Case Report

A rare case of pediatric intranasal lobular capillary hemangioma

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ABSTRACT

Pediatric nasal cavity vascular tumors express a wide variety of pathologies. Lobular capillary hemangioma (LCH) is an acquired benign vascular growth of skin and mucosa whose etiology remains unknown, though trauma and hormonal influences are implicated. Although well documented in the head and neck literature for children age five or less, it is a rarity within the nasal cavity and has yet to be documented in the mid-septum. We describe a unique case of intranasal LCH and review the current literature. A nine-year-old male presented with one week of profuse intermittent unilateral epistaxis and no history of nasal trauma. Rhinoscopy revealed a pink, pedunculated mass of the right mid-nasal septum at the bony-cartilaginous junction. CT and MRI imaging were consistent with an expansile vascular lesion receiving prominent bilateral sphenopalatine artery supply. Following embolization, en bloc endoscopic surgical excision of the lesion using cold dissection was performed with no bony or cartilaginous involvement noted. The epistaxis resolved following resection. Final histology confirmed the mass as a lobular capillary hemangioma. Paediatric intranasal LCH is a rare entity, yet warrants consideration in our differential diagnosis of pediatric vascular tumors. Our study indicates these lesions can develop in the mid-septum despite the absence of a vascular plexus. Preoperative embolization should be considered for pediatric nasal cavity tumors due to concern for hemorrhage. Endoscopic wide local excision is an appropriate and effective treatment.

Keywords: Pediatric nasal masses, Epistaxis, Embolization, Lobular capillary hemangioma, Pyogenic granuloma

INTRODUCTION

Pediatric nasal cavity masses pose a diagnostic challenge. Adequate differentials must encompass a spectrum of vascular, neural, and soft tissue lesions such as dermoid cyst, meningocoele, encephalocele, angiomatous polyp, vascular malformation, juvenile nasal angiofibroma (JNA), glioma, and benign or malignant neoplasm.1,2 History and physical, though critical, are often insufficient to establish a diagnosis. MRI and/or CT are essential, not only to characterize the lesion and thereby guide diagnosis, but also to identify intracranial extension, bony erosion, and vascularity whose presence significantly impacts treatment.1 Lobular capillary hemangioma (LCH), originally named ‘human botryomycosis’ and also known as pyogenic granuloma (PG), is an acquired benign vascular growth of skin and mucosa. The etiology of LCH remains unclear, though traumatic, hormonal, infectious, and congenital causes are all proposed.1,3 There is a well-established relationship with pregnancy.3 Although this lesion is well described in the head and neck literature for children aged five or less, it is a rarity within the nasal cavity and has yet to be documented in the mid-septum.4 We describe the diagnosis and management of a unique case of intranasal LCH.
CASE REPORT

A nine-year-old male with one week of acute onset profuse intermittent unilateral epistaxis and associated nasal obstruction was referred for evaluation. No history of nasal trauma and no known personal or familial history of bleeding diathesis were elucidated. A complete 12-system review of systems was otherwise negative. Exam was significant for copious and persistent bleeding from a pink, pedunculated mass obstructing the right nasal cavity. After conservative measures failed, the patient was sent to the Emergency Department for stabilization, urgent imaging, and further work-up.

CT and MRI imaging identified an expansile highly vascular lesion with a prominent bilateral sphenopalatine arterial supply and without intracranial abnormalities or bony erosion (Figure 1) and (Figure 2). Following multidisciplinary review, tissue diagnosis was deemed necessary to guide appropriate treatment. All teams acknowledged the potential risk for high volume blood loss given the friability of the lesion and the volume of bleeding previously observed. In a shared discussion with the family, the decision was made to perform preoperative embolization.

Four-vessel diagnostic cerebral angiography was performed successfully with intra-arterial embospheres and Onyx embolization of the right distal internal maxillary vessels. A platinum coil and Onyx plug was used in the left distal internal maxillary branches to prevent excessive soft tissue de-vascularization on the contralateral side (Figure 3).

The day following embolization, en bloc endoscopic excision of the lesion was performed using cold dissection with elevation of mucoperichondrial and mucoperiosteal flaps and minimal blood loss (Figure 4). The lesion was noted to originate in the mid septum at the bony-cartilaginous junction without bony or cartilaginous involvement. Minimal electrocautery was used. Final histology demonstrated lobular capillary hemangioma. At 12-month follow up, the patient remains without epistaxis or signs of regrowth.

