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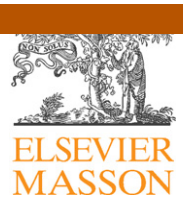

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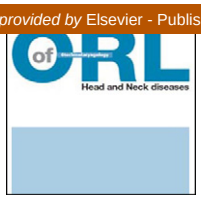

CASE REPORT

Nasal cavity lobular capillary hemangioma due to insect sting

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KEYWORDS
Lobular capillary hemangioma;
Pyogenic granuloma;
Nasal cavity;
Nasal foreign body

Summary
Introduction: Lobular capillary hemangioma is a frequent benign vascular inflammatory lesion of the skin tissue. It rarely reaches the mucous membrane, and the nasal fossa involvement is exceptional.
Case report: A 68-year-old woman presented with an ulcerous hemorrhagic mass blocking the left nasal fossa, which had appeared a few weeks after a wasp sting in the nose. The insect was evacuated only three weeks after the sting. The clinical and radiological data suggested malignancy. Biopsy under local anesthesia proved non-contributory and was complicated by 1 week's hospitalization for severe nosebleed. Surgical excision under video-endoscopy confirmed diagnosis. At 31 months' follow-up, the patient was free of recurrence.
Discussion/conclusion: The pathogeny of lobular capillary hemangioma is uncertain. No previous cases affecting the mucous membrane after insect sting have been reported. Except in the typical contexts of long-term packing or pregnancy, diagnosis can be difficult and misleading. It can mimic a malignant pathology. Its rich vascularization requires caution during biopsy, and the risk of recurrence requires excision to be complete.

Introduction

Lobular capillary hemangioma is a benign vascular lesion, very rarely involving the nasal cavities. The clinical presentation is usually suggestive. It is to be suspected in case of a unilateral ulcero-hemorrhagic fleshy lesion in the anterior part of the nasal fossa, with onset related to microtrauma such as prolonged packing [1,2]. We here report a case of atypical etiology following insect sting, showing a clinical and radiological aspect of neoplasia.

Case report

A 68-year-old woman presented with progressive left unilateral nasal obstruction of 6 weeks' evolution, associated with ipsilateral maxillary pain and iterative nosebleed.

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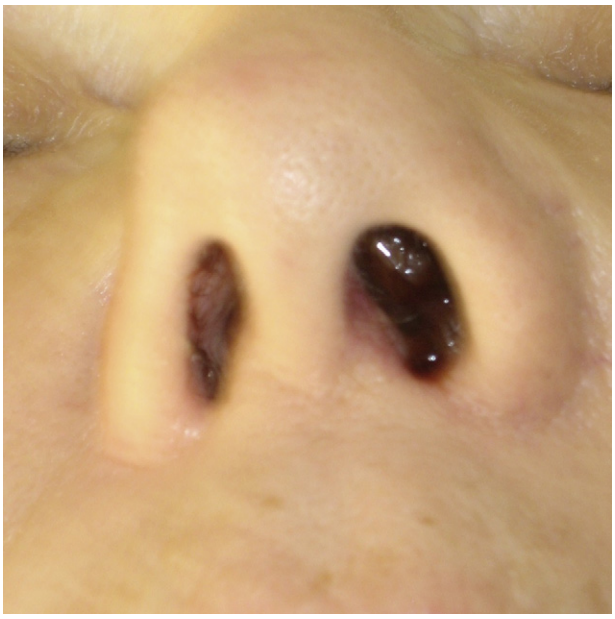

Figure 1 Left nasal cavity, clinical aspect.

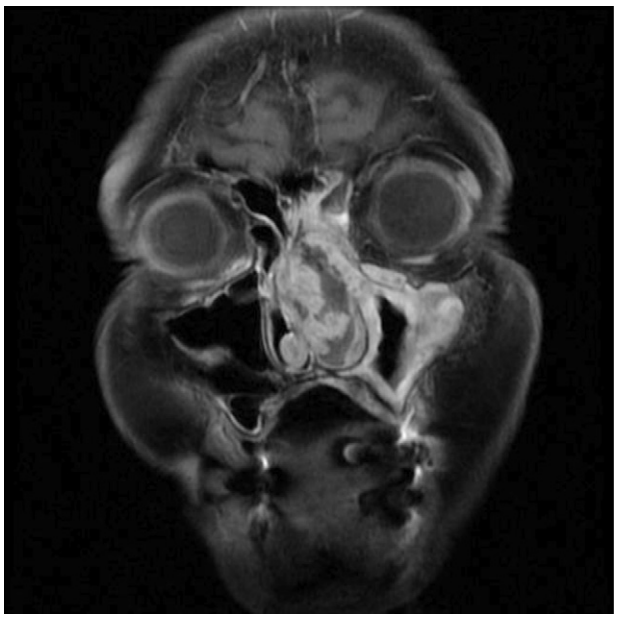

Figure 2 Sinus T1-weighted gadolinium-enhanced MRI, coronal slice.

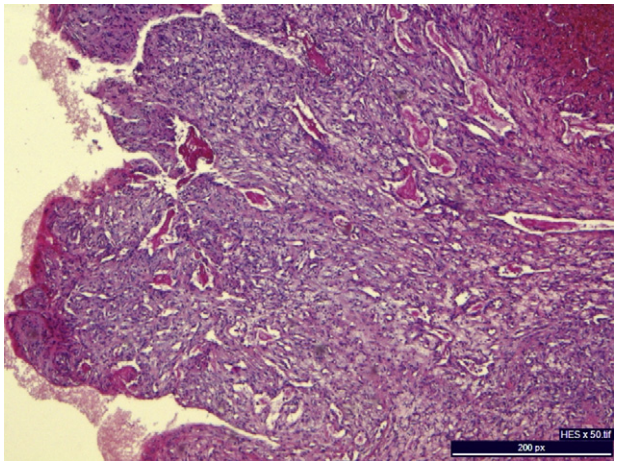

Figure 3 Standard histology. Aspect of polypoid vascular proliferation with superficial ulceration.

