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CASE REPORT

Cellular Angiofibroma of the Buccal Mucosa: A Rare Case Report and Review of Literature

Kamran Bokhari, Luqman Manikandan, Ajmal Mohammad, Mohammad Shahul, Mustafa Abdul Bagi, Zaheer Kota

ABSTRACT

Background: Cellular angiofibromas are rare benign mesenchymal neoplasms characterized by spindle cell component and numerous small vessels. They are commonly well-circumscribed, localized in the superficial soft tissues. The lesion is more frequently seen in the inguinoscrotal or vulvovaginal regions and occurs equally in men and women, and rarely involves oral cavity. Due to its clinical and histological similarity with other mesenchymal tumors, such as angiomyoma, hemangioma, lymphangioma and hemangiopericytoma, angiofibroma presents a diagnostic dilemma.

Case report: Very few cases of angiofibroma involving maxillofacial region have been reported in the literature. This case report involves cellular angiofibroma involving left buccal mucosa in a 23-year-old female patient. Following detailed clinical examination, radiological interpretation and histopathological diagnosis, surgical excision was performed. The patient was followed-up on a regular basis and was disease free.

Summary and conclusion: Benign angiofibroma involving oral cavity is a rare tumor. There is a close resemblance of this tumor with other mesenchymal lesions and, thus, faces a diagnostic challenge. Surgical excision with a long-term follow-up gives good prognosis to this lesion—benign cellular angiofibroma.

Keywords: Angiofibroma, Mesenchymal tumor, Oral soft tissue swelling.

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INTRODUCTION

Angiofibromas are uncommon, highly vascular, histologically benign but locally aggressive tumors.[1] Nucci et al are credited for describing cellular angiofibroma as an uncommon benign mesenchymal tumor in 1997.[2] These lesions usually arise in the inguinoscrotal or vulvovaginal regions and occur equally in men and women.[3] In the head and neck region, angiofibromas are more predominant in the nasopharyngeal region. Juvenile nasopharyngeal angiofibroma is the most common benign tumor of the nasopharynx as reported in the literature. Since this occurs more commonly in the second decade of life, it is proposed that nasopharyngeal angiofibroma is a testosterone dependent tumor.[4] Pathogenesis of angiofibromas is not very clear. Various etiological factors have been mentioned in the literature. It is proposed that these mesenchymal tumors can appear as a result of infection, trauma, hormones and arteriovenous malformation.[5,6]

Due to a wide spectrum of similar mesenchymal tumors reported in the literature with very close clinical, histological interrelations, these tumors pose a diagnostic challenge. This may cause relevant problems with the differential diagnosis in view of the tumor's rarity, ambiguous clinical presentation and histologic features. A definitive diagnosis and clear treatment plan can only be formulated after correlating all the three components (i.e. clinical, histological and radiological features). Angiomyoma, angiolipoma, mucocele and benign minor salivary gland tumors are few of the lesions which share close clinical correlation. Within the group of angiofibromas, the giant cell variant, lipomatous variant and the atypical variant are the ones which can be differentially diagnosed on the basis of histopathological examination.[7,8]

We report an interesting and rare case of benign cellular angiofibroma involving left buccal mucosa treated by surgical excision in a 23-year-old female patient.

CASE REPORT

A 23-year-old female patient reported to our center with a chief complaint of chronic swelling in the left buccal mucosa since 3 years (Fig. 1). On clinical examination, a reddish purple well-circumscribed swelling measuring approximately 2 × 2 cm was noticed. Anteriorly, the swelling was almost approaching the corner of mouth. There was no history of chronic cheek bite or irritation. No significant extraoral

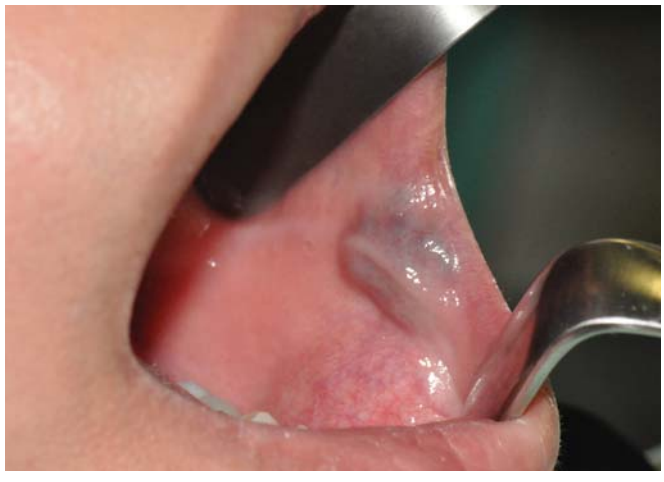

Fig. 1: Clinical view

swelling or asymmetry was evident. The swelling was firm in consistency and nontender on palpation. Mucosa over the swelling was normal with absence of any ulcerations or sinus discharge. There were no other secondary changes involved like paresthesia or cervical lymphadenopathy.

