



Case report

Nasal lobular capillary hemangioma: Report of a case managed by endoscopic excision and pre-operative angio-embolization

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ABSTRACT

Introduction: Lobular capillary hemangiomas are fast-growing benign vascular lesions with distinctive histopathological characteristics. The head and neck region is a common location for lobular capillary hemangiomas. However, the presence of such lesions in the nasal cavity is rare. Although several contributing factors have been identified in literature, the exact pathophysiology is not yet well understood. Predisposing factors include nasal trauma, pregnancy, and the use of contraceptive pills. Thus, the disease is more prevalent in females, with variable peak incidence in pediatric patients. Unilateral nasal obstruction and recurrent epistaxis are the most common symptoms of nasal lobular capillary hemangiomas. Radiological evaluation using contrast-enhanced computed tomography and magnetic resonance imaging is often required for large lesions.

Case presentation: We present a 30-year old female who presented to ENT clinics with two month complaint of left-sided nasal obstruction and epistaxis with left facial pain and headache. She had no predisposing risk factors. Imaging with CT and MRI revealed a large hypervascular mass in left nasal cavity. Surgical excision preceded by pre-operative embolization was done.

Discussion: Endoscopic endonasal excision is the standard of treatment. While some authors believe that pre-operative embolization is not required, others advocate its use. Based on literature, recurrence rate is variable.

Conclusion: We believe that use of pre-operative embolization for large nasal lobular capillary hemangioma would have an impact on perioperative morbidity.

1. Introduction and importance

Lobular capillary hemangiomas (LCH) are unique vascular tumors commonly found in the skin and mucous membranes of the head and neck. However, lobular capillary hemangioma in the nasal cavity is rare [1]. Owing to previous lack of histopathological knowledge of LCH, the lesion was termed human botryomycosis and later was termed pyogenic granuloma before 1980 [2]. The unique histopathological characteristics of capillaries arranged in a lobular pattern around feeder vessels were first described by Millis et al. in 1980 and were termed lobular capillary hemangiomas [3]. Clinically, nasal lobular capillary hemangiomas present with unilateral epistaxis and nasal obstruction. The etiopathology of LCH is not well understood. However, nasal picking, trauma to the region, pregnancy, hormonal imbalance, and use of oral contraceptives are predisposing factors that play a role in LCH formation [4]. Although different management options have been described in the

literature, endoscopic surgical excision is the standard treatment [5]. In this paper, we present a case of a patient with left large nasal lobular capillary hemangioma treated by endoscopic excision and pre-operative embolization. The role of angio-embolization is insufficiently discussed in the literature. We will also perform a review of the literature on nasal LCH. This work has been reported in line with the SCARE and PROCESS criteria [6,7].

2. Case presentation

A 30-year-old female with no known pre-existing medical conditions was referred by the primary health care clinic to the rhinology clinic. The patient's complaints were as follows: left-side nasal obstruction, recurrent left epistaxis, and headache over the left side of the head for the last two months. No other associated symptoms were noted. There was no previous history of nasal picking, nasal packing, or pregnancy.

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There was no medical or surgical history. Upon examination of the nasal cavity, the left soft tissue filling the nasal cavity appeared reddish to bluish, with a smooth surface, and bled on palpation. It appears to be attached to the nasal septum. Ear, throat, neck, and fiberoptic nasopharyngeolaryngeal endoscopy examinations were unremarkable. The patient had unremarkable basic blood tests. Contrast-enhanced computed tomography was performed to evaluate the nature and extent of the lesion. CT showed a 3.5×1.2 cm soft tissue lesion that showed heterogeneous enhancement in multiple patchy areas within the lesion. It appeared attached to both the lateral and medial nasal walls with mild deviation of the nasal septum and no bone erosions (Fig. 1). MRI was also performed for soft tissue delineation and revealed a hypointense lesion on the non-contrast T1 image and heterogeneously hyperintense lesions with multiple flow voids in the T2 image. The mass showed inhomogeneous enhancement, particularly at the periphery, after contrast administration (Figs. 2–3). Angiography revealed a hypervascularized left nasal cavity mass with 70–80 % of its supply from the anterior ethmoidal artery branches bilaterally and the rest from the internal maxillary artery (Fig. 4). Based on these radiological findings, endoscopic surgical excision preceded by pre-operative embolization was performed (Fig. 5). Owing to the high risk of complications associated with anterior ethmoid embolization, only embolization with 150–250 μ m PVA was performed on the left internal maxillary artery. After embolization, 20–30 % of the blood flow to the tumor was diminished. The removed mass was sent for histopathology, which confirmed the diagnosis of nasal lobular capillary hemangioma. No perioperative complications were noted. The patient was sent home the next day and was seen two weeks postoperatively for silastic sheet removal and examination, which showed a patent nasal cavity and healing mucosa. The patient was followed up after 18 months postoperatively and showed healed nasal mucosa, patent nasal cavity, and no recurrence. Subsequently, the patient became pregnant and delivered without disease recurrence. We believe that preoperative embolization has helped reducing the operative time and perioperative complications.



