Cukurova Medical Journal
ÇUKUROVA ÜNİVERSİTESİ TIP FAKÜLTESİ

Cukurova Med J 2018;43(2):471-474
DOI: 10.17826/cumj.342618

OLGU SUNUMU / CASE REPORT

Non–infiltrating angiolipoma of palate

Damağın infiltre olmayan anjiyolipoması

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Cukurova Medical Journal 2018;43(2):471-474

Abstract
Angiolipomas, histopathological variant of lipoma are benign mesenchymal tumor characterized by proliferation of mature adipocytes and interspersed with angiomatous proliferation composed of several congested blood vessels. Although they are the most common tumor of trunk and extremities but they are exceedingly rare in oral cavity according to their capsulation they can be classified into infiltrating and non infiltrating. Intraorally tongue, buccal mucosa and floor of the mouth are commonest sites, but hard palate is a very rare site for angiolipoma. Here we report a case of angiolipoma of palate in a 39 year old male. The present case also emphasized the cytopathological features of angiolipoma.
Key words: Angiolipoma, lipoma, fat tissue neoplasms, Non infiltrating, Intra oral, cytology, histopathology

Öz
Lipomların histopatolojik varyantı olan anjiyolipomalar, benign mezenkimal tümördür ve olgun adipositlerin çoğalması ile karakterizedir ve birkaç tıkalı kan damlasından oluşan anjiyomatöz proliferasyon serpiştirilmiştir. Gövde ve ekstremitelerin en sık görülen tümörü olmalarına rağmen, kapsülleşmelerine göre ağız boşluğunda çok nadir görülürler; infiltre olan ve olmayan olarak sınıflandırılabilirler. İntraoral dil, bukkal mukoza ve ağız zemini en sık görülen bölgelerdir ancak sert damak anjiolipom için çok nadir bir yerdir. Burada, 39 yaşındaki bir erkekte damak anjiyolipoması olan bir olguyu sunmaktayız. Mevcut olguda ayrıca anjiyolipomanın sitopatolojik özellikleri vurgulanmıştır.
Anahtar kelimeler: Anjiyolipom, lipom, yağ dokusu neoplazileri, sızdırmazlık, intra oral, sitoloji, histopatoloji

INTRODUCTION

Although not common in mouth, lipoma is one of the commonest benign mesenchymal tumors. Histopathologically they can be classified on the basis of the appearance of connective tissue stroma. Angiolipoma, osteolipoma, spindle cell lipoma, myolipoma, chondrolipoma and myxoid lipoma are some histopathological variants of lipomas[1]. Angiolipoma is a rare histopathological variant of lipoma characterized by sheets of mature adipocytes in a vascular stroma[2]. Very few cases of oral angiolipoma have been reported.

CASE

A 30 year old male presented to our institute for the evaluation of a painless swelling on the upper, left, posterior region of the jaw from 5 months. The swelling was initially small and gradually it reached the present size. Past medical and family history of the patient was not relevant to the persistent symptom. Intra–oral examination revealed a yellowish white swelling extending from the left maxillary tuberosity to the midline of the palate measuring about 4.5 × 2 cm. On palpation the swelling was found to be soft and fluctuant. The overlying mucosa was yellowish white in color. No sign of ulceration and drainage were present. (Figure 1) Computed tomographic scan revealed that the lesion was superficial and was not associated with the bone. Fine needle aspiration was done and a clear blood tinged fluid was yielded. FNAC exhibited a collection of mature fat cells with extra vasated RBCs and few plasma cells. (Figure 2) Cytopathological features were consistent to lipoma.

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Geliş tarihi/Received: 03.04.2017 Kabul tarihi/Accepted: 01.05.2017

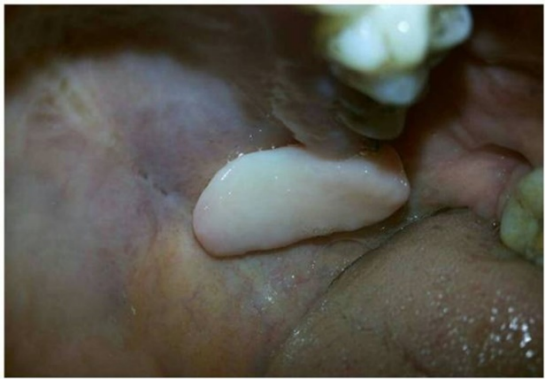

Figure 1. Clinical picture of the lesion.