DISCUSSION

Intranasal LCH generally presents with recurrent epistaxis, nasal obstruction, rhinorrhea, and pain. There are only 19 prior reported cases in the pediatric literature with five localized to the anterior nasal septum and none described at the bony-cartilaginous junction (Table 1). In the head and neck, LCH more commonly occurs on the skin or the mucosa of the oral cavity including the lips. Lesions in the anterior septum are commonly associated with little’s area. Histologically, the lesion classically has two distinct areas: a lobular region with capillary proliferation and an ulcerative region with inflammatory granulation tissue beneath an ulcer with neutrophilic infiltrates and irregularly dilated blood vessels. Radiographically, contrast-enhanced CT usually shows intense diffuse enhancement with a varied pattern of a well-circumscribed soft tissue mass without intrinsic calcification.
Table 1: Literature review of pediatric intranasal lobular capillary hemangioma.

| Study                  | Age  | Gender | Anatomic Origin          | Imaging Study | Treatment                        |
|------------------------|------|--------|--------------------------|---------------|----------------------------------|
| Mills SE et al         | 10   | Female | Septum                   | None          | Endoscopic excision              |
| Patrice SJ et al       | NR   | NR     | Nasal mucosa             | NR            | Endoscopic excision              |
| Simo R et al           | 7    | Male   | Right lateral wall       | NR            | Endoscopic excision              |
| Ogunleye and Nwaogu    | 45   | Male   | Roof of the left nasal cavity | CT           | Endoscopic excision              |
| Kapella et al          | 7    | Female | Left vestibule           | CT            | Endoscopic excision              |
| Karagama et al         | 8    | Male   | Left floor               | None          | Elliptical incision; 4/0 Vicryl stitches |
| Ozcan et al            | 6    | Female | Right floor              | CT            | Endoscopic excision              |
| Katori and Trukuda     | 11   | Male   | Right lateral wall       | CT and MRI    | Elliptical incision w/Nd Yag laser |
| Puxeddu et al          | NR   | NR     | NR                       | CT            | Endoscopic excision              |
| Benoit et al           | 5    | Male   | Right septum             | Unspecified   | Endoscopic excision              |
| Berlucchi et al        | 5    | Male   | Left septum              | MRI           | Endoscopic excision              |
| Ifeacho and Caulfield  | 14   | Male   | Right middle turbinate   | MRI           | Endoscopic excision              |
| Virbalas et al         | 12   | Female | Left lateral wall        | CT            | Endoscopic excision              |
| Virbalas et al         | 16   | Female | Right middle meatus      | CT            | Endoscopic excision              |
| Vijaya FA et al        | 14   | Male   | Left septum              | CT            | Endoscopic excision              |
| Marino-Sanchez et al   | 13   | Male   | Right inferior turbinate | CT            | Endoscopic excision              |
| Marino-Sanchez et al   | 12   | Female | Right septum             | CT            | Endoscopic excision              |
| Yildirium et al        | 9    | Male   | Posterior 1/3 right septum | CT           | Endoscopic excision              |
| Case 1                 | 9    | Male   | Right Mid-septum         | CT and MRI    | Preoperative embolization; Endoscopic excision |

MRI shows marked enhancement and is imperative to rule out intracranial extension of pediatric intranasal masses.

Preoperative embolization has occasionally been utilized for LCH but more commonly plays a role in other lesions such as JNA. In cases such as this one, where a large volume of bleeding is observed and the diagnosis is unclear, it can serve as a useful adjunct to prevent massive hemorrhage during surgical excision or biopsy. Tamaki et al notes preoperative embolization has made an otherwise unresectable LCH tumor resectable. Complications of embolization are rare but include soft tissue necrosis, cranial neuropathy, and stroke or blindness. The majority of reported cases underwent endoscopic surgical excision either through electrocoagulation, cryotherapy, LASER, excisional surgery, or excisional surgery following embolization. Recurrence rates range from 0-42% in various series.

This case report raises several interesting questions in the management of pediatric intranasal vascular tumors, specifically LCH. The fact that this lesion arose in the mid-septum where there is no vascular plexus and is less accessible to digital trauma lends support to the other proposed etiologies. Moreover, the role of preoperative embolization, especially in pediatric patients, is a somewhat contentious topic given the morbidity of any complications. In this case, it worked exceptionally well in providing an optimal surgical environment for a lesion where previously, hemostasis had been quite challenging.

**CONCLUSION**

Pediatric intranasal LCH is rare, yet warrants consideration in our differential diagnosis of pediatric vascular tumors. This case shows that LCH can develop in the mid-septum despite the absence of a vascular plexus. Preoperative embolization should be considered for pediatric nasal vascular neoplasms with high risk of hemorrhage. Endoscopic en-bloc excision is an appropriate and effective treatment.
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