Epithelioid hemangioma in the oral mucosa – A case report

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SUMMARY

Introduction Epithelioid hemangioma is an uncommon benign vasoproliferative neoplasm that usually manifests as multiple red nodules in middle-aged adults.

Case Outline A 52-year-old male patient presented with a one-year history of a nodular lesion in the left buccal mucosa measuring 3 cm. The clinical hypothesis was lipoma. An excisional biopsy revealed a circumscribed lesion composed of lobules of vessels with perceptible or poor lumina, associated with a prominent inflammatory infiltrate consisting of eosinophils, histiocytes and chronic inflammatory cells. The endothelial cells composing the lesion had an epithelioid morphology and contained abundant eosinophilic cytoplasm. Immunohistochemistry for CD34, factor VIII, collagen IV, alpha-smooth muscle actin, and mast cells, as well as histochemical staining with Weigert's orcein were performed.

Conclusion Vascular proliferations of soft tissues are a diverse and morphologically complex group of lesions that are difficult to diagnose. This report presents a case of oral epithelioid hemangioma, highlighting relevant morphological and immunohistochemical features that could help distinguish this condition from other neoplasms.

Keywords: angiolymphoid hyperplasia with eosinophilia; epithelioid hemangioma; oral mucosa

INTRODUCTION

First described by Wells and Whimser in 1969 as angiolymphoid hyperplasia with eosinophilia, epithelioid hemangioma (EH) is an uncommon benign vasoproliferative neoplasm whose etiology and pathogenesis continue to be a matter of debate [1, 2, 3]. The term epithelioid hemangioma was proposed by Enzinger and Weiss in 1983 [1]. Despite the acceptance of this term since 1983, other terms have been used to describe this lesion, including angioblastic hyperplasia with eosinophilia, nodular angioblastic lymphoid hyperplasia with eosinophilia, lymphofolliculosis, pseudopyogenic granuloma, atypical pyogenic granuloma, and inflammatory angiomatous nodule [4].

EH commonly affects young adults and manifests as red or brown multiple nodules or small papules in the intradermal or subcutaneous region of the head and neck [1, 2, 4, 5, 6]. Extracutaneous lesions located in muscles, bone, and salivary glands are uncommon, and lesions involving oral mucosa are rarely observed [1, 4, 6, 7].

Histologically, EH is composed of well-developed, but frequently immature, vascular structures lined by aggregates of epithelioid- or histiocytoid-like endothelial cells that contain abundant eosinophilic cytoplasm. The endothelial cells are usually associated with a prominent inflammatory infiltrate consisting of dispersed lymphocytes, eosinophils, histiocytes and mast cells [1, 3, 6]. Most lesions are well circumscribed and have a lobular architecture. Lymphoid follicles with a germinal center may be present [1, 4, 5]. Although recurrence is observed in one third of cases of EH, metastasis is extremely rare and surgical excision generally leads to cure [5].

This case report presents an oral EH, highlighting morphological and immunohistochemical features that are important for the differential diagnosis of this lesion.

CASE REPORT

A 52-year-old black male patient was admitted to the Surgery and Oral-Maxillofacial Traumatology Service with main complaint of having a lump in the cheek that started to appear one year previously. The patient was a non-smoker and non-drinker, had no history of systemic alterations, and laboratory parameters were within the normal range. Intraoral clinical examination revealed a sessile swelling in the left buccal mucosa with a smooth erythematous

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Figure 1. View of oral mucosa lesion; note the swelling in the left buccal mucosa with a smooth erythematous surface, which measured 3 cm in its major diameter

