

## Short Case Report

# Intravascular papillary endothelial hyperplasia: a case report

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**Abstract – Case report:** A 71-year-old patient, with no notable medical-surgical history, was referred for a specialized consultation with Oral Mucosal Pathology for tumefaction on the dorsal side of the tongue. It was a nodular lesion measuring one centimeter long, purplish, and painless. There was nothing found through palpation. Surgical excision with a laser diode was performed under local anesthesia. The postoperative follow-up was simple. The patient was seen 3 months later without any sign of recurrence. Histopathological analysis revealed a vegetative intravascular hemangioendothelioma. **Comments:** This case is rare because of the location of the tumor, and moreover because the subject is male. A laser diode was chosen to resect the lesion because of the purple color which could have resulted in a possible vascular lesion. Indeed, the limits of excision were coagulated, which facilitated the surgery time and any limited postoperative hemorrhagic complications that could have occurred.

## Clinical observation

A 71-year-old patient was referred to the Specialist Consultation for Oral Mucosal Pathology for the diagnosis and management of dorsal swelling on the tongue. His dentist had incidentally discovered the swelling a month earlier. During the interview, the patient did not report any change in size since the discovery of the lesion.

The patient was in good health and his medical history was not a contributing factor.

A clinical extraoral examination revealed no cervicofacial adenopathy. An intraoral clinical examination revealed a well-defined 1-cm swelling covered by a smooth purplish mucosa centered on the dorsal surface of the tongue (Fig. 1). The swelling was firm, non-fluctuating, and of a rubbery consistency with no color change when pressure was applied. No neurological disorder was associated with it.

Following clinical examination, several diagnostic hypotheses were proposed: a vascular malformation, a tumor of benign mesenchymal origin such as a fibroma, a neurofibroma, an Abrikossoff tumor, or an aggressive malignant neoplasia, such as angiosarcoma.

Surgical excision of the lesion was performed using a laser diode under local anesthesia, exposing the underlying glottic muscles.

The surgical procedure was without complications. One month after the operation, the mucosa had healed well (Fig. 2).

The anatomopathological examination of the operative specimen showed that it was coated with a multilayered, squamous, slightly hyperplastic keratinized epithelium. In the underlying capsule between the striated skeletal muscle cells, several dystrophic vessel-wall formations, including a Masson tumor and a nodular intraluminal fibrous formation, probably related to an old thrombus, were observed (Fig. 3). The histological diagnosis was an old thrombus and a benign Masson tumor.

## Comments

In 1923, Masson described the vegetative intravascular hemangioendothelioma [1] as a benign tumor pathology secondary to the reactive proliferation of endothelial cells with thrombus-related papillary formations [2]. Since its initial description it has been referred to as intravascular papillary endothelial hyperplasia (IPEH), or Masson tumor.

It is a benign pathology appearing in the form of a generally solitary nodule located in the deep dermis and the hypodermis. It represents 2% vascular tumors affecting the cutaneous and subcutaneous tissues. Although this lesion can be found in the cervicofacial region, its intraoral location is rarely reported in the literature [3].

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The histological formation of IPEH is still a topic for debate. However, three different types of IPEH have been reported: a primary (pure) form where a neoplasia is observed in distended vessels; a secondary (mixed) form that appears within pre-existing lesions such as hemangiomas, pyogenic granulomas, or lymphangiomas; and a rare form with an extravascular localization occurring in a hematoma. It has been observed that the great majority of lesions, whatever their type, are associated with a thrombus. The primary form is the most frequently encountered form [4].

Oral forms of IPEH seem to be more common in women, according to Pins *et al.*, the sex ratio F/M is 1.2:1 [5]. The most affected sites in the oral cavity are the cheeks and the lips. The general appearance of this lesion when it is present in the mouth is that of a centimeter-wide discreetly purplish nodule.

