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CASE REPORT

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Pyogenic Granuloma Gravidarum of the Nasal Cavity Causing Recurrent Epistaxis. A Case Report and Review of the Literature

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ABSTRACT

Pyogenic granuloma is a benign proliferative fibrovascular lesion commonly arising from the skin and mucous membranes of the head and neck region. Histologically, this tumor is characterized by vascular proliferation and a circumscribed group of capillaries organized in lobules. It is usually located in the oral cavity and nasal location is less frequent. When it occurs in pregnant women, it is usually referred as pyogenic granuloma gravidarum. In this article we present the case of a pyogenic granuloma gravidarum in a young woman with intermittent epistaxis during the last trimester of pregnancy that did not resolve after childbirth and was treated with transnasal endoscopic resection and cautery at the base of the lesion for hemostasis under local anesthesia.

Keywords: pyogenic granuloma, pregnancy, nose, trauma, endoscopy.

INTRODUCTION

Pyogenic granuloma is a benign proliferative fibrovascular lesion arising commonly from the skin and mucous membranes of the head and neck region (1, 2). It was first described in 1897, by Poncet and Dor, as a botryomycotic infection known as human botryomycosis. Later, in 1904, Hartzell assumed this lesion to be granulation tissue developing after bacterial infection and thus invented the term pyogenic granuloma (3, 4). According to Bhattacharyya *et al* (5) and Miller *et al* (6), pyogenic granuloma is a misnomer because this lesion is neither pyogenic nor granulomatous and the most accurate term for this pathology is lobular capillary hemangioma, which was proposed in 1980 by Mills *et al* (7). Lobular capillary

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hemangioma is believed to be the most appropriate descriptive term because histologically this tumor is characterized by vascular proliferation and a circumscribed group of capillaries organized in lobules. However, pyogenic granuloma remains the most popular term in the literature. These changes in terminology indicate an evolution in the understanding of the underlying etiology of this lesion. Although the exact pathogenesis of pyogenic granuloma is still unclear, it has been linked to a variety of factors like minor local trauma or chronic low-grade irritation, hormones and immunosuppression (8). It may develop at any age, but the prevalence is higher in the third decade, with a slight female predominance, especially in their reproductive years (9, 10). In the head and neck, it is usually located in the oral cavity and nasal location is less frequent. Intermittent epistaxis is the most common symptom due to the excessive vascularity of the tumor. Other symptoms include nasal obstruction, nasal discharge, epiphora, and less frequent nasal pain. Clinical examination reveals a pedunculate, grey to pink mass with irregular borders (11, 12).

When pyogenic granuloma occurs in pregnant women, it is called pyogenic granuloma gravidarum and is clinically and histologically identical. It affects 5% of pregnant patients and is most common during the last two trimesters (13-15). Most of these lesions are small and they usually regress spontaneously postpartum.

In this article, we present the case of a pyogenic granuloma gravidarum in a young woman with intermittent epistaxis during the last trimester of pregnancy that did not resolve after childbirth. ❑

CASE REPORT

A 33-year-old woman presented to our department in August 2022 due to intermittent epistaxis from the right nasal cavity during the past four months. From her medical history it is important to mention that, one month before presentation, she had given birth to her second child and was breastfeeding. Her first pregnancy was uneventful. According to the patient, the first episodes of epistaxis occurred at the beginning of the last trimester of pregnancy. She also mentioned having a nasal piercing for fifteen years, which she had recently removed due to the nose bleeding episodes. A mild nasal obstruction was also present. The rest of her medical history was unremarkable. Epistaxis was controlled with local pressure and nasal packing had not been used. Anterior rhinoscopy of the right nasal cavity revealed a red nasal mass with increased vascularity located on the anterior part of the nasal septum. Endoscopy with a 0° rigid

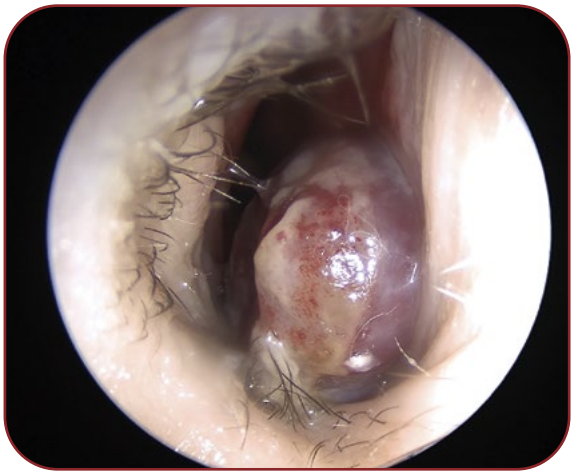

FIGURE 1. Endoscopic examination of the right nasal cavity with a 0° rigid endoscope revealing a hemorrhagic intranasal lesion located on the anterior part of the nasal septum