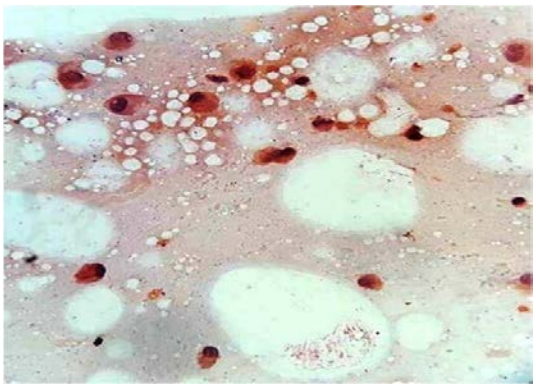

Figure 2. FNAC revealed mature adipocytes with few RBCs and plasma cells. (Hematoxylin and Eosin staining X 20)

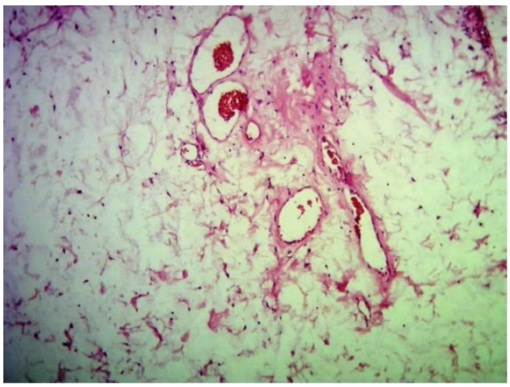

Figure 3 Sparse connective tissue stroma with mature adipocytes. (Hematoxylin and Eosin staining X20).

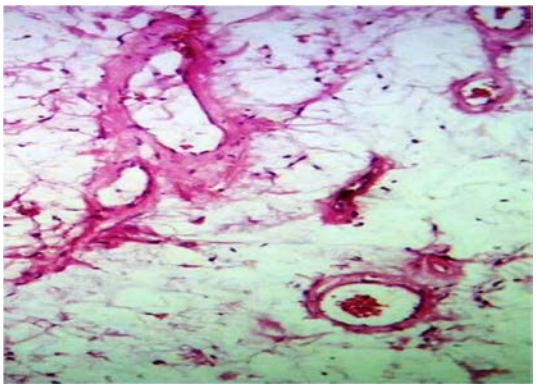

Figure 4. Dilated blood vessels lined by endothelial cells with mature adipocytes. (Hematoxylin and Eosin staining X40),

The lesion was completely excised under local anesthesia and tissue was sent to the Department of Oral and Maxillofacial Pathology of NIMS Dental College Jaipur (India), for histopathological evaluation. Follow up period of 1 year was uneventful.

Histopathological examination revealed a sparse connective tissue stroma with sheets of numerous mature adipocytes (Figure 3) and numerous dilated blood vessels lined by endothelial cells and having RBCs inside, (Figure 4) few areas showed hemosiderin pigmentation.A focal collection of inflammatory cells was noted, comprised of lymphocytes and plasma cells. The overlying epithelium was normal stratified squamous epithelium. Based on cytopathological and histopathological features, a final diagnosis of Non infiltrating angiolipoma was given.

DISCUSSION

Angiolipomas are benign tumor of fat tissue histopathologicaly composed of mature adipose tissue intermixed with dilated vascular channels[3]. They represent about 1 – 5 % of all neoplasm of oral cavity[2]. Angiolipoma was first described as a distinct entity by Bowen in 1912[4]. Angiolipoma has two histological types: infiltrating and non infiltrating. Non infiltrating angiolipomas are encapsulated[2]. Clinically non – infiltrating angiolipomas are usually painless while Infiltrating or diffuse neoplastic or non-neoplastic proliferations of mature fat may cause compression of vital structures or may be confused with atypical lipomatous neoplasm/well-differentiated liposarcoma[3,4].

They have been identified in various age groups ranging from infancy to ninth decade[4]. Their precise etiology and pathogenesis remains unclear. Trauma has been implicated in some cases. Lipomatous differentiation by hormones during puberty, fatty degeneration of a central hemangioma and vascular proliferation of a congenital lipoma (congenital origin) have been implicated as etiological factor, but most of the cases remain idiopathic[5]. The size of the angiolipomas can vary from 1 cm – 4 cm. The most common sites of angio;ipomas are forearms, upper arms, thighs and abdomen. Males are more commonly affected than females[6].