Radiographic examination was not significant with absence of any calcifications. Aspiration was performed under topical anesthesia and yielded collection of frank blood which was subjected to cytological examination. Initial diagnosis of mucocele which was made on the basis of clinical appearance of the lesion was ruled out based on the aspirate and histological report as angiomatous lesion. The patient was then advised to undergo color Doppler of the lesion to rule out any active feeder vessel characteristic of hemangioma. Ultrasound findings were negative in terms of vascularity or a feeder vessel (Fig. 2). Based on the clinical, histological and ultrasound findings, treatment was planned for excision under local anesthesia. Dissection in the submucosal plane revealed a well-encapsulated soft swelling which was friable but could be separated from the surrounding connective tissue and muscle plane (Fig. 3). Complete hemostasis was achieved, and primary closure performed. The excised specimen (Fig. 4) was sent for histological examination which confirmed the diagnosis of cellular angiofibroma. Figure 5 shows fibrous connective tissue with bundles of collagen with moderately proliferating fibloblasts along numerous dilated blood vessels.

There were no specific complaints postoperatively and the patient was discharged immediately. The patient was disease-free without any recurrence during the follow-up period.

DISCUSSION

Cellular angiofibroma represents a rare benign mesenchymal tumor which occurs mainly in the superficial soft tissue of the genital region.[9] Incidence in the head and neck region is rare and, within this region, the nasopharyngeal angiofibroma has been reported more in the literature. They are commonly well-circumscribed, localized in the superficial soft tissue and characterized by bland spindle-shaped cells arranged without any pattern in a stroma with wispy collagen and numerous small to medium sized thick

