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Recurrent Epistaxis and Unilateral Intranasal Mass in A Teenager

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Data Interpretation D
Manuscript Preparation E
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Patient: Male, 16-year-old
Final Diagnosis: Lobular capillary hemangioma
Symptoms: Epistaxis
Medication: —
Clinical Procedure: —
Specialty: Otolaryngology

Objective: Rare disease

Background: Epistaxis in children is a common problem encountered in outpatient clinics and emergency departments. A wide variety of conditions may cause recurrent epistaxis in children. We describe clinical, radiologic, and histologic features of a lobular capillary hemangioma presenting as a rapidly growing intranasal mass in a child with recurrent epistaxis.

Case Report: A 16-year-old male presented with a 2-month history of recurrent unilateral epistaxis requiring multiple visits to the emergency department. The child had nasal obstruction, snoring, no recurrent sinus infections, no anosmia nor hyposmia, no weight loss, no night sweats, no fever, no decreased activity, and no easy bruising. He denied any history of local trauma. On physical examination, a fleshy violaceous mass was found, protruding from and obliterating the right nasal cavity. Magnetic resonance imaging documented an avidly enhancing mass centered at the right nasal vestibule. Upon resection, histologic evaluation indicated a pyogenic granuloma. At the 2-month followup, the surgical site was healed with no evidence of recurrent lesion.

Conclusions: Lobular capillary hemangioma, although uncommon, should be considered in the differential diagnosis of recurrent epistaxis and intranasal mass in children.

Keywords: Child • Epistaxis • Granuloma, Pyogenic

Full-text PDF: https://www.amjcaserep.com/abstract/index/idArt/933075

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Background

Epistaxis in children is a common complaint encountered in outpatient clinics and emergency departments. The incidence of epistaxis is highest in patients who are less than 10 years old and greater than 70 years old [1-3]. While up to 60% of children experience at least 1 episode of epistaxis, recurrent nosebleeds occur in 9% of children [4,5]. The majority of epistaxis in children is self-resolving without intervention; however, timely identification of the cause of epistaxis is critical in children with recurrent or severe epistaxis.

A wide variety of local, systemic, and environmental factors as well as medications may cause epistaxis. Trauma, infection, inflammation, structural abnormality, and neoplasm are common local factors. Systemic factors such as hypertension, coagulopathy, and hemorrhagic telangiectasia should be included in the differential diagnosis. Environmental factors such as temperature and humidity or medications such as warfarin, aspirin, and nonsteroidal anti-inflammatory drugs can be contributing factors. Severe or recurrent epistaxis associated with a history of nasal obstruction among children and adolescents should prompt consideration of a more serious underlying etiology, such as a neoplasm [6]. We describe clinical, radiologic, and histological features of an intranasal lobular capillary hemangioma (LCH) presenting as a rapidly growing intranasal mass in a child with recurrent epistaxis.

Case Report

A 16-year-old male with no past medical history presented with a history of recurrent epistaxis. Epistaxis from the right nasal cavity began 2 months prior to clinical presentation. Epistaxis occurred 1-2 times daily and lasted 3 to 5 min. Applying direct pressure by pinching the nose over the cartilaginous tip for a few minutes controlled the bleeding. However, the frequency and duration of the epistaxis gradually increased over time. The patient went to several emergency departments for nosebleeds. Application of external pressure and oxymetazoline controlled the bleeding and eliminated the need for nasal packing. He also developed a post-nasal drip of blood that led to episodic nocturnal choking that would wake him up from sleep. At the last emergency department visit, a mass in the right nasal cavity was noted.

The adolescent had nasal obstruction and snoring. He did not have recurrent sinus infections, anosmia, hyposmia, weight loss, night sweats, fever, decreased activity, or easy bruising. He denied any history of local trauma. There was no family history of childhood cancers. On physical examination, a fleshy violaceous mass was seen protruding from and obliterating the right nasal cavity (**Figure 1**). The mass was non-tender and mobile.

