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Intravascular papillary endothelial hyperplasia in the lower lip: A case report

Hiperplasia endotelial papilar intravascular en el labio inferior: Reporte de caso

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ABSTRACT: Intravascular papillary endothelial hyperplasia or Masson's tumor is a benign, non-neoplastic lesion that can affect the torax, fingers, head, and neck. Its presence in the oral cavity is unusual; however, there are reports in the literature. A female patient in the 4th decade of life who attended the Oral and Maxillofacial Surgery Department of the Universidad Central de Venezuela, with a purplish lesion on the lower lip of two months of evolution associated with masticatory trauma is presented. An excisional biopsy was performed, and a histopathological study was performed. The wall of blood vessels made up of endothelial cells showing intraluminal papillary growth was observed. The papillary structures are lined by hyperplastic endothelial cells in the vascular lumen. The definitive diagnosis was intravascular papillary endothelial hyperplasia. Due to the great resemblance to other vascular lesions, it is essential to perform a histopathological study of suspicious lesions to rule them out.

KEY WORDS: Endothelial hyperplasia, Masson's tumor, benign oral lesions.

INTRODUCTION

Intravascular Papillary Endothelial Hyperplasia (IPEH), also known as Masson's tumor or intravascular vegetative hemangioendothelioma, is an unusual benign, non-neoplastic vascular lesion (Milhan *et al*., 2018). Which was described for the first time in 1923 by the pathologist Frances Pierre Masson (Cohen *et al*., 2009; Garcia-Guliarte *et al*., 2009; Milhan *et al*., 2018; Voruz *et al*., 2020) in a 68-year-old male patient with a painful, ulcerated hemorrhoid that could not be reduced. The hemorrhoid was excised and it was determined that histologically it was characterized by an atypical papillary endothelial proliferation that mimicked angiosarcoma (Tosios *et al*., 1994; Steffen, 2001; Milhan *et al*., 2018). IPEH was named again by Clearkin and Enzinger in 1976, as a benign tumor that arises in the oral region and other regions, where it is established that the entity is a reactive phenomenon rather than a true neoplasm (Yonezawa *et al*., 2009; Vieira *et al*., 2021), defining it as a reactive benign lesion of vascular origin that is produced by an excessive proliferation of endothelial cells (Cohen *et al*., 2009).

IPEH represents approximately 2% of all vascular tumors of the skin and subcutaneous tissue, presenting as predominant regions: trunk, fingers, head and neck (Cohen *et al*., 2009; Vieira *et al*., 2020). Tosios *et al*. (1994) point out that this lesion can occur in any part of the body, being more common in the head and neck region. In turn, Milhan *et al*. (2018) and Cohen *et al*. (2009) state that the most common sites of appearance in the oral cavity are: lips, buccal mucosa, and tongue. Its age range in terms of its appearance is wide; however, it is considered to be more prevalent in the fifth decade of life and is more frequent in women (Cohen *et al*., 2009).

The lesion can arise in normal blood vessels and be called the "pure form", or in a pre-existing vascular lesion, called the "mixed form" (Tosios *et al*., 1994). It may or may

not be associated with other lesions such as hemangiomas, pyogenic granulomas or lymphangiomas. IPEH in the oral area is relatively rare and usually presents as a skin lesion (Matsuzaka *et al*., 2003).

Clinically, a firm, raised and demarcated nodular lesion or mass is observed, slow growing and violaceous in color (Eguchi *et al*., 2020) blue or reddish approximately 2 centimeters. Histologically, it presents as pure or mixed in appearance, with the presence of confined papillary structures attached to the walls between the vascular spaces. These papillae are lined by enlarged endothelial cells without polymorphism, mitotic activity, or necrosis. There is also presence of thrombi of different sizes and organizations between the lesions (Cohen *et al*., 2009).

