International Journal of Otorhinolaryngology and Head and Neck Surgery
Schild SD et al. Int J Otorhinolaryngol Head Neck Surg. 2020 Nov;6(11):2115-2118
http://www.ijorl.com
pISSN 2454-5929 | eISSN 2454-5937

DOI: https://dx.doi.org/10.18203/issn.2454-5929.ijohns20204641

Case Report

A rare case of pediatric intranasal lobular capillary hemangioma

Sam D. Schild*, Rachel Irizarry, Ann Plum

Department of Otolaryngology, State University of New York, Downstate Health Sciences University, Brooklyn, NY, USA

Received: 20 May 2020
Accepted: 18 September 2020

***Correspondence:**
Dr. Sam D. Schild,
E-mail: sam.schild@downstate.edu

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, meningocele, 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.

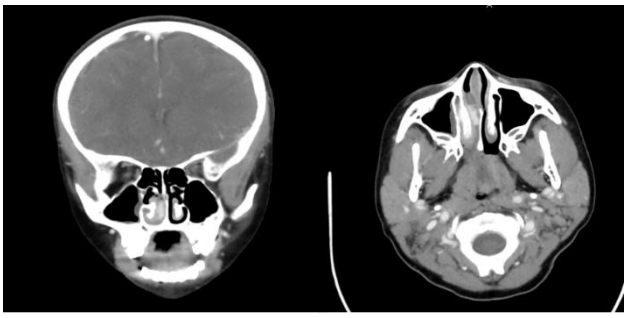

Figure 1: CT head and neck with contrast.

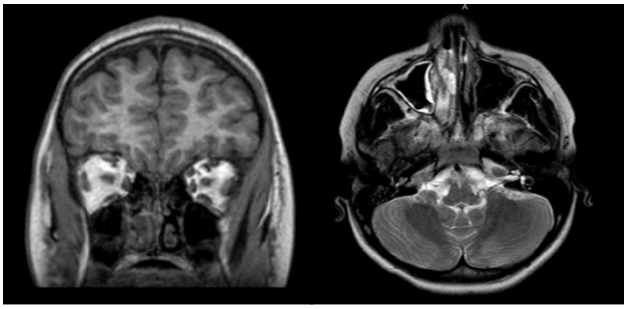

Figure 2: MRI head and neck with contrast.

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).

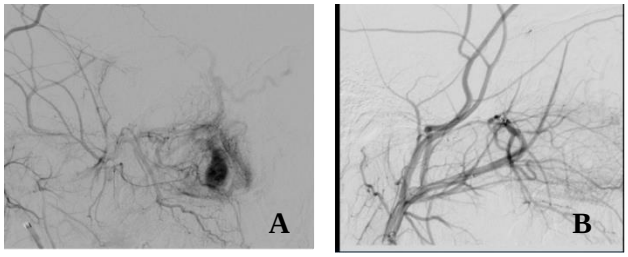

Figure 3: Angiography and embolization (A) Pre-embolization (B) Post-embolization.

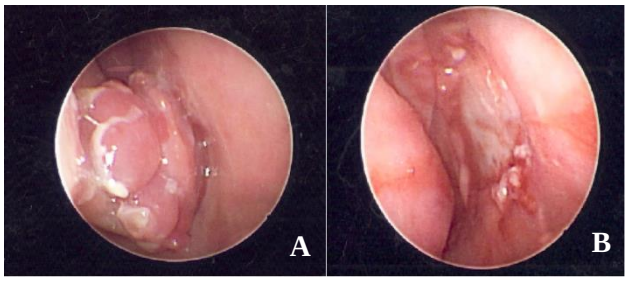

Figure 4: Intraoperative images (A) Pre-excision (B) Post-excision

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.[2] 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).[2-17] In the head and neck, LCH more commonly occurs on the skin or the mucosa of the oral cavity including the lips.3 The anterior nasal septum and tip of the inferior turbinate are the two most common intranasal locations lending support to the trauma etiology theory.[3] Lesions in the anterior septum are commonly associated with little's area.[2] 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.[3] Radiographically, contrast-enhanced CT usually shows intense diffuse enhancement with a varied pattern of a well-circums cribed soft tissue mass without intrinsic calcification.[2]

Table 1: Literature review of pediatric intranasal lobular capillary hemangioma.