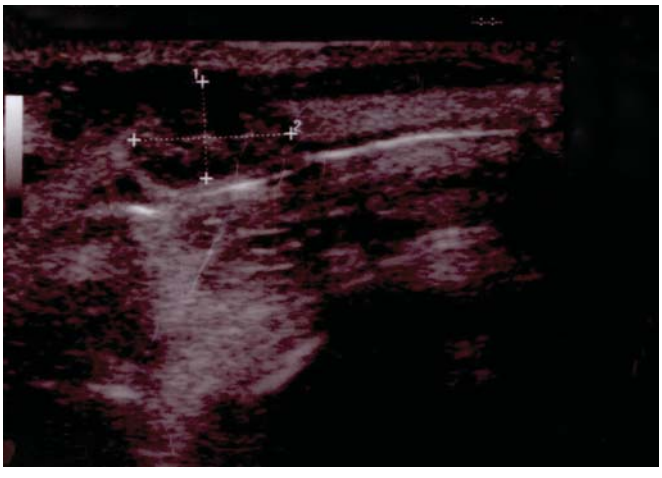

Fig. 2: Ultrasound report

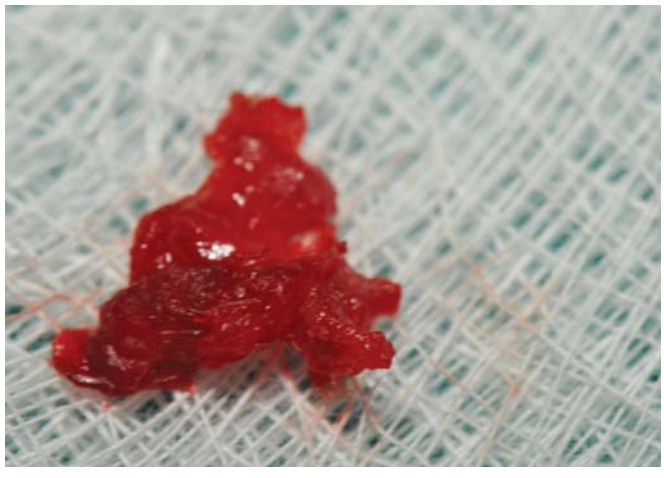

Fig. 4: Excised specimen

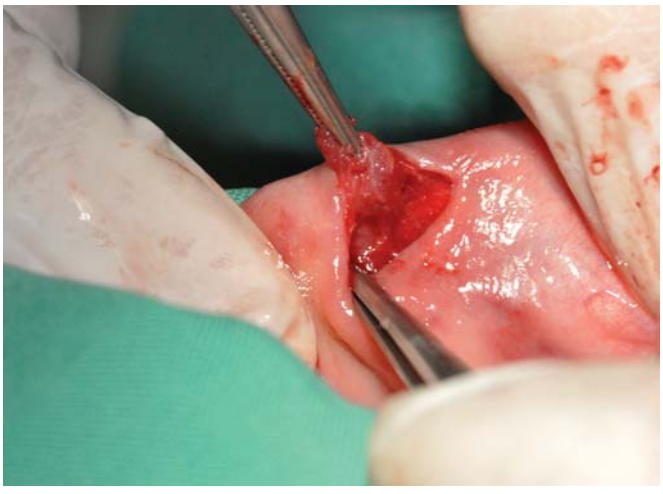

Fig. 3: Intraoperative view

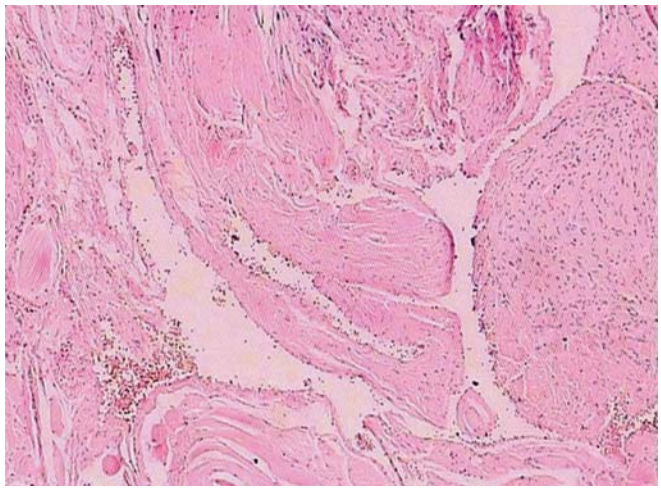

Fig. 5: Histopathological findings

walled vessels.[10] A close interrelation occurs within this set of mesenchymal tumors. Flucke U et al analyzed clinicopathological and immunohistochemical features of 25 cases of cellular angiofibroma. They concluded that there is a link between cellular angiofibroma, spindle cell lipoma and mammary type myofibroblastoma showing a spectrum of one entity with morphological variations dependent on anatomic location.[9] Angiofibromas though histologically benign are locally aggressive tumors and to elaborate their aggressive nature. Barnes L et al studied a series of 31 cases of angiofibromas by flow cytometry to determine, if they exhibited any abnormalities in DNA content.[1] They concluded that although angiofibromas are benign tumors, they can be locally destructive if not recognized early and treated aggressively. Their findings were based on the fact that six of the 31 nasopahryngeal tumors had intracranial extension despite surgical approaches that were selected for total removal.

The diagnosis of angiofibroma is dependent more on histological confirmation due to its close resemblance with other lesions, such as angiomyoma, hemangioma, lymph-angioma and hemangiopericytoma. Treatment of choice for a benign case of cellular angiofibroma is surgical excision.[10] Eleanor C et al studied clinicopathologic features in 13 cases of cellular angiofibroma with morphologic atypia or sarcomatous transformation.[3] They concluded that the biologic significance of atypia or sarcomatous transformation in cellular angiofibroma remains uncertain.

Most of the cases of cellular angiofibroma reported in the literature are the nasopharyngeal ones and the surgical guidelines, recurrence potential imply them. Those cases reported with occurrence in the buccal mucosa are the giant cell variant and thus no clear characteristic features, treatment guidelines are specific to cellular angiofibroma of the buccal mucosa. However, based on the clinical presentation, histological features, it can be concluded that cellular angiofibromas in the buccal mucosa are benign lesions and can safely be treated with local surgical excision with an adequate follow-up period.

CONCLUSION

1. The incidence of cellular angiofibroma in the bucccal mucosa is rare.
2. This tumor can be easily misdiagnosed solely on the basis of clinical examination.
3. The tumor follows a benign course and can safely be treated by local surgical excision.

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