Case report

Lobular capillary haemangioma of nasal septum with remodelling of bony lateral wall

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ABSTRACT

The diagnosis of intranasal masses can often be confusing due to the diversity of inflammatory and neoplastic lesions affecting the sinonasal tract, and the ambiguity of symptoms. Haemangiomas have been described in head and neck. However, uncommon findings such as bony remodelling and unusual clinical appearance of mass were noted in the present case. The description of such non-classical presentations is essential in deciding further management and preventing plausible hemodynamic imbalance.

Keywords: Lobular capillary haemangioma; pyogenic granuloma; nasal septum; epistaxis; nasal neoplasms

INTRODUCTION

S inonasal lobular capillary haemangioma (LCH) is infrequent, classically arising from anterior nasal septal mucosa (1). The diagnosis can often be confusing due to the diversity of inflammatory and neoplastic lesions affecting the sinonasal tract, and the ambiguity of symptoms; the differentials include inverted papilloma, angiofibroma, angiomatous polyp, and hemangiopericytoma (1). Imaging techniques play a vital role in such cases; however, definitive diagnosis is usually histopathological. We report a case of lobular capillary haemangioma (LCH) of nasal cavity arising from anterior part of septum.

Case report

A 52-year-old lady presented with left-sided nasal bleeding for four months, followed by nasal obstruction for three months and nasal mass for two months. Nasal bleed was sudden, spontaneous, intermittent, with no history of local trauma or forceful sneezing. There were 2-3 episodes per week, each episode consisting of frank blood of around 10-15 ml; bleeding subsided with local pressure and topical medications. Nasal obstruction was gradually progressive, left-sided, occasionally associated with mucoid discharge, following which a painless mass appeared in the left nasal cavity. Within two weeks, a diffuse, painless, slowly progressive swelling appeared over left side of nasal dorsum.

There was no history suggestive of sinusitis, allergy, ocular or aural disease; she was postmenopausal, without comorbidities. She had undergone a lesional biopsy elsewhere, following which pain and excessive bleeding occurred, for which nasal packing was done.

Examination revealed a diffuse 2x1 cm swelling over left side of dorsum, obliterating nasomaxillary groove laterally (Fig. 1). It was non-tender, hard, with no local rise of temperature, not involving orbit or superficial skin. Right-sided nasal dorsum deviation was present. Anterior rhinoscopy revealed a single, pinkish-white, smooth, globular, mucosa covered mass reaching the vestibule causing complete left nasal cavity obliteration (Fig. 2). Posterior extent and site of attachment could not be assessed. Septum was pushed to the right with gross cartilaginous deviation. Posterior rhinoscopy and rest of examination were normal.



Fig. 1: Clinical photograph of lesion obliterating left nasomaxillary groove (white arrow)



Fig. 2: Clinical photograph of anterior rhinoscopic examination showing pale white mass (black arrow) in the left nasal cavity extending up to the vestibule

Plain computed tomography (CT) scan performed at another hospital a month ago showed a well-

circumscribed mass in anterior part of nasal cavity with no bony destruction. Contrast-enhanced CT (CECT) was done; an enhancing soft tissue mass of 3.2x1.4 cm occupying anterior part of left nasal cavity in front of inferior turbinate, obscuring anterior part of nasal septum, causing disruption and splaying of frontal process of maxilla on left side was seen (Fig. 3).



Fig. 3: Axial section of contrast-enhanced CT scan showing mild heterogeneously enhancing soft tissue mass (long white arrow) in the anterior part of the left nasal cavity, and erosion or splaying of the nasal bones on the left side (short white arrow)

Biopsy was done; histopathology showed bits of tissue lined by respiratory epithelium with ulceration covered by acute inflammatory exudate and underlying granulation tissue. The lesion was composed of proliferating capillaries with presence of plump reactive endothelial and stromal cells (Fig. 4). Immunohistochemistry performed to rule out mesenchymal tumours showed CD 34 (Fig. 5) in lobular pattern of endothelial cells and positive smooth muscle actin (SMA) in capillary wall, MIB-1 index of 10% suggesting a diagnosis of lobular capillary haemangioma. CD117 was weakly positive, vimentin was positive, and beta-catenin was negative in endothelial and stromal cells, excluding the possibility of other mesenchymal tumours. However, because of bony destruction and remodelling noted on CT imaging, a differential was angiofibroma could not be ruled out.