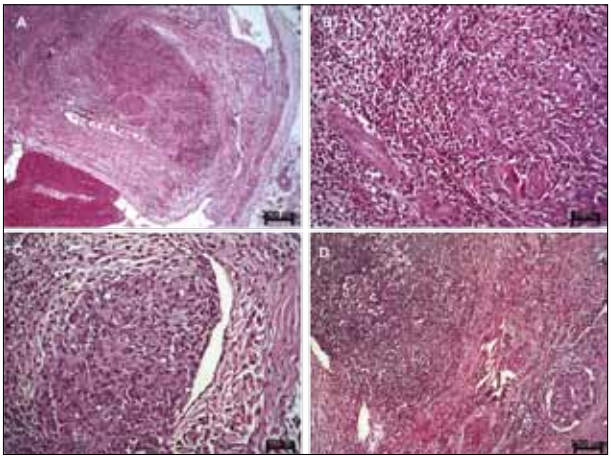

Figure 2. Histological sections of EH stained with hematoxylin-eosin: (A) well-delimited lesion exhibiting proliferation of endothelial cells in a lobular arrangement; (B) note the lobule composed of epithelioid endothelial cells near vascular spaces and associated with numerous eosinophils; (C) detail of the blood vessel wall surrounded by epithelioid cells; (D) left, lymphoid follicles; right, blood vessels with a thickened wall surrounded by epithelioid cells

surface, which was tender to palpation and measured 3 cm in its major diameter (Figure 1). The patient reported no pain to manipulation and there were no signs of an association with dental trauma or infection. Under the clinical hypothesis of lipoma, the patient was submitted to an excisional biopsy. The lesion was removed with safety margins and exhibited defined limits, no adherence to deeper planes, and no signs of excessive bleeding.

The specimen was sent for histopathological analysis, which showed a well-delimited lesion characterized by lobular proliferations. The lesion was composed of epithelioid endothelial cells with eosinophilic cytoplasm located near large vascular spaces or immature vessels. Vascular damage characterized by thrombosis and fibrointimal proliferation was seen in some wider vessels. Another finding was the abundant presence of eosinophils, sometimes near

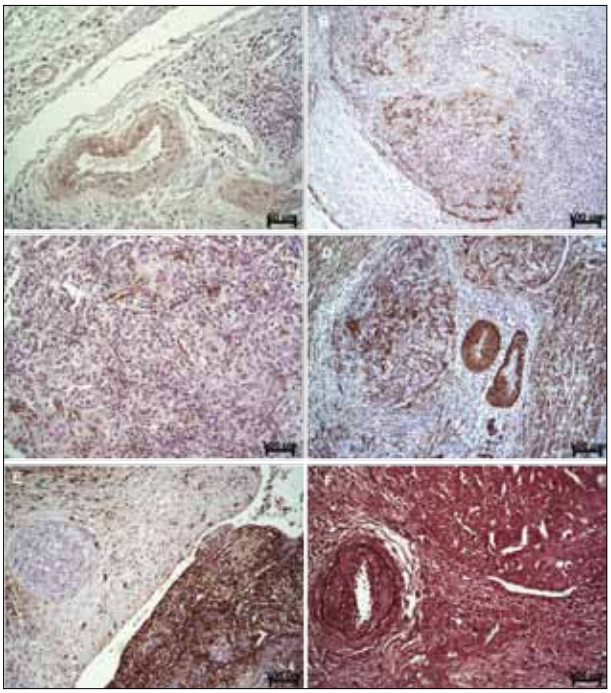

Figure 3. Histological sections of EH submitted to immunohistochemistry (A–E) and histochemistry (F): (A) note the immunostaining for collagen IV around blood vessels; (B) epithelioid endothelial cells and small-caliber vessels immunostained for factor VIII in a lobular arrangement; (C) CD34-positive vessels in an area rich in epithelioid endothelial cells and eosinophils; (D) strong immunostaining for α-SMA in vessels of different calibers; (E) right, note the presence of mast cells amidst numerous epithelioid endothelial cells; (F) Weigert's orcein staining around intact blood vessels, demonstrating the presence of elastic fibers; note the absence of elastic fibers in an area rich in endothelial cells.

lymphocytes, as well as formation of lymphoid follicles (Figures 2A–D). Mitotic figures and cellular atypia were absent. Immunohistochemistry for collagen IV, factor VIII, CD34, alpha-smooth muscle actin (α-SMA) and mast cells was performed, as well as histochemical staining with Weigert's orcein (Figures 3A–F). The antibodies, sources, dilutions and antigen retrieval methods used are shown in Table 1.