Anterior rhinoscopy found a large soft, fleshy ulcero-hemorrhagic tumor totally obstructing the left nasal cavity as of the vestibule (Fig. 1). Flexible endoscopy failed to determine the base of implantation or extension.

Four weeks previously, in a different center, biopsy had been performed under local anesthesia for a rapidly evolving fleshy mass, discovered in the left nostril, and led to severe nosebleed, requiring 1 week's hospitalization, while itself proving non-contributive.

Facial sinus CT with and without contrast enhancement found a richly vascularized tissular lesion pushing back the septal wall, lacrimal bone and nasal pyramid, associated with mild demineralization of the intersinonasal wall, suggestive of malignancy.

MRI found a lesion of 4 cm anteroposterior diameter and 2.5 cm height, extending from the vestibule to the tail of the middle concha and from the floor of the nasal fossa to the olfactory groove. There was left frontal-ethmoid-maxillary sinusitis and lacrimal duct ectasia.

The mass showed heterogeneous contrast enhancement and heterogeneous cerebriform T2 iso-signal, suggestive of inverted papilloma (Fig. 2). The syndrome comprising a mass involving the intersinonasal wall, nasal bones and lacrimal bone, however, rather suggested a malignant neoformation.

Complete excision was performed by 30° endoscope under general anesthesia, including a ring of healthy tissue around the middle concha implantation base.

Anatomopathology found a lesion formed of superficially eroded, richly vascularized granulation tissue, lifting up highly congestive nasal mucosa, with multiple polymorphic inflammatory elements, suggestive of lobular capillary hemangioma (Fig. 3).

On further interview, the patient recalled a wasp sting in the same nostril, 5 days before symptom onset; she had evacuated the insect, a mason wasp (Odynerus reniformis), only 3 weeks later.

At 31 months' follow-up, the patient had shown no recurrence.

Discussion

Lobular capillary hemangioma is also known as pyogenic granuloma or botryomycoma. It is a benign, richly vascularized inflammatory lesion of the skin or mucosa. Intravascular and subcutaneous locations have been reported.

Mucosal lobular capillary hemangioma is generally found on the gums, lips, tongue or oral mucosa; intranasal locations are very rare [1,2]. Patrice et al. reported a 21% rate of mucosal involvement in a series of 178 patients, with only one intranasal location [3].

Cutaneous locations show male predominance, whereas mucosal involvement is twice as frequent in women [4], especially during pregnancy.

Clinically, lobular capillary hemangioma can mimic malignancy in its fleshy, unilateral, ulcerated hemorrhagic aspect and rapid growth.

There is no specific aspect on imaging.

The intranasal extension and implantation base can be determined on CT and MRI if lesion size prevents endoscopic exploration.

Lobular capillary hemangioma shows few radiologic criteria of malignancy. It may distort the nasal septum [5], but generally respects bone structure [6]. Lee, however, reported bone erosion in 50% of cases and bone displacement in one-third [7]; there was no sinusal, ocular, cerebral or nervous extension.

Positive diagnosis is only made on histologic findings of an aspect of neoangiogenesis with vessels oriented perpendicularly to the surface, associated with accompanying inflammatory tissue. Ulceration of the covering mucosa definitively rules out benign or malignant vascular tumor.

Biopsy and total excision should preferably be performed in a single step in the operating theater under general anesthesia. Hemorrhage may require efficient means of hemostasis. Incomplete excision may mislead anatomopathologic diagnosis if the specimen includes no mucosal ulceration.

Lobular capillary hemangioma pathogeny [8] is uncertain, involving an angiogenic disorder of unknown origin. There is increased expression of phosphorylated transcription activating molecules (p-ATF2 and p-STAT3) and of tumor suppressor gene. Hormonal changes, notably during pregnancy, impact vascular systems by the action of vascular endothelial growth factor (VEGF) and estrogens. Medical etiologies exist, notably implicating epidermal growth factor receptor (EGFR) inhibitors but also antivirals, immunosuppressors and chemotherapy drugs.

Intranasal locations were reported following microtrauma [9], prolonged packing [1] or foreign-body presence [10], and trauma by finger. Their frequent location in the anterior part of the septum (Little's area) or head of the inferior concha corresponds to such traumatic causes, mainly affecting the anterior part of the nasal cavities. Our review of the literature retrieved no other cases of mucosal lobular capillary hemangioma secondary to an insect sting.

The main differential diagnoses are: nasal polyp, inverted papilloma, capillary hemangioma, fibroma, meningoencephalocele, sarcoidosis, Wegener's granulomatosis, histiocytoma, adenocarcinoma, glioma, fibrosarcoma, achromic melanoma, Kaposi's sarcoma, lymphoma, hemangiopericytoma, esthesioneuroblastoma and angiosarcoma.

Lobular capillary hemangioma shows frequent recurrence after treatment [4]. Subperichondrial or subperiosteal excision with healthy mucosal margins minimizes recurrence (3.7%) compared to simple excision [6].

Conclusion

In nasal locations, lobular capillary hemangioma may be hard to differentiate from a malignant lesion on clinical examination and imaging, especially when there is no suggestive etiology. History taking should look for microtrauma and mucosal aggression, of however unexpected a cause, to guide diagnosis. Biopsy incurs a risk of nosebleed. Excision should be complete in first intention, performed in the operating theater under general anesthesia; this allows diagnosis and treatment in a single step and avoids secondary hemorrhage.

Disclosure of interest

The authors declare that they have no conflicts of interest concerning this article.

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