Fig. 4: Histopathology showing lobular architecture of proliferating capillaries with central ectatic feeding vessels surrounded by mixed inflammatory infiltrate



Fig. 5: Immunohistochemistry with CD34 highlighting the endothelial cells

Transnasal endoscopic excision was performed; Left septal mucoperichondrial flap was elevated; mass was removed in-toto with 1 cm margin of normal mucoperichondrium. No involvement of cartilaginous or posterior part of septum or lateral nasal wall was seen. The external swelling resulted from splaying of frontal process of maxilla due to mass effect. The mucoperichondrial defect was allowed to heal with secondary intention; post-operative period was uneventful, nasal packs were removed after a day. The final histopathology showed a lesion lined by respiratory epithelium composed of an ectatic feeding blood vessel surrounded by small-sized capillaries arranged in lobular pattern with mixed inflammatory infiltrate with stroma showing myxoid change and collagenisation, diagnostic of LCH.

The patient is under regular follow-up, with no evidence of recurrence one year after surgery.

DISCUSSION

Haemangiomas are benign vascular tumours composed of blood vessels and connective tissue. While head and neck is a frequent site of occurrence (12.2%), they less commonly affect the sinonasal tract (2.5%) (2). They are classified as cavernous, capillary, mixed based on the histopathological (3). Lobular capillary haemangioma is a type of haemangioma first described by Poncet and Dor in 1897 due to fungal infection by Botryomycosis hominis, followed by Frank and Blahd, who called it pyogenic granuloma in 1940 (4.5). It was later found to be neither granulomatous nor infectious; hence the term pyogenic granuloma was disregarded and termed lobular capillary haemangioma by Mills in 1980 due to the lobular arrangement of capillaries encompassing angulated feeding vessels without endothelial atypia (4,5). Plausible etiological factors include local trauma, viral oncogenes, hormones, angiogenic growth factors, arteriovenous malformations and hypertension (2,6).

They most commonly affect women in the third decade of life (3,6). Typical clinical features include nasal bleed, nasal obstruction and nasal discharge (6). Sites of involvement in the nasal cavity in order of

decreasing frequency are nasal septum (55%), nasal vestibule (17%), inferior turbinate (12.5%), middle turbinate and uncinate process (7.5% each) (2,6). They appear as vascular, reddish, polypoidal, painless masses with contrast enhancement. On magnetic resonance imaging, they are homogenous isointense on T1, heterogenous hyperintense on T2 with thin peripheral hypointense ring and flow voids (7).

Histopathologically they are characterised by widespread endothelial proliferation and conspicuous vascular spaces, lobular arrangement of capillaries, fibrovascular tissue base, epithelial ulceration, and surrounding inflammatory infiltrate of neutrophils, lymphocytes, plasma cells and eosinophils (4,8). Immunohistochemistry shows CD31 positivity in endothelial cells and smooth muscle actin (SMA) positivity in the smooth muscle cells (4). Treatment is wide surgical resection via endoscopic or external approach, with or without embolisation, with good prognosis and 15.8% recurrence (9).

The present case was not very characteristic of LCH; inverted papilloma, angiomatous polyp and angiofibroma were considered differentials. Female gender, presenting complaint of epistaxis, and involvement only of anterior part of nasal cavity favoured a diagnosis of LCH. Whereas, absence of risk factors such as history of trauma, postmenopausal patient with pale white mass lacking features of vascular lesion clinically and radiologically negated the diagnosis.

Bony remodelling was also noted, which has not been reported in such cases to the best of our knowledge; it is more likely to occur in large LCHs (1). Studies show that bony remodelling depends more on tumour size than histological type (7). A pale, rapidly growing mass with associated external swelling and bony remodelling made it difficult to clinically differentiate the present case from more aggressive and malignant tumours. Despite the vascular nature, there was no need for preoperative embolisation or blood transfusion.

CONCLUSION

Lobular capillary haemangioma is an uncommon nasal cavity tumour, often confused with more aggressive lesions due to its abrupt growth; differential diagnosis of LCH must be kept in mind while dealing with rapidly progressive vascular nasal masses, even in the absence of risk factors.

CONFLICT OF INTEREST

Authors declare that there is no conflict of interest.

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