Immunohistochemical staining was positive for collagen IV and factor VIII around blood vessels (Figures 3A and 3B). The epithelioid endothelial cells were positive for factor VIII (Figure 3B), and CD34-positive vessels were observed in areas rich in epithelioid endothelial cells and eosinophils (Figure 3C). Strong α-SMA immunostaining was detected in small- and medium-caliber vessels (Figure 3D). Numerous mast cells were also found, especially in areas rich in epithelioid cells (Figure 3E). Staining with Weigert's orcein revealed the absence of elastic fibers in areas rich in epithelioid endothelial cells (Figure 3F).

The diagnosis of EH was based on the clinical and histopathological features. The patient has been followed up for approximately 18 months and shows good healing and no signs of recurrence.

DISCUSSION

This case report presents a 52-year-old patient with EH involving the cheek mucosa. Although uncommon, a search

Table 1. Specifications of the antibodies used

Antibody	Manufacturer	Clone	Antigen retrieval	Dilution	Incubation
Collagen IV	Dako	CIV22	Citrate pH6	1:25	1 hour / room temperature
Factor VIII	Dako	F8/86	Citrate pH6	1:100	1 hour / room temperature
CD34	Dako	QBEND10	Citrate pH6	1:200	1 hour / room temperature
α-SMA	Dako	1A4	Citrate pH6	1:150	1 hour / room temperature
Tryptase	Dako	AA1	Trypsin 1%	1:50	1 hour / room temperature

of the English-language literature for cases of EH involving the oral cavity identified 40 case reports, including the present case. The mean age of the patients was 37.7 years (range: 3–82 years). There was a slight predilection for young adult men (24 of 40 cases) (Table 2). In contrast, cutaneous EH more commonly affects women in the third and fourth decades of life [6]. There was a predominance of cases involving the lips (18 of 40 cases), while the cheek mucosa was affected in nine of the cases reported (Table 2). Involvement of the cheek mucosa seems to be less common and usually manifests as a nodule, features observed in the present case.

Clinical presentation of EH is nonspecific and is therefore often confused with epidermal cyst, angioma, pyogenic granuloma, Kaposi's sarcoma, salivary gland tumor, lymphoma and squamous cell carcinoma, among other less frequently cited conditions [1, 6]. In the present case, the nodular feature mimicked lipoma, an entity not yet suggested by other authors (Table 2). EH can also manifest as pruritus, macula and ulcer; however, the nodular form is the most common [1, 7, 9, 13, 16, 19, 21, 22, 23, 27].

In view of clinical similarities that EH shares with other diseases, histopathological examination is essential for establishment of the diagnosis. In the present case, the diagnosis was based on the typical histopathological features described in other studies [1, 2, 3, 6]. The morphological characteristics consisted of well-developed vessels and some immature vessels lined by aggregates of epithelioid endothelial cells associated with an exuberant component of inflammatory cells, particularly eosinophils and lymphocytes. According to Aggarwal and Keluskar [6], eosinophils typically account for 5–15% of the infiltrate, but can reach up to 50% in rare cases. Occasionally, the infiltrate is devoid of eosinophils. An interesting finding reported by Sun et al. [1] and Aggarwal and Keluskar [6] was the presence of cytoplasmic vacuoles in endothelial cells, a feature not observed in the present case. At low magnification, most of the lesions described by Sun et al. [1] also exhibited a characteristic lobular arrangement similar to the case reported here. Other features shared with the present case were the presence of lymphoid follicles, which are typical of EH, and the absence of cellular atypia [1, 2, 3]. In contrast to the seven cases reported by Sun et al. [1] which were non-encapsulated lesions, a well-defined capsule was observed in the present case. Furthermore, similar to the case reported recently by Kumar et al. [3], evidence of thrombi inside dilated vessels was found in the present patient, suggesting the occurrence of vascular damage.

