Routine blood and urine investigations were found to be within normal limits.

Plain film radiograph suggested no bony changes. Ultrasound showed a hypoechoic lesion more in favor of soft tissue swelling. FNAC was done and revealed cohesive clusters of spindle and ovoid cells. Some cells having cigar shaped nuclei with blunt ends. With the above investigation a final diagnosis of benign spindle cell tumor possibly a leiomyoma was done.

The surgery was carried out under general anesthesia (Fig. 3). Extraoral elliptical incision was given enclosing the scar and fibrosed tissue. Further dissection was carried out to reach the capsule of the mass and it was released from surrounding attachments. The lesion was excised in toto and sent for histopathological examination. Fibrosed scar tissue was also removed and closure done in layers.

Histopathology of the stained H & E sections showed that cells were spindle shaped and arranged in long fascicles and in a swirling fashion. The cells had eosinophilic cytoplasm with blunt ends with cigar shaped nuclei. Many blood vessels with thickened walls were also found (Fig. 4a).

The special stain with Masson-trichrome stain showed that cells were of smooth muscle origin confirming the diagnosis of angioleiomyoma (Fig. 4b).

Patient is on regular follow-up and no recurrence seen in one year.

Discussion

Leiomyoma is the most common benign neoplasm of the uterus and smooth muscle. The first case of oral leiomyoma was reported by Blanc in 1884. Because of the scarcity of smooth muscle in the oral cavity, the Leiomyoma is relatively uncommon in that location. Leiomyomas are divided histologically into 3 major subtypes: Leiomyoma (solid), Angiomyoma (angioleiomyoma, vascular leiomyoma), and Epithelioid leiomyoma (leiomyoblastoma or bizarre leiomyoma). Angioleiomyoma are infrequently found in oral cavity, representing only 0.06% of all the lesions [3].

The review by Baden et al. [3] showed mean age of 41 years for all types of oral leiomyomas, where as Brook et al. [6] in their study found a mean age of 47 years. In contrast both of our cases were in early third decade. The most frequent site of occurrence of oral Angioleiomyoma was the lip seen in 48.6%, palatal lesions occurred in 21.1%, buccal mucosa and tongue were next in frequency with 9.2% of cases.

Oral angioleiomyomas generally present as a painless slow growing lesion, as seen in our cases. Makoto et al. [4] reported a case of painful Angioleiomyoma of lower lip. They discussed the mechanism of pain production by tumor, and said it may involve contraction of tumor vessel, producing ischaemia within the neoplastic tissue. The length of time before the patients seek medical attention may span months to years.

The clinical differential diagnosis relevant to the angioleiomyomas usually include other benign mesenchymal tumors like fibroma, lipoma, and neurofibroma, vascular lesions including lymphangioma and hemangioma, benign lesions of salivary gland origin, such as deep-seated mucocele, and soft tissue cysts, such as dermoid cyst.

The plain radiography in case of soft tissue tumors does not give the relevant information. CT scan will be of great importance in such cases and shows non-homogenous enhancing mixed density lesion. But in the cases involving the bone, Burke EJ [5] reported an asymptomatic radiolucency in the mandible on routine panoramic radiograph. Surgical specimen was made to reach the diagnosis of vascular leiomyoma.

Histologically, the proliferation of smooth muscle cells may resemble other benign lesions composed primarily of spindle shaped cells, such as neurofibroma, neurileiomyoma, and fibrous histiocytoma. In these cases, routine histochemical stains such as Masson Trichrome, Mallory's phosphotungstic acid and immunohistochemical markers like SMA (smooth muscle actin) can be useful in identification of smooth muscle nature of the cells [6]. Hemangiopericytoma, hemangioendothelioma, and hamartomatous lesions containing smooth muscle, such as vascular and neurovascular hamartomas, may also be considered in a histopathologic differential diagnosis [7].

The malignant counterpart, the leiomyosarcoma should be taken into account when the number of mitotic figures per field is over ten. Differentiation between leiomyoma and low grade leiomyosarcoma is not always easy. Some leiomyomas might be difficult to differentiate from the malignant leiomyosarcoma; the number of mitotic figures, anaplasia, and bizarre cell forms are considered as the features that can help in predicting the degree of malignancy. Most investigators agree that if fewer than 5 mitoses per 20 high-power fields are found, the prognosis is generally good, but that the prognosis worsens progressively as more mitoses became apparent. In one of our cases 3–4 mitotically active cells were seen and patient was kept on a regular follow up. However, the use of only mitotic figure counts for the distinction of benign from malignant lesions is not always reliable.

Mesquita et al. [8] reported a case of leiomyosarcoma of the buccal mucosa in a 23-year-old male patient and described the immunohistological features that were useful in arriving at the final diagnosis. Surgical excision with a margin of normal tissue is preferred for leiomyosarcoma and regional lymph node dissection is only recommended if there is clinical evidence of lymphadenopathy.

The treatment of choice of oral angioleiomyomas is of surgical excision. Despite the vascular nature, the profuse bleeding during removal is rarely seen. Intraoral approach gives the good esthetic result which was followed in one of our case, but in second case extraoral approach was preferred over the intraoral as the patient had a scar extraorally because of an attempted incision and drainage. The scar tissue was included in the incision to give an esthetic result.

Recurrence is uncommon. It has been noted that, however, that the smooth muscle neoplasm in oro-facial region have a higher proportional incidence of malignancy than their counter parts in the female genital tract [9]. In addition, the differentiation of leiomyoma from a low grade leiomyosarcoma can be difficult histopathologically. This suggests that careful long-term follow up may be warranted for all the smooth muscle neoplasms of oral cavity. In our cases there has been no evidence of recurrence seen as yet after complete surgical excision and patient are being followed up regularly.

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