Thus, it can be confused with other benign lesions or vascular malformations because of its smooth and regular nodular appearance and its low prevalence in the oral cavity. No malignant transformation of IPEH has been reported. However, some Masson tumors can grow to large sizes and be confused histologically with an angiosarcoma, whose treatment is more aggressive because of its malignant nature [6].

It is essential to rule out the differential diagnosis of angiosarcoma to avoid unnecessarily invasive surgery. The management of IPEH is based on its removal. This tumor has a low rate of recurrence [7].

The reported case is rare because of the location of the tumor and because the subject is male. The choice to remove the tumor with a laser diode was based on the purplish hue of the lesion, which suggested a possible vascular lesion. This technique ensures that incised tissue is already coagulated, which decreases the operating time and limits postoperative bleeding complications.

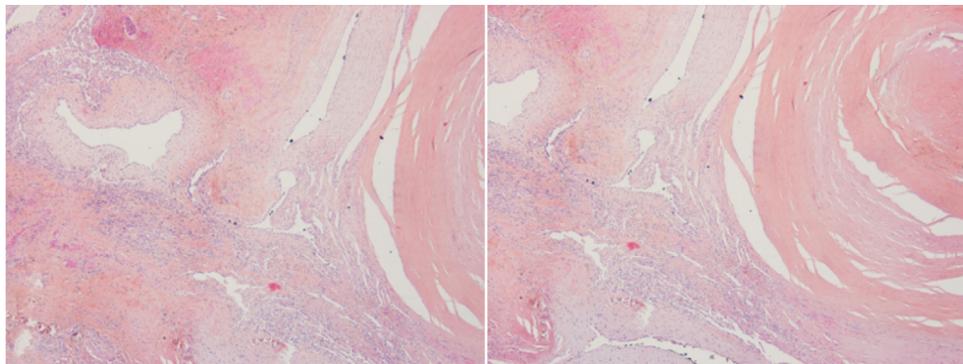
**Conflicts of interest:** The authors declare that they have no conflicts of interest in relation to this article.



**Fig. 1.** Lingual tumor with a purplish appearance during the initial consultation at the Univeristy Hospital Center of Bordeaux.



**Fig. 2.** Healing on the tongue, 1 month after the surgery.



**Fig. 3.** Distended blood vessel with intravascular fibrous formation. Hematoxylin–eosin staining; color image; enlarged  $\times 20$ .

## References

1. Masson P. Hemangioendotheliome végétant intravasculaire. *Bull Soc Anat Paris* 1923;93:517–523.
2. Mahapatra QS, Sahai K, Malik A, Mani NS. Intravascular papillary endothelial hyperplasia: an unusual histopathological entity. *Indian Dermatol Online J* 2015;6:277–279.
3. Korkolis DP, Papaevangelou M, Koulaxouzidis G, Zirganos N, Psychogiou H, Vassilopoulos PP. Intravascular papillary endothelial hyperplasia (Masson's hemangioma) presenting as a soft-tissue sarcoma. *Anticancer Res* 2005;25:1409–1412.
4. Bologna-Molina R, Amezcua-Rosas G, Guardado-Luevanos I, Mendoza-Roaf PL, González-Montemayor T, Molina-Frechero N. Intravascular papillary endothelial hyperplasia (Masson's tumor) of the mouth—a case report. *Case Rep Dermatol* 2010;2:22–26.
5. Pins MR, Rosenthal DI, Springfield DS, Rosenberg AE. Florid extravascular papillary endothelial hyperplasia (Masson's pseudoangiosarcoma) presenting as a soft-tissue sarcoma. *Arch Pathol Lab Med* 1993;117:259–263.
6. Patel PB, Kuan EC, Peng KA, Yoo F, Nelson SD, Abemayor E. Angiosarcoma of the tongue: a case series and literature review. *Am J Otolaryngol* 2017;38:475–478.
7. Akdur NC, Donmez M, Gozel S, Ustun H, Hucumenoglu S. Intravascular papillary endothelial hyperplasia: histomorphological and immunohistochemical features. *Diagn Pathol* 2013;8:167.