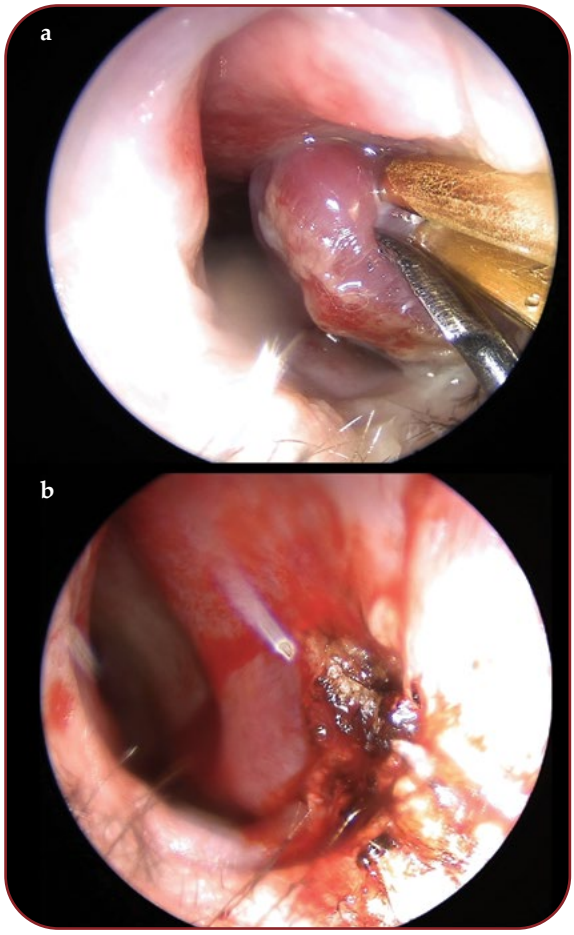

FIGURE 2a, b. (a) Transanasal endoscopic resection of the lesion under local anesthesia using a Freer elevator and bipolar electrocautery; (b) endoscopic view after excision of the lesion

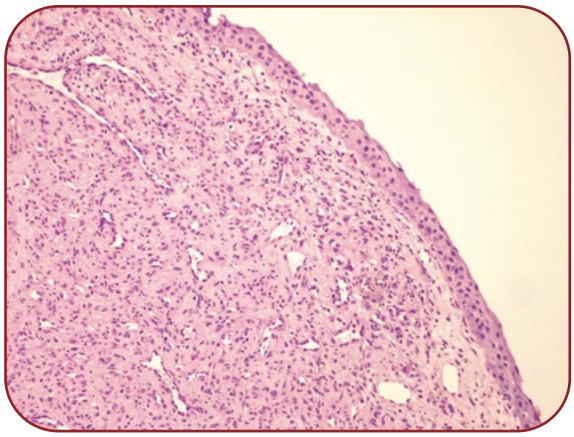

FIGURE 3. Histological section showing the typical pattern of pyogenic granuloma (lobular capillary hemangioma, hematoxylin and eosin x 10)

nasal endoscope revealed the presence of the above-mentioned tumor without other pathological findings (Figure 1). The tumor was confined to the anterior part of the right nasal cavity with a pedunculated base arising from the Little's area of the nasal septum. The patient was scheduled for an endoscopic excisional biopsy under local anesthesia. Bipolar electrocautery and a Freer elevator were used, and the entire lesion was resected and sent for histopathological examination (Figure 2a, b). Biopsy revealed a pyogenic granuloma (Figure 3). There were no signs of recurrency until the patient's most recent follow-up. ❑

DISCUSSION

Pyogenic granuloma is a benign vascular lesion with a poorly understood pathogenesis. When it occurs in pregnant women, it is usually referred as pyogenic granuloma gravidarum or "pregnancy tumor". Although the exact etiology remains unclear, nasal cavity trauma and increased levels of estrogen and progesterone have been usually implicated in the development of pyogenic granuloma (16). Other risk factors that have been described in the literature include preexisting skin conditions, viral oncogenes, underlying microscopic arteriovenous malformations and angiogenic growth factor production (4, 9). The most common traumatic factors include nose picking and nasal packing (5, 17, 18). There are also other uncommon traumatic factors such as nasal surgery, nasal intubation, nose piercing and intranasal cocaine use that have been related to pyogenic granuloma. Neves-Pinto *et al* described a case of a nasal septum giant pyogenic granuloma after long lasting nasal intubation (19). Alghamdi Jr. *et al* reported an interesting case of a recurrent cutaneous pyogenic granuloma as a complication of rhinoplasty (20). Another interesting study presents a case series of eight patients with nasal pyogenic granuloma after decorative nose piercing (21).

Regarding hormonal factors, higher levels of estrogen and progesterone during pregnancy have been associated with the pathogenesis of pyogenic granuloma gravidarum. It is believed that elevated levels of estrogen and progesterone could stimulate endothelial proliferation within the oral and nasal mucosa in response to preexisting local trauma resulting in granulation tissue formation (4, 14, 16). Oral contraceptives have also been related to a higher incidence of pyogenic granuloma (4). The exact molecular mechanism is not yet known, but it seems that higher levels of circulating hormones are more important than the number of progesterone and estrogen hormone receptors which has been found to be similar in tissue samples of pyogenic granuloma from non-pregnant women, pregnant women and men (16). Another theory that has been proposed suggests that progesterone and estrogen receptors are transiently expressed in an early phase of the development of these lesions (4, 22). Also, progesterone regulates the VEGF-A-VEGFR2 signaling, which is a promoter of angiogenesis and elevated levels might cause an imbalance of promoters and inhibitors during the angiogenic process (23). In our case, the patient was pregnant and had a history of nose piercing that could probably be related to chronic local trauma.