Regarding its treatment, it can be simple surgical excision if it is a primary IPEH; in the case of secondary IPEH, it should be treated as a vascular lesion. Its prognosis is good and recurrence occurs in its mixed forms (Tosios *et al*., 1994). The objective of this clinical case is to describe a nodular lesion on the lower right lip in a 45-year-old female patient.

CASE REPORT

This is a 45-year-old female patient, who attended the Bucomaxillofacial Surgery Department, Faculty of Dentistry of the Universidad Central de Venezuela in October 2022, for assessment, diagnosis, and treatment due to presenting volume increase in the lower right lip, where he presented an increase in volume due to involuntary bite-type trauma during the chewing act. After this, the patient refers change in color to purplish red and a progressive increase in size with 1 year of evolution up to the present. She denies a contributing systemic disease history, denies medication associatedwith the reason for consultation.

Extraoral clinical examination revealed an increase in volume in the lower right labial area (Fig. 1A and 1B). The intraoral clinical examination revealed a nodular lesion in the lower right lip involving dry lip and wet lip up to the labial commissure, approximately 2 cm x 2 cm in diameter. Violet-colored, oval-shaped lesion with a smooth surface, soft consistency, adherent implantation, asymptomatic and primary presentation.

A vitro-pressure test is performed in which a positive result is obtained. In addition, the possible differential diagnoses (mucocele, vascular lesion) and her treatment were explained to the patient, according to what was compiled in the anamnesis. Likewise, the authors presented a written informed consent to the patient where the nature of the treatment was explained and authorization was obtained for the photographic record and scientific publication of the case. An excisional biopsy was performed under perilesional infiltrative local anesthesia with 2% lidocaine, approached through an elliptical wedge-shaped incision (Fig. 2A), Hilton maneuver to debride the lesion, excision of the lesion (Fig. 2B), and tissue synthesis with 000 black silk suture (Fig. 3A). The sample fixed in 10% formaldehyde was sent for histopathological study (Fig. 2C).

Under the microscope, a non-encapsulated benign lesion made up of numerous blood vessels was observed. In some vessels, the wall made up of endothelial cells showing intraluminal papillary growth is observed. The papillary structures are lined by hyperplastic endothelial cells in the vascular lumen. Presence of prominent intravascular fibrin thrombi and extensive areas of hemorrhage. There is no evidence of malignancy in the sections studied (Fig. 2D). The diagnostic conclusion was obtained: Intravascular Papillary Endothelial Hyperplasia (Masson's Tumor). Control appointments were made at 7 days (Fig. 3B) and 3 months (Fig. 3C).

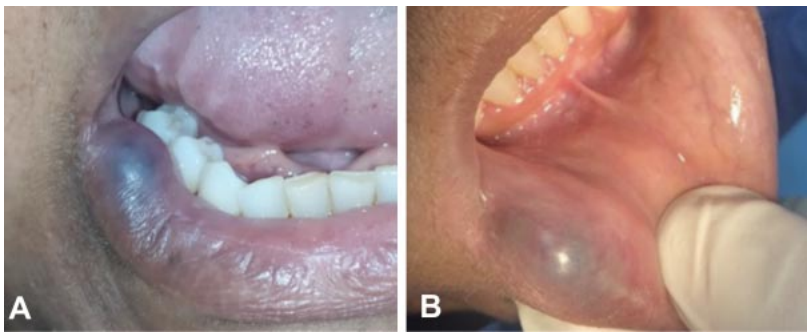

Fig. 1. Clinical photograph of the lesion (A and B).