Although these morphological features are well established, it is important to differentiate oral EH from other vascular tumors that also consist of epithelioid-like endothelial cells since some of these tumors exhibit a different biological behavior, prognosis and treatment. The differential diagnosis of EH includes epithelioid angiosarcoma, epithelioid hemangioendothelioma, hobnail hemangioma, and epithelioid angiomatous nodule [1, 4, 5]. Epithelioid angiosarcoma is characterized by an infiltrative and destructive growth pattern and is composed of highly pleomorphic cells, in addition to the presence of atypical mitotic figures and areas of necrosis [5]. Epithelioid hemangioendothelioma also exhibits an infiltrative growth pattern in the form of cords and nests of spindle-shaped, discretely pleomorphic endothelial cells amidst a hyaline and myxoid stroma [6]. However, well-defined vascular canals are rarely found [6]. In contrast to EH, epithelioid hemangioendothelioma is associated with high rates of morbidity and mortality due to recurrence and regional metastasis [5]. Hobnail hemangioma is characterized by a biphasic growth pattern consisting of well-defined dilated vessels lined by prominent "hobnailed" endothelial cells and neoplastic vessels in the deep portion of the lesion. This tumor is extremely rare in the oral mucosa [5]. Finally, epithelioid angiomatous nodule shares many clinical and histopathological features with EH; however, this lesion seems to be confined to the dermis [5].

EH should also be differentiated from Kimura's disease and bacillary angiomatosis. Kimura's disease also exhibits a prominent inflammatory infiltrates associated with blood vessels and lymphoid follicles with germinal centers, but aggregates of epithelioid endothelial cells with abundant eosinophilic cytoplasm are absent [5, 27]. Moreover, Kimura's disease manifests as nodules or deep tumors accompanied by lymphadenopathy and marked peripheral eosinophilia, representing a chronic inflammatory process and not originating from proliferation of endothelial cells [5, 6]. There has been extensive debate whether EH is a variant of Kimura's disease. However, today it is accepted that the two diseases are independent and represent separate entities with distinct clinical and histopathological features [6]. Bacillary angiomatosis usually develops in immunodeficient patients and is mainly caused by infection with *Bartonella henselae*. Microscopic analysis shows abundant neutrophils associated with clusters of bacteria [5].

Although some peculiar morphological features help recognize EH and exclude other vascular lesions, immunohistochemical markers can be used to confirm the vascular nature of epithelioid-like cells or even to better characterize the lesion. These epithelioid endothelial cells are positive for endothelial cell markers such as factor VIII and CD34, a fact observed in the present study. Similarly,

Sun et al. [1] detected less prominent staining for CD34 compared to factor VIII. Immunostaining for α-SMA was useful in demonstrating the myopericytic layer around immature blood vessels. A similar immunoreactivity pattern for these markers has been observed in other studies [1, 2]. In the present case, collagen IV staining permitted identification of small immature vessels with indistinguishable or inconspicuous lumina. Positive immunostaining for mast cells confirmed the presence of these cells, which have also been detected in EH by other authors [1, 3].

The pathogenesis of EH continues uncertain and two strong hypothesis have been proposed, which consider it to be a reactive lesion or true neoplasm. The features that support the reactive condition would be the preference of EH for the surface of soft tissues, the presence of central areas containing epithelioid endothelial cells in an