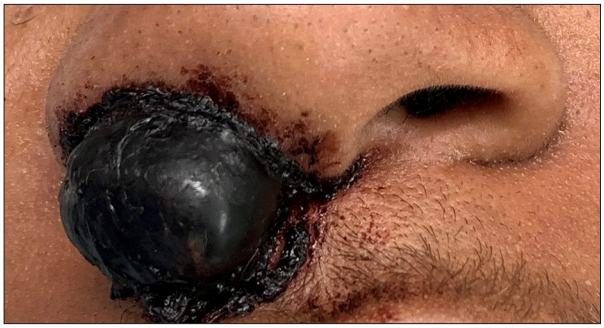

Figure 1. A fleshy violaceous mass protruding from and obliterating the right nasal cavity. (*Microsoft Photos, Version 2020.20120.4004.0, Microsoft*).

Magnetic resonance imaging (MRI) demonstrated an avidly enhancing mass centered at the right nasal vestibule (**Figure 2**). The mass was closely associated with the nasal septum and lateral nasal wall, and displaced adjacent mucosa without evidence of gross invasion. A hypointense rim around the mass was noted. A pedunculated mass arising from the right anterior nasal septum was excised via endoscopic approach. The patient recovered well during the immediate postoperative period. He experienced no recurrent epistaxis and was subsequently discharged at day 1 postoperatively.

A 4.5×2.8×1.3 cm polypoid mass with a dark red to brown mucosal surface and focal fibrinous exudate was received in pathology. Serial sectioning showed a dark red to tan brown spongy cut surface with strands of fibrous bands (**Figure 3A**). Histologic evaluation showed proliferating vessels in a lobular pattern admixed with larger vessels. The mass was lined by respiratory mucosa with surface ulceration. Within the tumor, there was prominent granulation tissue, edema, and acute inflammatory infiltrate as well as areas of intraparenchymal hemorrhage (**Figure 3B-3D**). The final diagnosis was LCH. The patient was followed up at 2 months. He denied any episodes of epistaxis or nasal obstruction and the surgical site was healed with no evidence of recurrent lesion.

Discussion

LCH is a benign vascular lesion of unclear etiology that usually affects the skin and mucous membranes of the oral cavity. The underlying cause of LCH is thought to be a localized, exuberant inflammatory response most commonly occurring in response to trauma or hormonal stimulation, such as pregnancy or hormone therapy [7]. The nasal cavity is an infrequent site of origin, and when present is typically seen in the anterior portion of the nasal septum, though LCH has also been demonstrated at the posterior nasal septum, inferior turbinate, vestibule, and middle turbinate [8,9]. Lesions can affect individuals of all ages, with no preference with regard to sex.

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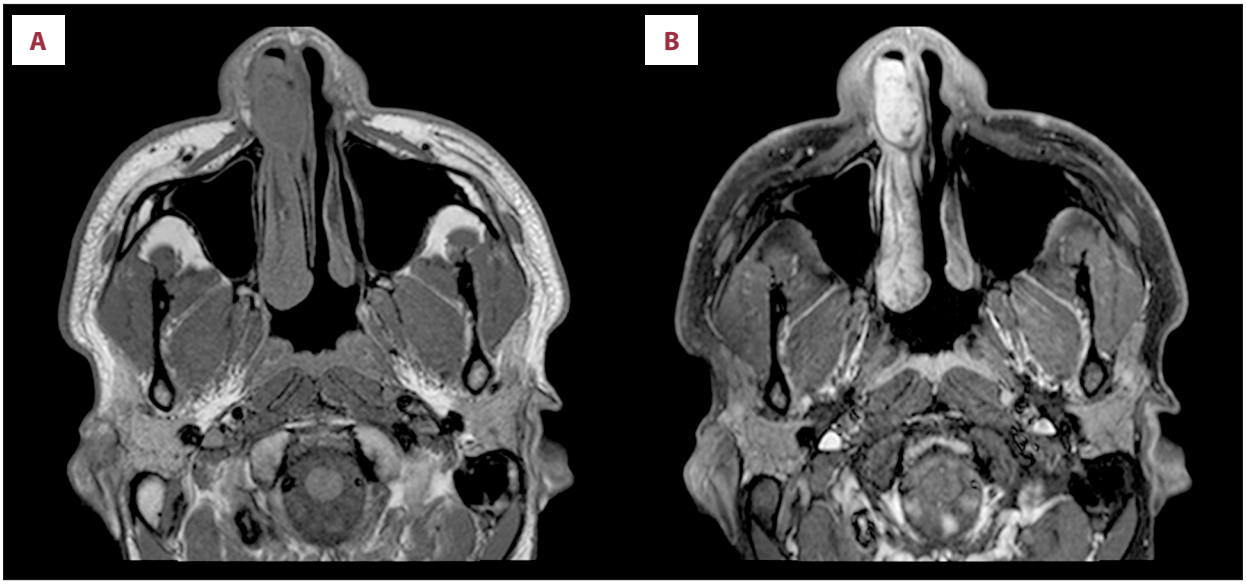

Figure 2. Axial T1-weighted pre contrast (**A**) and post contrast (**B**) images show a mass centered at the right nasal vestibule intimately associated with the nasal septum and lateral nasal wall. Post-contrast administration, the mass showed enhancement.

