

CASE REPORT

Masquerading dacryocystitis

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Abstract

Intravascular papillary endothelial hyperplasia (IPEH), otherwise known as Masson's tumor, is a benign lesion of the skin with no known malignant transformations reported. It is considered a non-neoplastic lesion, as it is reactive to some vascular insult, usually a thrombus in a vein leading to vascular endothelial cell proliferation. Masson's tumor typically presents as a slow-growing, indolent lesion commonly affecting the head, neck and arms with a female predominance. It comprises ~2% of all malignant and non-malignant vascular tumors of the skin and subcutaneous tissue. We are presenting the first case of Masson's tumor of the right medial canthal subcutaneous tissue, masquerading as dacryocystitis.

INTRODUCTION

Intravascular papillary endothelial hyperplasia, or Masson's tumor, is a reactive lesion with several histological variants. The primary form of the tumor is a non-neoplastic lesion involving a vascular insult to a normal vessel (usually a vein) and thrombus formation [2, 1]. It is thought that, in the typical disease etiology, some vascular insult forms a thrombus in a vein, the thrombus and vascular stasis attracts macrophages and the macrophages release basic endothelial fibroblast growth factor leading to the proliferation of vascular endothelial cells and formation of the Masson's tumor [4, 5].

Microscopically, Masson's tumor is primarily characterized as a papillary structure projecting into the vascular lumen covered in hyperplastic endothelial cells. There should be an absence of mitotic figures, cellular pleomorphism and necrosis [6]. A distinguishing factor of Masson's tumor is that it does not express CD 105, differentiating it from well-differentiated primary vascular neoplasms [6].

Masson's tumor presents as an indolent, slowly growing reddish-purple soft tissue mass. It is typically asymptomatic, and there have been no reports of malignant transformation [7]. The standard treatment is complete surgical excision; because of its benign nature, there is no indication for excision with

wide surgical margins. If all the tissue is not excised, however, Masson's tumor may recur [8].

Masson's tumor most commonly occurs on the head, neck and arms but is rarely reported involving the ocular adnexa. In fact, this is the first time in the literature that it has ever been reported occurring in the medial canthal subcutaneous tissue.

Case report

A 59-year-old female presented to her referring ophthalmologist with a 7-month history of a right medial lower eyelid lesion, which fluctuated in size with associated tenderness and episodic right tearing. She had no preceding history of trauma, nasal surgery or radiotherapy. The referring ophthalmologist diagnosed her with right dacryocystitis.

On examination, she had right subcutaneous swelling inferior to the medial canthal tendon, which is where swelling secondary to dacryocystitis characteristically occurs. The lesion was a 5 mm × 5 mm nodular, non-erythematous area with associated tenderness. There were no associated overlying cutaneous signs and no signs of purulence from the puncta. On irrigation of the right lower eyelid punctum, there was minimal resistance; reflux and purulence were not evident. However, there was a delay in irrigation fluid reaching the pharynx indicating a

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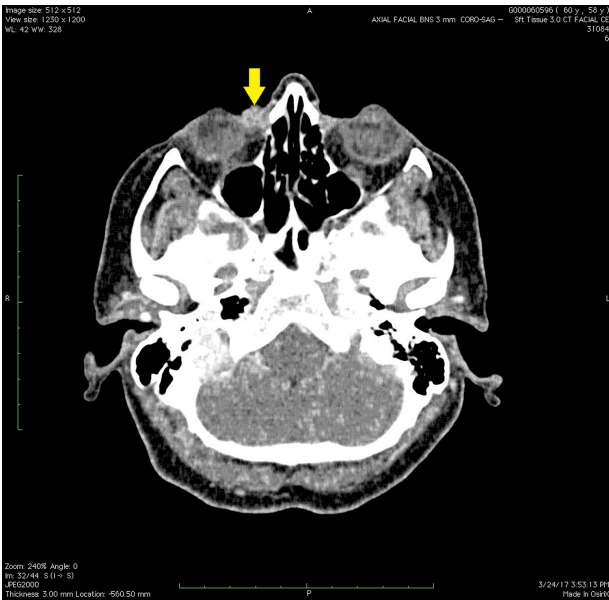


Figure 1: A non-contrast CT scan showing a well-circumscribed hyperintense circular area in the right medial canthal preseptal area.

partial obstruction of the nasolacrimal duct system. A computed tomography (CT) scan without contrast showed a well-circumscribed hyperintense area in the right medial canthal preseptal region, confluent with the lacrimal sac (Fig. 1).

The patient then underwent a right medial orbitotomy for exploration. Upon exploration, no visible lesion or pathology was present. Then, right nasolacrimal duct probing was performed, and right nasolacrimal duct obstruction was evident. A right dacryocystorhinostomy with Crawford tube placement was then performed to correct the nasolacrimal duct obstruction secondary to impingement by the mass lesion. Additionally, the right medial canthal subcutaneous tissue/lacrimal tissue was biopsied, which was subsequently identified pathologically as a Masson's tumor (Figs 2 and 3). The mass lesion was never visualized; neither was it excised. Upon pathological determination of the mass lesion, it was determined that, due to the indolent nature of the tumor, the risk of excision outweighed the benefit. Postoperatively, the patient experienced relief from the tenderness and tearing. The mass lesion has not progressed clinically since stent placement, and her symptoms remain completely resolved over the last 6 months.

DISCUSSION

Masson's tumor typically presents as a slow-growing, indolent and nodular area [7]. Its etiology is not well described, but it appears that it is a reactive proliferation of vascular endothelial cells mediated by the chemoattraction of macrophages and associated release of basic endothelial fibroblast growth factor [4, 5]. It can present primarily with an insult and thrombus formation to a vein, secondarily from vascular malformations or, rarely, extravascularly from an organizing hematoma [2, 1].