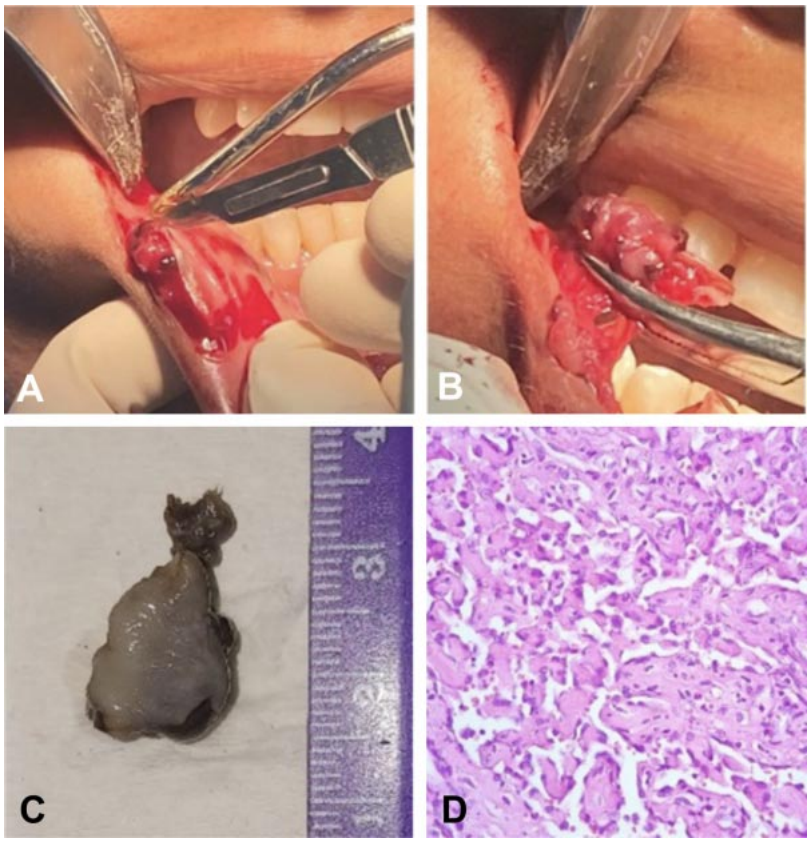

Fig. 2. Procedure for taking a biopsy sample (A and B), macrophotography of the lesion (C), and microphotography of the lesion at 10X magnification with H-E staining (D).

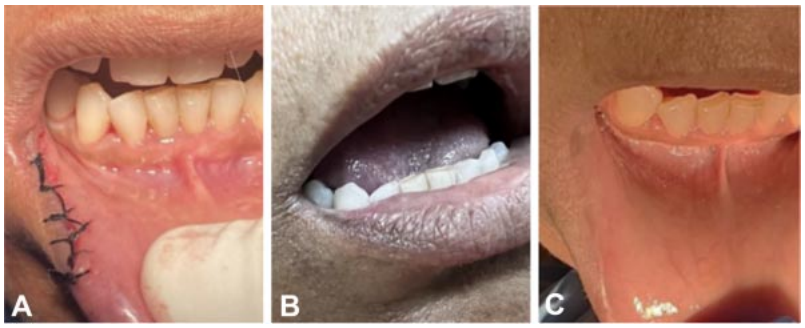

Fig. 3. Immediate postoperative control (A), control seven days (B) and three months (B) after surgery.

DISCUSSION

IPEH represents 2 % of all vascular tumors of the skin and subcutaneous tissue, in the oral cavity it is described as a rare lesion (Cohen *et al*., 2009; Garcia-Guliarte *et al*., 2009; Milhan *et al*., 2018; Voruz *et al*., 2020). As exposed by Vieira *et al*. (2021), there are only 20 reported cases in the literature to 2020, which remarks the importance of the present case. According to its epidemiology, it can present in the fifth decade of life and in female patients, very similar to what was presented previously, since the patient is in the 4th decade of life (Cohen *et al*., 2009); and, in turn, in contrast to the case presented by Murugaraj *et al*. (2010) of a 14-year-old female patient. There's also differences reported by de Courten *et al*. (1999) which in their six cases report, most of the patients were males in their 50's.