Prior local trauma is also frequently reported; however, it is not considered necessary for development of LCH.

Patients with nasal LCH typically present with recurrent epistaxis, though congestion, nasal obstruction, and pain are common. Manual or endoscopic visualization reveals a mobile, hypervascular polypoid mass with a red to purple, friable surface with or without superficial ulceration. Lesions frequently bleed when manipulated. Differential diagnoses for recurrent epistaxis associated with an intranasal mass in a teenager include benign tumors such as choanal polyps, angiofibromas, angiomatous polyps, capillary hemangiomas, paragangliomas, hemangiopericytomas, and sinonasal papillomas as well as malignant neoplasms such as nasopharyngeal carcinoma, nasopharyngeal teratoma, angiosarcoma, and Kaposi's sarcoma [10-12]. Inflammatory conditions such as sarcoidosis and granulomatosis with polyangiitis are possible, though typically display more destructive features [13].

Imaging of LCH shows a well-defined mass with soft tissue attenuation. Since they are highly vascular, these lesions demonstrate avid, though diffuse, arterial enhancement. Because they do not have large draining veins, they typically display somewhat delayed washout. Bony destruction is rare [14]. MRI shows T1 hypointensity with possible flow voids and heterogeneous hyperintensity on T2 with a hypointense rim [14]. Masses enhance intensely with gadolinium.

Histologically, LCH appear as capillaries of varying sizes in a lobular arrangement with fibrous septa. Vessels range from tightly packed with scant lumens to larger dilated cavernous spaces. The septa typically contain scattered inflammatory cells while the lobules are often surrounded by a loose spindle cell proliferation of pericytic cells [9,10,15]. Ulceration or atrophy of the overlying stratified squamous epithelium is common.

A wide variety of treatment options, including but not limited to, intralesional corticosteroid injection, laser therapy, cryotherapy, and sclerotherapy, have been suggested; however, the lack of availability of a specimen for histopathological examination, an increased risk for recurrence, and the need for repeated treatments have limited the broad use of these treatment choices. Definitive management involves local excision. Paranasal endoscopic techniques are the most common and may be accompanied by electrocautery or absorbable hemostatic agents [9]. Complete excision with a margin of normal mucosa is recommended to prevent recurrence. Larger lesions have typically required open craniofacial excision due to concerns for bleeding. Preoperative embolization can be an attractive option for particularly hemorrhagic lesions. Recurrence rates are as high as 40% for those previously biopsied or treated with electrocauterization [9,16]. Whether this reflects a failure to completely excise the lesion or involves host factors prompting susceptibility to spontaneous cellular changes is unclear. The recurrence rate is 8% with an open excisional technique [7]. Additionally, in adolescents and especially in gravid women, lesions may spontaneously regress over a period of time, possibly reflecting resolution of the underlying inflammatory insult or hormonal drivers [17].

Counseling is a critical component of management of patients with LCH. Preoperative counseling should include discussion about the possibility of partial excision, depending on the size of the lesion, potential for recurrence, and need for additional

