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Case Series

Nasal lobular capillary hemangioma: etiologic factors

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ABSTRACT

Nasal lobular capillary hemangioma (LCH) is a rare and benign vascular tumor of unknown etiology, often associated with hormonal factors and a history of trauma. In this case series we present twelve cases of nasal LCH diagnosed on patients treated at the Hospital da Senhora da Oliveira from January 2010 and December 2019. Of the 12 patients, 66, 7% were males. The most common clinical presentation was epistaxis (75%) followed by nasal obstruction (50%). In 75% of patients the most commonly affected area was the nasal septum. Regarding proposed contributing factors, nasal trauma was the most prevalent (33%), followed by the use of oral contraceptives (25%) and pregnancy (8.3%). Endoscopic sinus surgery was performed in all patients, with complete excision of the lesion, and no post-operative complications were observed. Nasal LCH should be considered in the differential diagnosis of nasal vascular lesions associated with unilateral epistaxis and nasal obstruction. Complete surgical resection should be the treatment of choice, since it's associated with lower risk of recurrence.

Keywords: Epistaxis, Lobular capillary hemangioma, Nasal obstruction

INTRODUCTION

Lobular capillary hemangioma (LCH) is a benign vascular neoformation, of rapid growth, which mainly affects the skin and mucous membranes of the head and neck (gingiva, lips, tongue and buccal mucosa). It is a rare entity in the nasal cavity. The most commonly affected area in the nasal cavity is the nasal septum, followed by the lateral nasal wall and vestibule.[1-4]

Initially described by Poncet and Dor in 1897 as human botryomycosis, speculating a fungal origin. It was later classified by Hartzell et al as pyogenic granuloma, assuming the presence of granulation tissue that grew in response to a bacterial infection. Based on microscopic features and the absence of evidence supporting an infectious or granulomatous origin it was subsequent designated lobular capillary hemangioma.[5-7]

In epidemiological terms, the incidence of nasal LCH increases in the third decade of life, with a predominance in females, however, after 40 years of age, the proportion of affected men and women appears to be similar.[7] Nasal LCH in children is uncommon.[8] The etiology of nasal LCH remains unknown, although there is evidence to support the role of local trauma and hormonal factors as possible contributing factors for its development.[1,5,9] Patients with LCH reported nasal trauma between 7% and 23% of cases. The most frequently described traumas were nose-picking, nasal packing and lower turbinate surgery. Most nasal LCH caused by trauma usually occurs in the anterior part of the nasal septum or in the anterior aspect of the lateral nasal wall.[5,10] Hormonal factors also appear to be a trigger, given the well-established association of increased estrogen and progesterone levels during pregnancy and the development of nasal LCH.[7,10]

Nasal LCH usually presents with epistaxis and nasal obstruction, depending on the size and location of the lesion.[1,2] Macroscopically, it presents as a solitary red-purple papule or nodule, with a friable surface with or without ulceration, and may be pedunculated or broad-

based. Microscopically, it has a typical architecture, with vascular proliferation of endothelial cells organized in a characteristic pattern of capillaries circumscribed and organized in lobules.[5-7] Several investigations as local nasal examination, diagnostic nasal endoscopy and imaging like computed tomography (CT) with contrast, may be necessary to assess the extent, suggest the possible nature of the lesion (site of origin, growth pattern and vascularization) and help in the differential diagnosis.[6,9] Surgery excision is the treatment of choice for LCH of the nasal cavity. Regardless of size, endoscopic sinus surgery can be considered the ideal approach, with low surgical morbidity.[5] Complete excision of the lesion with resection in the subperichondrial or subperiosteal plane and with a margin of normal mucosa around it should be the recommended treatment in order to minimize the possibility of recurrence.[1]

CASE SERIES

We presented twelve cases of nasal LCH or pyogenic granuloma diagnosed on patients treated at the Hospital da Senhora da Oliveira, Guimarães from January 2010 and December 2019. All the patients were evaluated with anterior rhinoscopy, endonasal endoscopy and nasal CT scan prior to surgery to study the lesion. Endoscopic resection in general anaesthesia followed the recommended approach, with complete excision of the lesion with resection in the subperichondrial or subperiosteal plane and with a margin of normal mucosa. All the patients had a histological diagnosis of LCH from surgical excision.

This study was conducted in a manner that warrants confidentiality of all included patients. Data collected had been deidentified prior to being stored. Permission was granted by the institution's ethical committee before starting the study and the data collection. Of the 12 patients, 8 were male (66.7%) and 4 were female (33.3%) (Figure 1). The mean age was 34.1 years old (SD±22.8 years old) with a range from 12 to 79 years old.

Epistaxis was the most common presenting symptom in 75% (n=9) of the patients, followed by nasal obstruction (50%, n=6), ipsilateral to the location of the lesion. The most common site of the LCH was the nasal septum (n=9, 75%), followed by the nasal cavity (n=2, 16.7%) and the inferior turbinate (n=1, 8.3%). Demographic and clinical characteristics are summarized in Table 1.

Nasal trauma was the most prevalent contributing factor, with a history of nasal packing in 33.3% (n=4) of the patients. The use of oral contraceptives was found in 25% (n=3) of the cases and only one patient was pregnant at the time of the diagnosis.

All patients were evaluated with anterior rhinoscopy and endonasal endoscopy and underwent nasal CT evaluation prior to surgery. All patients had unilateral LCH and there were no cases with multiple lesions.

Endoscopic sinus surgery was performed in all patients, with complete excision of the lesion with resection in the subperichondrial or subperiosteal plane and with a margin of normal mucosa around it. No post-operative complications were observed in any patients.

Patients were followed up for a period ranging from 12 months to 36 months and there was no record of recurrences in any patients.

Table 1: Demographic and clinical characteristics.