Masson's tumor is typically symptomless but will continue to grow until it is surgically resected. In rare cases, Masson's tumor may continue to grow until it causes mass-associated issues. In this case, we see impingement by the lesion and associated nasolacrimal duct obstruction. This patient's tumor

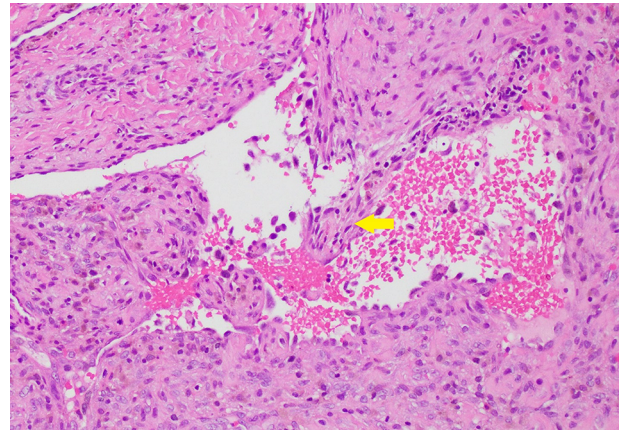


Figure 2: A histopathology slide showing papillary projections into the vascular lumen at $\times 20$ magnification, view 1. Note the absence of necrosis.

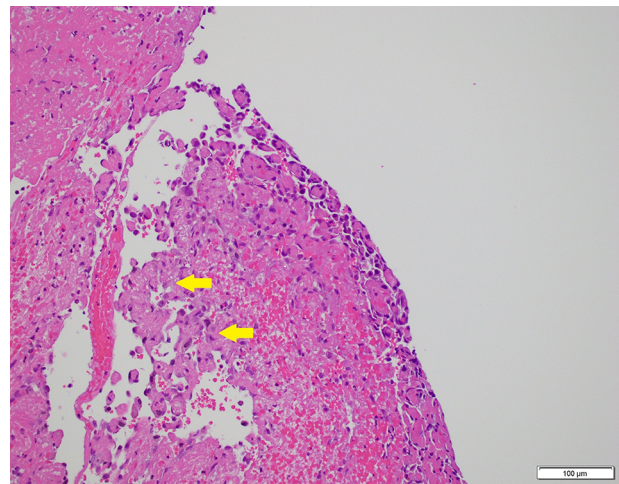


Figure 3: A histopathology slide showing papillary projections into the vascular lumen at $\times 40$ magnification, view 2. Note the lack of mitotic figures and cellular pleomorphism.

probably grew as a result of some thrombus secondary to a seemingly innocuous trauma to the face. This case could easily have been misdiagnosed and treated as dacryocystitis were it not for careful examination and resulting pathological findings.

It is not a common lesion, only occurring as 2% of all malignant and non-malignant vascular tumors of the skin [9]. Although rare, it is prudent for the oculoplastic surgeon/ophthalmologist to be familiar with Masson's tumor, especially because of its similarity to primary vascular neoplasms such as angiosarcoma. Entities on the differential diagnosis should include angioma, vascular malformation, pyogenic granuloma, angiosarcoma and Kaposi's sarcoma. It is important to differentiate these from Masson's tumor as it can be treated only with complete excision [8]. This is the first case report in the literature of Masson's tumor masquerading as dacryocystitis. This case highlights the importance of a thorough examination and having a low threshold to biopsy surrounding nasolacrimal tissue in atypical cases such as this.

CONFLICT OF INTEREST STATEMENT

None declared.

FUNDING

There was no funding.

ETHICAL APPROVAL

No ethical approval was required.

CONSENT

Written consent was obtained from the patient.

GUARANTOR

Peter Timoney is the guarantor of this article.

REFERENCES

1. Pins MR, Rosenthal D, Springfield DS, Rosenberg AE. Florid extravascular papillary endothelial hyperplasia (Masson's pseudoangiosarcoma) presenting as a soft-tissue sarcoma. *Arch Pathol Lab Med* 1993;**8**:259–63 PMID: 8442671.
2. Soares AB, Altemani A, Furuse C et al. Intravascular papillary endothelial hyperplasia: report of 2 cases and immunohistochemical study. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2008;**106**:708–11.
3. Levere SM, Barsky SH, Meals RA. Intravascular papillary endothelial hyperplasia: a neoplastic “actor” representing an exaggerated attempt at recanalization mediated by basic fibroblast growth factor. *J Hand Surg Am* 1994;**19**: 559–64.
4. Moriyama S, Kunitomo R, Sakaguchi H et al. Intravascular papillary endothelial hyperplasia in an aneurysm of the superficial temporal artery: report of a case. *Surg Today* 2011;**41**:1450–4.
5. Weiss SW, Goldblum JR. Benign tumors and tumor-like lesions of blood vessels. In: *Enzingers and Weiss's Soft Tissue Tumors*. New York: Mosby-Elsevier, 2008, 668–71.
6. Akdur NC, Donmez M, Gozel S et al. Intravascular papillary endothelial hyperplasia: histomorphological and immunohistochemical features. *Diagn Pathol* 2013;**8**:167.
7. Clearkin KP, Enzinger FM. Intravascular papillary endothelial hyperplasia. *Arch Pathol Lab Med* 1976;**100**:441–4 PMID: 947306.
8. Avellino AM, Grant GA, Harris AB et al. Recurrent intracranial Masson's vegetant intravascular hemangioendothelioma. Case report and review of the literature. *J Neurosurg* 1999;**91**:308–12.
9. Tedla M, Bežová M, Biró C et al. Intravascular papillary endothelial hyperplasia of larynx: case report and literature review of all head and neck cases. *Otolaryngol Pol* 2014;**68**:200–3.