Study	Age	Gender	Anatomic Origin	Imaging Study	Treatment
Mills SE et al	10 years	Female	Septum	None	Endoscopic excision
Patrice SJ et al	NR	NR	Nasal mucosa	NR	NR
Simo R et al	7 years	Male	Right lateral wall	NR	Endoscopic excision
Ogunleye and Nwaorgu	45 days	Male	Roof of the left nasal cavity	CT	Endoscopic excision
Kapella et al	7 years	Female	Left vestibule	CT	Endoscopic excision
Karagama et al	8 years	Male	Left floor	None	Elliptical incision; 4/0 Vicryl stitches
Ozcan et al	6 years	Female	Right floor	CT	Endoscopic excision
Katori and Trukuda	11 years	Male	Right lateral wall	CT and MRI	Ellipitical incision w/Nd Yag laser
Puxeddu et al	NR	NR	NR	CT	Endoscopic excision
Puxeddu et al	NR	NR	NR	CT	Endoscopic excision
Benoit et al	5 years	Male	Right septum	Unspecified	Endoscopic excision
Berlucchi et al	5 months	Male	Left septum	MRI	Endoscopic excision
Ifeacho and Caulfield	14 years	Male	Right middle turbinate	MRI	Endoscopic excision
Virbalas et al	12 years	Female	Left lateral wall	CT	Endoscopic excision
Virbalas et al	16 years	Female	Right middle meatus	CT	Endoscopic excision
Vijaya FA et al	14 years	Male	Left septum	CT	Endoscopic excision
Marino-Sanchez et al	13 years	Male	Right inferior turbinate	CT	Endoscopic excision
Marino-Sanchez et al	12 years	Female	Right septum	CT	Endoscopic excision
Yildirium et al	9 years	Male	Posterior 1/3 right septum	CT	Endoscopic excision
Case 1	9 years	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.[3,18] 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.18 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 emboli zation.[1,18] Recurrence rates range from 0-42% in various series.[1,3,18]

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.

Funding: No funding sources
Conflict of interest: None declared
Ethical approval: Not required

REFERENCES

1. Rodriguez DP, Orscheln ES, Koch BL. Masses of the Nose, Nasal Cavity, and Nasopharynx in Children. Radiographics: a review publication of the Radiol Soc Nor Americ, Inc. 2017;37(6):1704-30.
2. Virbalas JM, Bent JP, Parikh SR. Pediatric nasal lobular capillary hemangioma. Cas Repor Medic. 2012;2012:769630.
3. Puxeddu R, Berlucchi M, Ledda GP, Parodo G, Farina D, Nicolai P. Lobular capillary hemangioma of the nasal cavity: A retrospective study on 40 patients. Americ J Rhinol. 2006;20(4):480-4.
4. Marino-Sanchez F, Lopez-Chacon M, Jou C, Haag O. Pediatric intranasal lobular capillary heman gioma: Report of two new cases and review of the literature. Respirat Medic Cas Report. 2016;18: 31-4.
5. Mills SE, Cooper PH, Fechner RE. Lobular capillary hemangioma: the underlying lesion of pyogenic granuloma. A study of 73 cases from the oral and nasal mucous membranes. Americ J Surgic Pathol. 1980;4(5):470-9.
6. Patrice SJ, Wiss K, Mulliken JB. Pyogenic granuloma (lobular capillary hemangioma): a clinicopathologic study of 178 cases. Pediatric dermatol. 1991;8(4):267-76.
7. Simo R, de Carpentier J, Rejali D, Gunawardena WJ. Paediatric pyogenic granuloma presenting as a unilateral nasal polyp. Rhinol. 1998;36(3):136-8.
8. Ogunleye AO, Nwaorgu OG. Pyogenic granuloma, a cause of congenital nasal mass: case report. Anna Tropic Paediat. 2000;20(2):137-9.
9. Kapella M, Panosetti E, Rombaux P, Delos M, Weynand B. Lobular capillary haemangioma of the nasal cavity: observation of three specific cases. Acta Oto Rhino Laryngolog Belgica. 2001;55(3): 241-6.
10. Karagama YG, Howarth K, Steel PR, Spencer MG. Lobular capillary haemangioma of the nasal vestibule: a rare entity. Int J Pediat Otorhinolaryngol. 2002;66(1):71-5.
11. Ozcan C, Apa DD, Gorur K. Pediatric lobular capillary hemangioma of the nasal cavity. European archives of oto-rhino-laryngology. J Europ Federat Hea Nec Surg. 2004;261(8):449-51.
12. Katori H, Tsukuda M. Lobular capillary hemangioma of the nasal cavity in child. Auris, nasus, laryn. 2005;32(2):185-8.
13. Benoit MM, Fink DS, Brigger MT, Keamy DG, Jr. Lobular capillary hemangioma of the nasal cavity in a five-year-old boy. Otolaryngol Hea Nec Surg. 2010;142(2):290-1.
14. Berlucchi M, Pedruzzi B, Farina D. Radiology quiz case 2. Lobular capillary hemangioma (LCH). Archiv Otolaryngol Hea Nec Surg. 2010;136(11): 1141-3.
15. Ifeacho SN, Caulfield HM. A rare cause of paediatric epistaxis: lobular capillary haemangioma of the nasal cavity. Brit Med J case reports. 2011;2011.
16. M. Vijaya FA, G. Umarani, A. Sultan, Lobular capillary hemangioma of, nasal septum - a rare presentation Ie.
17. Yildirim U, Karli R, Gun S. Pediatric Intranasal Lobular Capillary Hemangioma: A Rare Clinical Entity. Balk Medic J. 2017;34(6):586-7.
18. Tamaki A, Babajanian E, D'Anza B, Rodriguez K. Lobular capillary hemangiomas: Case report and review of literature of vascular lesions of the nasal cavity. Americ J Otolaryngol. 2017;38(3):363-6.

Cite this article as: Schild SD, Irizarry R, Plum A. A rare case of paediatric intranasal lobular capillary hemangioma. Int J Otorhinolaryngol Head Neck Surg 2020;6:2115-8.