As pointed out by different authors (Tosios *et al*., 1994; Matsuzaka *et al*., 2003; Cohen *et al*., 2009; Inoue *et al*., 2011; Milhan *et al*., 2018), its location, although infrequent, can occur in the head and neck region; specifically in the oral cavity, it can be found in the lower lip as in the present case similar to the findings of de Courten *et al*. (1999), Chen *et al*. (2018) and Sarode & Sarode (2015) and in contrast to Matsuzaka *et al*. (2003) where it appeared on the upper lip, Voruz *et al*. (2022) who described three cases of this lesion in the nostrils, Milhan *et al*. (2018) where the lesion occurred in the anterolingual region, Murugaraj *et al*. (2010) in buccal mucosa and Eguchi *et al*. (2020), Luigi *et al*. (2022), Mirmohammadsadeghi *et al*. (2019) where it presented in the mandibular region and Cho *et al*. (2021) in the submandibular gland.

Its diagnosis is usually difficult due to its similarity with other lesions (Matsuzaka *et al*., 2003; Cohen *et al*., 2009; García-Guilarte *et al*., 2009; Milhan *et al*., 2018) since both clinically and histologically it has a great resemblance to other lesions of vascular origin. Tosios *et al*. (1994) studied 18 lesions where hemangiomas, venous trauma, mucoceles, pyogenic granulomas, and nevus were clinically presumptive diagnoses.

In the present case, vitro-pressure was used during the clinical diagnosis, which was positive. In the report by Murugaraj *et al*. (2010) also performed this technique but their result was negative.

Clinically, the lesion presented is observed as an increase in volume in the lower lip on the right side, with a nodular appearance, involving both dry and wet lips, of a purplish color, smooth surface and soft consistency, as reported by Cohen *et al*. (2009).

Histologically, endothelial cells were observed in some vessels on their walls with intraluminal papillary growth, which were lined by hyperplastic endothelial cells and the presence of intravascular fibrin thrombi, similar to what was found by different authors (Tosios *et al*., 1994; Cohen *et al*., 2009; Inoue *et al*., 2011; Milhan *et al*., 2018; Eguchi *et al*., 2020). Inoue *et al*. (2011) pointed that immunohistochemical agents like antibodies against CD34, vimentin, factor VIII antigen, a-smooth muscle actin (a-SMA), podoplanin, CD105, and ki-67 antigen can be used for its diagnosis.

Surgical resection is the treatment of choice for these cases (Cohen *et al*., 2009; Sarode & Sarode, 2015; García-Guilarte *et al*., 2018; Eguchi *et al*., 2020). In the report presented, the lesion was completely removed, and tissue debridement was performed, in addition to suturing.

CONCLUSION

IPEH or Masson's tumor is a very rare benign lesion of vascular origin. Its correct diagnosis is important to differentiate it from angiosarcomas, for which it is essential to perform a biopsy on suspicious lesions to rule it out and thus ensure correct management of the lesion.

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Conflict of interest: The authors declare that they have no conflicts of interest.

Ethical approval: The study was conducted in accordance with the Declaration of Helsinki. Informed consent was obtained from the subject involved in the study.

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RESUMEN: La hiperplasia endotelial papilar intravascular o tumor de Masson es una lesión benigna no neoplásica que puede afectar el tórax, los dedos, la cabeza y el cuello. Su presencia en la cavidad bucal es inusual; sin embargo, hay informes en la literatura. Se presenta una paciente femenina de la 4ta década de la vida que acude al Departamento de Cirugía Oral y Maxilofacial de la Universidad Central de Venezuela, con una lesión violácea en el labio inferior de dos meses de evolución asociada a un traumatismo masticatorio. Se realizó biopsia excisional y estudio histopatológico. Se observó la pared de vasos sanguíneos formada por células endoteliales que muestran crecimiento papilar intraluminal. Las estructuras papilares están revestidas por células endoteliales hiperplásicas en la luz vascular. El diagnóstico definitivo fue hiperplasia endotelial papilar intravascular. Debido al gran parecido con otras lesiones vasculares, es imprescindible realizar un estudio histopatológico de las lesiones sospechosas para descartarlas.

PALABRAS CLAVE: Hiperplasia endotelial, tumor de Masson, lesiones orales benignas.

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