Table 2. Summary of clinical data of previously reported EH in oral mucosa

Age (years)	Sex	Lesion	Location	Preoperative diagnosis	Reference
37	Female	Nodule	Lower lip	Kaposi's sarcoma	Rosai and Akerman [8]
28	Female	Macule	Palate	Lymphoma	Saxe and Kahn [9]
22	Male	Nodules	Upper and lower lip	-	Dickens [10]
28	Male	Nodule	Upper lip	Salivary gland adenoma	Buckerffeld and Edwards [11]
46	Male	Nodule	Upper lip	-	Eveson and Lucas [12]
13	Male	Pruritus	Lower lip	-	Buchner et al. [13]
28	Male	Nodules	Oral mucosa	Pyogenic granuloma	Massa et al. [14]
32	Male	Nodule	Upper lip	-	Peters et al. [15]
31	Male	Macule	Togue	-	Iguchi et al. [16]
25	Female	Nodule	Palate	-	Moran et al. [17]
42	Female	Nodule	Buccal mucosa	-	Kabani et al. [18]
82	Male	Ulcer	Togue	Squamous cell carcinoma	Razquin et al. [19]
48	Male	Nodule	Togue	-	Artazkoz et al. [20]
55	Male	Ulcer	Lower lip	Squamous cell carcinoma	Lopez and Battaglino [21]
17	Male	Nodule	Buccal mucosa	Pyogenic granuloma	Toeg et al. [22]
12	Male	Ulcer	Buccal mucosa	-	Toeg et al. [22]
65	Male	Nodule	Upper lip	EH	Renshaw and Rosai [23]
43	Female	Nodule	Upper lip	EH	Renshaw and Rosai [23]
3	Male	Nodule	Upper lip	EH	Renshaw and Rosai [23]
34	Female	Pruritus	Lower lip	EH	Renshaw and Rosai [23]
59	Male	Nodule	Buccal mucosa	-	Misselevich et al. [24]
30	Female	Nodule	Upper lip	-	Bartralot et al. [25]
27	Female	Nodule	Buccal mucosa	-	Martin-Granizor et al. [26]
54	Male	Nodule	Lower lip	-	Martin-Granizor et al. [26]
43	Female	Nodule	Togue	-	Martin-Granizor et al. [26]
30	Female	Nodule	Upper lip	-	Mariatos et al. [4]
23	Male	Ulcer	Togue	Malignant tumor	Shimoyama et al. [27]
60	Male	Nodule	Buccal mucosa	-	Tsuboi et al. [28]
56	Male	Ulcer	Togue	-	Park et al. [7]
42	Male	Nodule	Palate	Salivary gland adenoma	Sun et al. [1]
48	Male	Nodule	Togue	Vascular tumor	Sun et al. [1]
52	Male	Nodules	Togue	Vascular tumor	Sun et al. [1]
65	Female	Nodule	Togue	Pyogenic granuloma	Sun et al. [1]
31	Female	Nodule	Upper lip	Vascular tumor	Sun et al. [1]
8	Male	Nodule	Lower lip	Pyogenic granuloma	Sun et al. [1]
32	Female	Ulcer	Togue	Squamous cell carcinoma	Sun et al. [1]
65	Female	Nodule	Lower lip	-	Miteva et al. [2]
25	Female	Nodule	Buccal mucosa, upper and lower lip	Atypical granuloma	Aggarwal and Keluskar [6]
30	Female	Nodule	Gingiva	-	Kumar et al. [3]
52	Male	Nodule	Buccal mucosa	Lipoma	Present

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indeterminate pattern, peripheral vessels surrounded by the proliferation of epithelioid cells, and vascular damage characterized by fragmentation of the elastic lamina, fibrointimal proliferation and rupture of the muscle wall [29]. From this perspective, some authors believe that local trauma may trigger cell proliferation of the reactive condition [3, 26]. On the other hand, the invasive growth of EH observed in some cases and possible recurrence suggest a true neoplastic process [1, 3, 4].

Sun et al. [29] suggested local hypoxia mediated by congenital vascular malformation or trauma to play an important role in the pathogenesis of EH. Hypoxia would be responsible for proliferation of endothelial cells, especially by promoting synthesis of vascular endothelial growth factor (VEGF) that results in formation of a vascular tumor. Additionally, inflammatory cells, such as eosinophils and mast cells, contribute to proliferation of endothelial cells in EH. Hypoxia has been shown to stimulate degranulation of mast cells and eosinophils, increasing the expression of hypoxia-inducible factor 1 (HIF-1) and VEGF and thus inducing the proliferation of endothelial cells [30].