Characteristics	N (%)
Age (years)	
Mean	34.1
Minimum	12
Maximum	79
Gender	
Men	88 (66.7)
Women	4 (33.3)
Presenting symptom	
Epistaxis	9 (75)
Obstruction	6 (50)
Location	
Septum	9 (75)
Nasal floor/still	2 (16.7)
Inferior turbinate	1 (8.3)

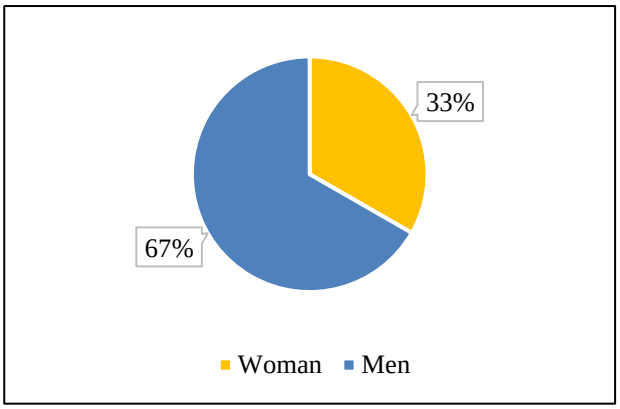

Figure 1: Distribution by gender.

DISCUSSION

Nasal LCH is a benign tumor of vascular origin, which presents as a friable nasal mass, sometimes with an ulcerated surface, associated with symptoms such as epistaxis and, depending on the size and location, unilateral nasal obstruction.[1,2] As corroborated in our case series, epistaxis was the initial clinical manifestation in the majority of the cases (75%). It can occur at any age, with cases reported from 10 months to 72 years of age, though with a higher incidence between the third and fifth decade of life.[1,11] In our case series the average age was 34 years, with a range of 12 to 79 years old, which is in agreement with the published literature. Previous studies reported that

nasal LCH occurs equally in males and females. Smith et al in their study of 34 patients with LCH reported an incidence of 50% in females and Puxeddu et al reported a 47.5% incidence in females in a study of 40 patients.[1,11] More recently, Alexis Lopes et al in their study with 38 patients reported an incidence of 65.8% in females.[7] Contrary to the literature, in our cases the incidence of nasal LCH was higher in males (66.7%). Nasal LCH usually grows from the nasal septum, predominantly from the little area, and/or inferior turbinate and nasal vestibule.[11] Little's area is located in the most anterior portion of the nasal septum and here we found the anastomosis of four arteries (anterior ethmoidal artery, sphenopalatine artery, greater palatine artery and septal branch of the superior labial artery).

Our results were similar to the literature with the most common site being the nasal septum (75%), followed by the inferior turbinate (16.7%). The etiology of nasal LCH remains unknown, however several etiological factors can contribute to its development, like local trauma being present in various studies in 7% and 23% of cases. Trauma can cause vascular rupture and changes in the surrounding soft tissues, with subsequent tissue necrosis and recruitment of inflammatory and angiogenic mediators, eventually leading to the formation of a hemangioma.[12] In our case series, nasal trauma was documented in 33% of cases, a higher percentage than reported in the literature. Pexuddu et al and Smith et al reported an incidence of nasal LCH in pregnant women of 5% and 29.4%, respectively.[1,11] There was only one pregnant woman in our study. The use of oral contraceptives was present in 25% of the cases.

The use of imaging studies like CT scan is controverse, being considered mandatory in some studies.[13] Pexeddu et al recommended the use of CT scan only in large lesions and with superior extension towards the base of the skull. In terms of imaging features, the diagnosis of LCH consists of the identification of a mass with intense contrast uptake surrounded by a circumferential halo.[1,6,14] In our case series, the imaging study was performed in all patients and no bone erosion or extension to the paranasal or intracranial sinuses was observed. Histopathological examination is crucial for the diagnosis of LCH. It is characterized by endothelial proliferation with prominent vascular spaces, capillary in a lobular pattern, ulceration of the epithelium and a pedicle composed of fibrovascular tissue.[9,10]

The differential diagnosis of an intranasal mass includes a wide range of congenital, inflammatory and neoplastic pathologies. The most important differential diagnosis is nasal angiofibroma, especially in young male patients. Other differential diagnoses to consider are antrochoanal polyp, meningocele, meningoencephalocele, sarcoidosis, granulomatosis with polyangiitis, papilloma and some less common malignant lesions such as squamous cell carcinoma, nasopharyngeal carcinoma or nasopharyngeal teratoma.[1,7,9] Surgical excision is the preferred treatment for nasal LCH, as recommended in our population. Other therapeutic options include cauterization, embolization and cryotherapy.[3] There are no comparative studies on the different surgical methods used for the LCH. Complete resection of the lesion, complemented by the removal of the perichondrium surrounding the base of the lesion, minimizes the possibility of recurrence which varies between 0% and 42%.[1,9,11] Regarding risk factors for recurrence evidence is scarce, however, Smith et al reported a higher local recurrence in older patients.[11] When nasal LCH occurs during pregnancy it may regress after delivery, with surgery being reserved for lesions that have not completely reverted.[7,9]

CONCLUSION

In our case series, nasal LCH was diagnosed more frequently in males. About 33% of the population had a history of nasal trauma, hormonal factors like oral contraceptives and pregnancy were observed in 25% and 8% of cases, respectively. Contrary to the literature, our results suggested male gender and a history of nasal trauma as the main contributing factors for the development of nasal LCH. Although it is a rare lesion, particularly in the pediatric population, LCH should be considered in the differential diagnosis of nasal vascular lesions associated with unilateral epistaxis and nasal obstruction. Complete surgical resection should be the treatment of choice, with an associated lower risk of recurrence.

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