Fig. 1. Coronal CT showed heterogeneous enhancement in multiple patchy areas within the lesion.

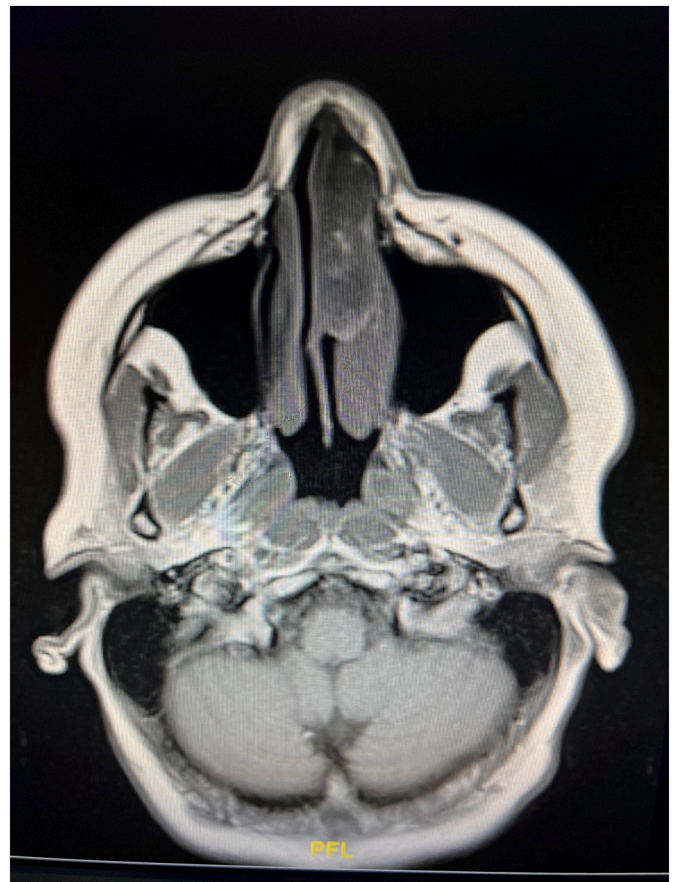


Fig. 2. MRI T1 revealed a hypointense lesion on the non-contrast T1 image.

3. Clinical discussion

Lobular capillary hemangioma, also referred to as pyogenic granuloma, represents a vascular lesion of rapid growth and benign histology. It affects skin and mucous membranes, most commonly in the head and neck region. Location of lobular capillary hemangioma in nasal cavity has rarely been reported in the literature [8]. Incidence of lobular capillary hemangioma has been reported in different age groups, including pediatric population [9]. Roberto et al. found in their retrospective study that the peak incidence was in the fifth decade of life [10].

The pathophysiology of lobular capillary hemangiomas is not well understood. In the literature, the disease is linked to trauma, pregnancy, hormonal factors, oncological and angiogenic factors, and underlying vascular malformation [11]. LCH from trauma can be caused by nasal picking or nasal packing for epistaxis management. Hormonal factors with elevated estrogen and progesterone levels were observed in pregnant females and patients taking oral contraceptives [4].

Clinically, patients commonly present with unilateral epistaxis, nasal obstruction, and rhinorrhea. Endoscopic examination shows polypoidal, hypervascular masses of variable size, which may bleed on palpation [11]. Reported common nasal locations of LCH include the nasal septum, vestibule, inferior turbinate, middle turbinate and uncinat process [10]. Nasal LCH with multiple sites of origin has been also reported [12]. Differential diagnoses include granulomatous disease, angiofibroma, hemangiopericytoma, inverting papilloma, or malignant sinonasal neoplasms [13].