Some features observed in the present case, such as evidence of vascular damage including thrombosis and fibrointimal proliferation, seem to support the reactive condition. However, the positive staining of epithelioid endothelial cells, which lined the vessels or formed aggregates, for the vascular markers cited above favors the neoplastic nature of this lesion. Another finding that complements and supports the neoplastic nature of the present case is the absence of elastic fibers, demonstrated by staining with Weigert's orcein amidst epithelioid endothelial cell proliferations, which rules out the hypothesis that the proliferation of these cells would have been triggered by vascular damage. In the present case, elastic fibers were only found surrounding the wall of intact blood vessels.

Considering the possible neoplastic nature of the lesion, according to Miteva et al. [2] it remains unclear whether EH arises from the exclusive proliferation of endothelial cells derived from blood vessels or, in fact, exhibits a biphasic pattern, also involving proliferation of endothelial cells derived from lymphatic vessels. The authors therefore performed an immunohistochemical analysis using D2-40 and observed an increase in the number of lymphatic vessels inside the tumor. On the basis of these results, the authors suggested that the lymphoid component of EH may result from lymphangiogenesis.

Treatment of EH is often a challenge. Intralesional corticosteroids, cryotherapy, laser cauterization and irradiation have been used, but are not very effective. Surgical excision and follow-up continue to be the most recommended treatment [3, 4, 6]. Most patients with oral EH (Table 2) were submitted to complete surgical resection (37 of 40 cases) without additional therapy, and recurrence was relatively rare, reported in only five studies published in the literature [3, 8, 9, 14, 20]. Local recurrence is common in one third of patients with cutaneous EH, but is rare in the oral mucosa [1, 3]. The present case is consistent with the literature since the lesion was treated by complete surgical excision, showing no signs of recurrence so far. As expected, none of the cases of oral EH was associated with metastases.

EH in the oral mucosa is uncommon and the clinical and histopathological differential diagnosis with other lesions is necessary, including non-neoplastic proliferative processes or neoplasms of vascular, lymphoid, glandular and epithelial origin. For this purpose, careful morphological analysis should be performed based on the peculiarities of each entity. Immunohistochemical analysis may be used in some cases.

NOTE

The abstract of the paper has been published in Oral Surgery, Oral Medicine, Oral Pathology, and Oral Radiology (Vidal MTA, Gurgel CA, Leitão ÁCGH, Ramos EAG, Xavier FCA, Ramalho LMP, Santos JN. Epithelioid Hemangioma Located in The Buccal Mucosa: Case Report. Oral Surg Oral Med Oral Pathol Oral Radiol. 2014; 117(2):e155).

Written informed consent for publication of this case report and any accompanying images was obtained from the patient.

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Епителиоидни хемангиом слузокоже уста – приказ болесника

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КРАТАК САДРЖАЈ

Увод Епителиоидни хемангиом је ретка бенигна вазопролиферативна неоплазма која се обично испољава код одраслих особа средњег животног доба у виду црвених израслина.

Приказ болесника Болесник стар 52 године јавио се са квржастом лезијом пречника 3 *cm* с леве стране слузокоже уста, насталом годину дана раније. Клиничка претпоставка била је да се ради о липому. Ексцизиона бипсија показала је омеђену лезију сачињену од режњева судова са слабо приметним или малим луменом, који се доводе у везу са израженим инфламаторним инфилтратом сачињеним од еозинофила, хистиоцита и хроничних инфламаторних ћелија. Ендотелијске ћелије које су чиниле лезију имале су епителиоидну морфологију и садржале су веће количине еозинофилне цитоплазме. Извршена је имунохистхемијска анализа за *CD*34, фактор VIII, колаген VI, *α*-актин глатких мишића и мастоците, као и хистохемијско бојење орцеином Вајгертовом методом.

Закључак Разнородна и морфолошки сложена група лезија тешка за дијагностиковање сачињава васкуларне пролиферације меког ткива. Овај приказ представља случај оралног епителиоидног хемангиома, при чему је акценат на релевантним морфолошким и имунохистохемијским особинама које могу помоћи да се ово стање диференцира од других неоплазми.

Кључне речи: ангиолимфоидна хиперплазија са еозинофилијом; епителиоидни хемангиом; слузокожа уста

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