CASE REPORT

Calibre Persistent Labial Artery: Clinical Features and Immunohistochemistry Diagnosis

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Abstract Calibre persistent labial artery (CPLA) usually presents as an asymptomatic papule on the lower lip and can be easily misdiagnosed as a mucocele, haemangioma, venous lake, varix or fibroma. When it is ulcerated, squamous cell carcinoma is the most usual differential diagnosis. Here, we report a case of a 25-year-old woman with no previous relevant medical history who presented with a complaint of an asymptomatic, non-ulcerated, progressively growing nodule (over the last 5 months) on the upper lip. In this case, the diagnosis was made clinically and confirmed by immunohistochemical analysis. We conclude that clinicians should be aware of CPLA and it should be included in the differential diagnosis of labial mucosal papules. Sometimes, the immunohistochemical analysis is necessary to make a correct diagnosis.

Keywords Calibre-persistent labial artery · Mucocele · Atherosclerosis · Immunohistochemical analysis · Labial artery

Introduction

Voth first used the term “calibre persistent artery” in 1962 to describe abnormal vessels in the stomach wall [1].

Calibre persistent artery specifically of the lip was first described by Miko et al. in 1980 [2]. The authors described

arteries with a diameter larger than normal, near a mucosal or external surface, which were similar to the lesions described in the gastrointestinal tract [2, 3].

Calibre persistent labial artery (CPLA) is a vascular anomaly in which a vessel penetrates into the submucosal tissue with a larger diameter and continues without division or loss of calibre [4].

Although rarely reported to date, the incidence of CPLA is probably far greater than suggested and many healthcare providers, particularly dermatologists, oral pathologists and dentists, meet this condition. However, detailed diagnostic data are lacking in the literature.

To date, there have been 17 cases of CPLA reported in the literature [2–9].

We report this case to supplement the clinical information available, and draw attention to the importance of immunohistochemistry in order to diagnose, without doubt, this clinical condition that can lead to problems of differential diagnosis with chronic ulcer, mucocele, and squamous cell carcinoma of the lower lip.

Case Report

A 25-year-old woman with no previous relevant medical history presented with the complaint of an asymptomatic, non-ulcerated, progressively growing nodule (over the last 5 months) on the upper lip. She complained mainly aesthetic discomfort that it entailed and interference with chewing functions; the swelling had suddenly grown recently.

Physical examination of the upper lip showed an elevated, soft, 5 mm in diameter, serpiginous lesion under normal-appearing mucosa of the oral cavity that emerged on the left side of the upper lip as a nodule (Fig. 1). On

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Fig. 1 Clinical examination: linear elevated area on the upper labial mucosa

Fig. 2 The large labial artery that was detected during the procedure

palpation it was fluid-filled, slightly painful and no pulsation was noted.

Excision of the neoformation was done under local anesthesia (mepivacaine and epinephrine 1:200,000). During the operation, just after the lesion was excised, profuse and pulsating bleeding was observed. A large labial artery was detected and successfully ligated with a suture with no. 4-0 monocryl sutures. The excised lesion was sent for histopathological examination (Fig. 2).

Analgesic (ibuprofen 400 mg) and antibiotics (amoxicillin 1,000 mg, twice a day) were prescribed to be taken for 1 week.

At follow-up 1 month post-operatively the upper lip was completely asymptomatic and no pulsation was felt in the region.

Histopathological examination showed that the surface of the specimen was covered by intact stratified squamous epithelium. Taking up much of the submucosa was a large artery with marked intimal thickening and focal atheromatous plaques. Flap of mucosa and submucosa site of a

large artery with marked intimal thickening, many perilesional vessels and blood extravasation (Fig. 3). The internal elastic lamina (IEL) was disrupted in one region and the media was thinned in this area. Adjacent to the artery was normal fibrous connective tissue with fat and small vessels and nerves. There was no evidence of vasculitis, malignancy or other features.

Antibody used for immunohistochemical analysis was Monoclonal CD34, dilution 1/16 (Immunotech; Marseilles, France) (Fig. 4). The strongest expression of CD34 was found in endothelial cells (EC) of capillaries, followed by arteries, veins, arterioles, and venules.

Discussion

A calibre persistent labial artery (CPLA) is an artery in the submucosal tissue with a diameter that is larger than normal. It is usually visibly or palpably pulsatile. Thus, correct diagnosis can easily be made clinically if the lesion presents with characteristic features. However, it can sometimes occur as an asymptomatic papule without evident pulsation and in this case poses problems of differential diagnosis with various other conditions: mucocele, varicosities, venous lakes, hemangiomas and vascular malformations. CPLA can be easily differentiated from varicose veins and venous lakes, when examined histologically by comparing the architecture of vessels affected, particularly the thickness of the muscle wall. Other differential diagnoses include mucocele, hemangiomas (benign vascular tumors with proliferation of endothelial cells) and vascular malformations (structural vascular abnormalities without endothelial proliferation, as simple capillary malformation,

Fig. 3 Flap of mucosa and submucosa. It highlights major artery with thickened walls with stenotic lumen and numerous vessels around the wound and widespread extravasation (hematoxylin-eosin stain, original magnification $\times 100$)

Fig. 4 Immunohistochemical staining for CD34 (original magnification $\times 200$)

arteriovenous malformation) that require support for further investigation [10, 11].

For this reason we consider the immunohistochemical analysis a further support for the differential diagnosis from other similar lesions to the CPLA.

To completely resolve any doubt is immunohistochemical analysis was done using monoclonal antibodies to the CD34.

The monoclonal antibody, CD34 is a 110-kDa transmembrane glycoprotein present on leukemic cells, EC, and stem cells. In addition, it is localized on cells of the splenic marginal zone, dendritic interstitial cells around vessels, nerves, hair follicles, muscle bundles, and sweat glands in a variety of tissues and organs. Its function is still unclear. It is used for leukemia diagnosis and subclassification and for diagnosis of vascular tumors. Antibodies to CD34 also strongly label gastrointestinal stromal tumors, and the antigen is invariably found in solitary fibrous tumor and dermatofibrosarcoma protuberans [12, 13].

We conclude that clinicians should be aware of CPLA and it should be included in the differential diagnosis of

labial mucosal papules. Sometimes, the immunohistochemical analysis is necessary to make a correct diagnosis.

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