Usually, pyogenic granuloma gravidarum affects the oral mucosa, especially the gingiva, while the nasal cavity is an unusual site of location with a reported incidence of 7%-10% (1). Most cases of nasal pyogenic granuloma occur on the nasal septum (76%) and the inferior turbinate (12%-20%) (9, 16). In our case, the lesion was located on the anterior part of the nasal septum.

The most common symptoms include recurrent unilateral epistaxis and nasal obstruction as in our patient. Symptoms like headache, smell disorder and facial pain are very rare. Epistaxis is a frequent ear, nose and throat emergency and in most cases, it is anterior originating from the Kiesselbach plexus. Regarding etiology, epistaxis can be idiopathic or secondary due to trauma,

surgery, vascular abnormalities, coagulopathies, anticoagulant use and neoplasms (24). Trauma is one of the most common causes of epistaxis, with nasal bone fractures being the most common cause of traumatic nosebleeds in the emergency department (25). In our patient, recurrent episodes of anterior epistaxis were the main symptom. On endoscopic examination, pyogenic granuloma appears as a gray-to-pink, red or dark, irregular and hypervascularized tumor with a pedunculated base (16, 23). Usually, these lesions are small, but giant lesions have also been described in the literature (17, 23).

Imaging plays a very significant role in determining the extent of a nasal mass and in guiding surgical planning. Due to its rapid growth, pyogenic granuloma might be easily misdiagnosed as a malignant tumor. Computed tomography (CT) findings of pyogenic granuloma include a well-defined unilateral tumor of soft tissue density, with marked enhancement and no calcification (14, 26). On magnetic resonance imaging (MRI), characteristic features of nasal pyogenic granuloma comprise high heterogeneous T2 signal intensity with a thin peripheral isointense or hypointense ring and spontaneous T1 hypointensity (4, 14, 16). Our patient presented with a small lesion that was entirely visible on endoscopy with a rigid 0° endoscope. There were no signs of local invasion, and delineation of the intranasal tumor extension was possible with endoscopy, so imaging was not performed before surgery. Also, our patient had recently given birth and was breastfeeding, so she preferred to avoid a CT scan. The patient was claustrophobic and did not consent for an MRI. Ultrasonography has been used for the examination of nasal bone fractures and sinusitis in the emergency department (27). Due to lack of irradiation, ultrasonography would be an option, but the intranasal location of the lesion on the nasal septum, and not on the external nasal pyramid, did not favor the use of this imaging technique.

The differential diagnosis of pyogenic granuloma includes angiofibroma especially in male adolescents, inverted papilloma, low-grade angiosarcoma, squamous cell carcinoma, hemangioma and hemangiopericytoma (4, 9). When a family history of hereditary hemorrhagic telangiectasia is present, differential diagnosis from telangiectasia is necessary. Also, in immunocompromised patient's Kaposi sarcoma should be excluded (23).

A variety of treatment methods have been described in the literature, including surgical excision, diode laser, cautery, embolization, drug therapy, cryotherapy and silver nitrate cautery. Drug therapy is usually used for small lesions and includes treatment with topical b-blocker (28), imiquimod (29) and ingenol mebutate (30). Surgery remains the treatment of choice due to the low recurrence rate of pyogenic granulomas after surgical resection. Preoperative superselective embolization in order to reduce the risk of hemorrhage, followed by lateral rhinotomy has been described in the literature for giant nasal pyogenic granuloma gravidarum (31), but in most cases, transnasal endoscopic surgical excision is considered the ideal approach (4). In adult patients, small lesions can be resected under local anesthesia as in our patient. In pregnant women, the decision for surgery depends on the gestational age and amount of blood loss; it is usually postponed until after childbirth and is reserved for lesions that fail to regress postpartum (4, 31, 32).

The lack of imaging modalities is the main limitation of this case study, which was discussed earlier in the text. Also, the relatively short postoperative time (15 months) could be considered a limitation regarding recurrence concerns. ❑

CONCLUSION

Nasal pyogenic granuloma gravidarum is less common and is associated with unilateral epistaxis and nasal obstruction. This case adds to the literature an intranasal occurrence of pyogenic granuloma associated with pregnancy and local trauma probably caused by nasal piercing. Through this case study we demonstrate that pyogenic granulomas sometimes do not regress after childbirth and can affect patients' quality of life. Also, this case emphasizes the fact that recurrent epistaxis in women who have just given birth and are breastfeeding needs a more careful and detailed endoscopic examination and should be referred to a specialist ENT doctor. Transnasal endoscopic resection under local or general anesthesia depending on location and size of the lesion is the treatment of choice for pyogenic granuloma. Otorhinolaryngologists and obstetrician-gynecologists should be aware of

this pathological entity in order to guide proper diagnosis and management. ❑

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