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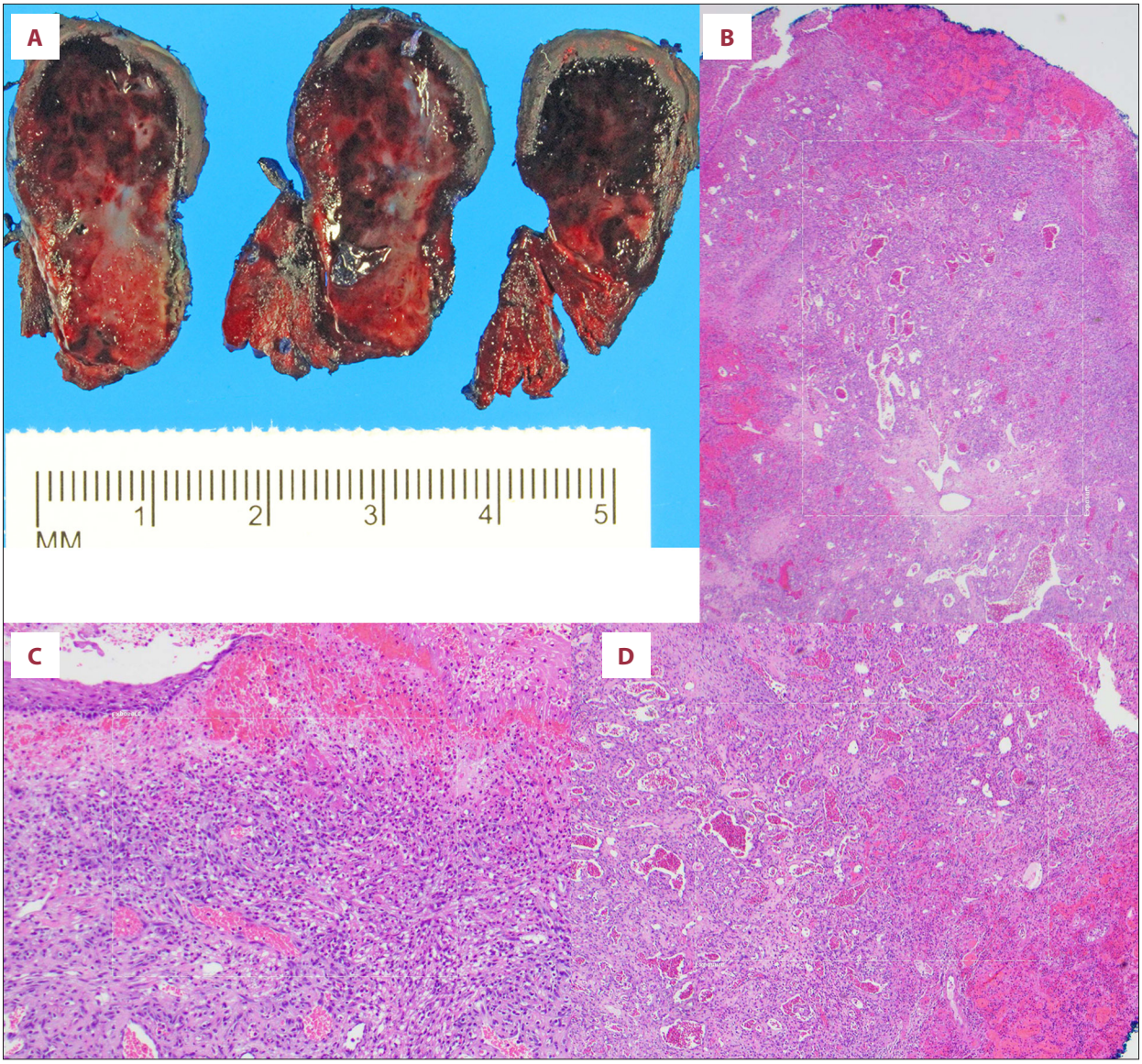

Figure 3. Sections of the mass showing tan-brown to dark red, hemorrhagic, spongy parenchyma with tan-white fibrous tissue (**A**). Low magnification showing superficial mucosal ulceration with hemorrhage and granulation tissue, deep lobular proliferation of capillaries with central dilated vessels, separated by fibrous bands (Hematoxylin and Eosin) (**B**). Higher magnification showing ulceration of respiratory mucosa with hemorrhage, granulation tissue, and inflammation, (Hematoxylin and Eosin) (**C**). Lobular proliferation of capillaries with central dilated larger vessels (Hematoxylin and Eosin) (**D**). (*Microsoft Photos, Version 2020.20120.4004.0, Microsoft*).

surgery, as well as the possibilities of scar formation and deformity. The role of local trauma as a cause of LCH needs to be discussed and patients should be advised to avoid trauma.

Conclusions

LCH may present in variety of ways. The present case suggests that, although very rare, intranasal LCH should be included in the differential diagnosis when children present with epistaxis and intranasal mass. Comprehensive evaluation along with early detection and treatment of intranasal LCH may prevent complications and contribute to satisfactory outcomes.

Declaration of Figures Authenticity

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