Oral angiolipomas are very rare. Intraorally hard palate is the very rare site for the occurrence of angiolipoma[2, 3, 6]. The first case of the angiolipoma of the palate was reported by Davis GB et al in 1976 [2]. The second case was reported by Flaggert JJ et al in 1986 in an 8 year old girl[7]. In present case patient was 29 years old without any history of trauma. The size of the tumor was 4.5 × 2 (cm). As clinical differential diagnosis hemangioma, leiomyoma, fibroma, neurilemmoma, kaposis sarcoma can be considered; certain differentiating features help in making the definitive diagnosis (Table 1)[3,4,7-11]. Histopathological differential diagnoses include angiomyxolipoma, angiofibroma, angioleiomyoma, hemangioma (Table 2) [7, 8, 10, 11].

The following is the hisopathological diagnostic criteria for angiolipoma(Table 3)[12]. In present case the diagnosis was made by the correlation of the clinical, cytological and histopathologiacal features. The present case fulfilled all the diagnostic hitopathological features of non – infiltrating angiolipoma. There is no evidence that angiolipomas undergo malignant transformation[2, 7]. In present case also there were no malignant changes noted. Surgical excision is the treatment of choice for angiolipoma. Carbon dioxide laser and liposuction may be alternative treatment options. Present case was treated by surgical excision[3]. A pubmed database was searched and only 3 cases of angiolipoma have been found affecting palate. Hence the case presented here is a rare occurrence of angiolipoma. Angiolipomas of oral cavity has not been reported to recur.

Table 1. Clinical differential diagnosis of angiolipoma with characteristic features [3, 4, 7-11]

S. No	Differential diagnosis	Characteristic features
1.	Hemangioma	a) Diascopy is positive. b) Presence of phleboliths in computed tomography. c) Pulsation and fluctuations are present. d) Usually bluish to slight purplish in color.
2	Irritational fibroma	a) Association of any traumatic or irritational factor is present clinically.
3	Neurilemmoma	a) Clinical demarcation is not possible, histopathological evaluation is required.
4	Kaposis Sarcoma	a) Occurs as an ulcerative exophytic growth. b) Usually reddish in color.
5	Pyogenic granuloma	a) Usually occurs on gingival. b) Highly vascular and telangiactatic lesions, tend to bleed on palpation.
6	Benign salivary gland tumors	a) Histopathological evaluation is required.
7	Peripheral osteoma	a) Bony hard swelling. b) Shows patchy radiopacity on radiographs.
8	Leiomyoma	a) Clinical demarcation is not possible. Histopathological evaluation is required.

Table 2. Histopathological differential diagnoses of aniolipoma with characteristic features [7,8,10,11] (

S. No	Differential diagnoses	Characteristic features
1	Angiomyxolipoma	Histopathologically Angiomyxolipomas show an admix of mature adipocytes with dilated blood vessels in a myxoid stroma.
2	Angiofibroma	Histopathologically angiofibromas show a pronounced vascular tissue in a fibrocellular stroma. Lipomatous tissue is absent
3	Angioleiomyoma	Histopathologically Anggioleiomyomas consist of fassicles of spindle shaped cells having cigar shaped nuclei, with multiple torturous blood

		vessels. Lipomatous tissue is absent.
4	Hemangioma	Histopathologically, hemangiomas show multiple vascular channels, they are devoid of lipomatous tissue.
5	Liposarcoma	Liposarcomas can be confused with angiolipomas but can usually be differentiated by the presence of embryonal adipose tissue, pleomorphism, increased number of mitosis, and metastasis
6	Angiomyolipma	Angiomyolipomas are the commonest benign tumor of the kidney and is strongly associated with tuberous sclerosis. Histopathological differentiation between angiomyolipoma and angiolipoma is difficult. Immunohistochemically angiolipomas show positive expression for HMB 45 and Smooth muscle actin.
7	Infiltrating lipoma	Infiltrating lipomas consist of lesional fat tissue infiltrating in the deeper tissue, in the form of long thin streaks radiating from the intratumoral mass.

Table 3. Histopathological diagnostic criteria of angiolipoma [12]

S. NO	Diagnostic criteria
1	Histopathologically the lesion is encapsulated (Non infiltrating angiolipoma) or poorly encapsulated (Infiltrating angiolipoma)
2	Evidence of 50% mature adipocytes in the tumor
3.	Interspersed angiomatous proliferation in the tumor.
4.	Fibrinous microthrombi.
5.	Absence of other mesenchymal elements (smooth muscle) or pleomorphism

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