Radiological evaluation of large lobular capillary hemangiomas is generally advocated in the literature. Computed tomography features include a well-circumscribed soft tissue mass and the absence of calcification with marked enhancement in the early phase and decreased

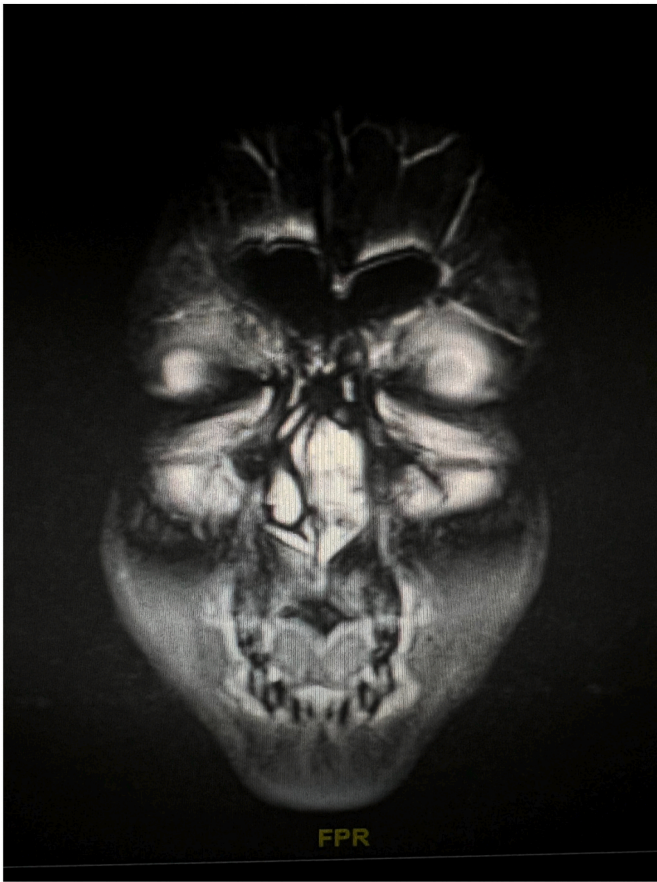


Fig. 3. T2 MRI showed heterogeneously hyperintense lesions with multiple flow voids.



Fig. 4. Angiography revealed a hypervascularized left nasal cavity mass with 70–80 % of its supply from the anterior ethmoidal artery branches.

enhancement in the delayed phase in dual-phase [14]. Tzu-hang et al. observed no bone erosion in a retrospective study on computed tomography [13]. On magnetic resonance imaging, LCH appears hypointense in T1 and hyperintense in T2 with a hypointense rim with low voids, and areas of hemorrhage can be depicted [15].

Histopathologically, LCH appears as vascular proliferation in lobular arrangements with stroma characterized by inflammation and edema, similar to granulation tissue. Variations may be attributed to the chronological phase of the lesion [16].

The mainstay treatment of LCH is endoscopic surgical excision. Several studies have advocated pre-operative embolization of lesions, specifically for large lesions [17,18]. In their retrospective study of 40 patients with nasal LCH, Puxeddu et al. used pre-operative angiography and embolization in 2 patients. Though no preoperative embolization was performed in the remaining patients, no blood transfusion was required. This led to the suggestion that no embolization was required even for large lesions [10]. Tzu-hang et al. did not use pre-operative embolization in 15 patients [13]. Successful surgical outcomes were reported when resection of the lesion was carried out along the sub-perichondral or the subperiosteum plane with a margin of healthy mucosa. Although some retrospective studies reported no recurrence, others reported a recurrence rate of 42 % with no malignant transformation potential [19]. In a retrospective cohort study, Sireci et al. proposed a management approach for nasal septum tumors based on their histology and types. In their review, 32 patients with nasal septum tumors were included. Large tumors, more than 2 mm, were biopsied under local anesthesia and then excised endoscopically under general anesthesia. In cases of benign nasal septum tumors, LCH tumors were resected with only the mucoperichondrium whereas other benign tumors included resection of the bone or cartilage in addition. In cases of



Fig. 5. Mass after excision.

malignant tumors of nasal septum, all layers of nasal septum were excised along with other adjuvant treatment based on disease extent and histopathology [20].

4. Conclusion

Nasal lobular capillary hemangioma shares common presentation with other sinonasal neoplasms. Its rarity and the lack of established guidelines represent diagnosis and management challenges for the treating physicians. Role of pre-operative embolization is debatable and no available high evidence research in the literature. We believe that use

of pre-operative embolization for large nasal lobular capillary hemangioma would have an impact on perioperative morbidity.

Consent

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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2. Unique identifying number or registration ID:
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Guarantor

Dr. Meshal B Albeshar.

CRediT authorship contribution statement

Dr. Meshal B Albeshar: writing, literature review, review, corresponding and first author.

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Dr. Mohammad Badr Alsumairi: writing, literature review, review.

Dr. Nedhal Muhammed Hussein: writing, literature review, review.

Declaration of competing interest

None.

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