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Case report

Pediatric intranasal lobular capillary hemangioma: Report of two new cases and review of the literature

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ABSTRACT

Background: Lobular capillary hemangioma (LCH) is an acquired benign vascular tumor of unknown origin. It usually affects skin and mucous membranes of the oropharynx. It rarely involves the nasal cavity which most commonly manifests as epistaxis. To our knowledge, only fifteen pediatric intranasal LCH cases have been reported in the literature. None of these occurred in the inferior turbinate. We report two new pediatric cases of LCH, one of them on the inferior turbinate and the other one on the anterior nasal septum. Our principal aim was to highlight the importance of considering this lesion as a differential diagnosis for pediatric unilateral nasal obstruction and epistaxis.

Methods: Retrospective case series and review of current literature regarding the possible causes, diagnosis, and treatment of nasal LCH.

Description of cases: Two adolescents presented with symptoms of unilateral nasal obstruction and epistaxis. Plain and contrast enhanced computed tomography revealed a well-defined intensely enhancing lesion in both cases. Patients underwent transnasal endoscopic excision and bipolar electrocautery at the base of the tumor for hemostasis. Histopathological examination confirmed the diagnosis of LCH.

Discussion: Current epidemiological and pathophysiological data suggests that the development of LCH may be associated to previous nasal trauma or endocrine disorders. LCH should be considered in the differential diagnosis of all pediatric endonasal masses associated with unilateral epistaxis and nasal obstruction. Endoscopic total excision with bipolar electrocautery for hemostasis is an appropriate treatment.

1. Introduction

Lobular capillary hemangioma (LCH), formerly known as pyogenic granuloma, is an acquired benign vascular proliferation of unknown origin, with a characteristic lobular architecture on microscopy [1]. It usually affects skin of the head and neck, and mucous membranes of the oral cavity. It is rare in the nasal cavity of children [2].

To the best of our knowledge, only fifteen pediatric intranasal LCH cases have been reported in the literature (Table 1). None of these occurred in the inferior turbinate.

We report two new pediatric cases of LCH, one of them on the inferior turbinate and the other one on the anterior nasal septum.

Our principal aim was to highlight the importance of considering this lesion as a differential diagnosis for pediatric unilateral nasal obstruction and epistaxis.

2. Description of cases

2.1. Case 1

A 13-year-old male presented to us with complete obstruction of the right nasal cavity, on a 2 months background of recurrent anterior epistaxis and purulent discharge, after nasal trauma (accidental contusion while nose picking).

Nasal endoscopy revealed a big red smooth surfaced mass with abundant purulent rhinorrhea filling the right nasal cavity (Fig. 1A). The mass was mobile and appeared pedunculated with its base

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Table 1
Pediatric intranasal lobular capillary hemangioma in the literature.

Study	Age	Gender	Origin in the nasal cavity	Imaging study	Treatment
Mills et al., 1980 [14]	10 years	Female	Septum	None	Endoscopic excision
Simo et al., 1998 [15]	7 years	Male	Right lateral wall	NR	Endoscopic excision
Kapella et al., 2001 [16]	7 years	Female	Left vestibule	CT	Endoscopic excision
Ogunleye and Nwaorgu, 2000 [17]	45 days	Male	Roof of the left nasal cavity	CT	Endoscopic excision
Karagama et al., 2002 [18]	8 years	Male	Left floor	None	Elliptical incision + 4/0 Vicryl stitches
Ozcan et al., 2004 [19]	6 years	Female	Right floor	CT	Antibiotic and decongestant 20 days prior to endoscopic excision
Katori and Tsukuda, 2005 [2]	11 years	Male	Right lateral wall	CT and MRI	Elliptical incision with Nd Yag Laser
Puxeddu et al., 2006 [7]	NR	NR	NR	CT	Endoscopic excision
Puxeddu et al., 2006 [7]	NR	NR	NR	CT	Endoscopic excision
Benoit et al., 2010 [20]	5 years	Male	Right Septum	Imaging studies	Endoscopic excision
Burlucchi et al., 2010 [4]	5 months	Male	Left Septum	MRI	Endoscopic excision
Ifeacho and Caulfield, 2011 [21]	14 years	Male	Right middle turbinate	MRI	Endoscopic excision
Virbalas et al., 2012 [11]	12 years	Female	Left middle meatus	CT	Endoscopic excision
Virbalas et al., 2012 [11]	16 years	Female	Right middle Turbinate	CT	Endoscopic excision
Vijaya et al., 2015 [22]	14 years	Male	Left septum	CT	Endoscopic excision
Case 1	13 years	Male	Right inferior turbinate	CT	Endoscopic excision
Case 2	12 years	Female	Right septum	CT	Endoscopic excision

NR = Not reported; CT = Computed tomography; MRI = Magnetic resonance imaging.

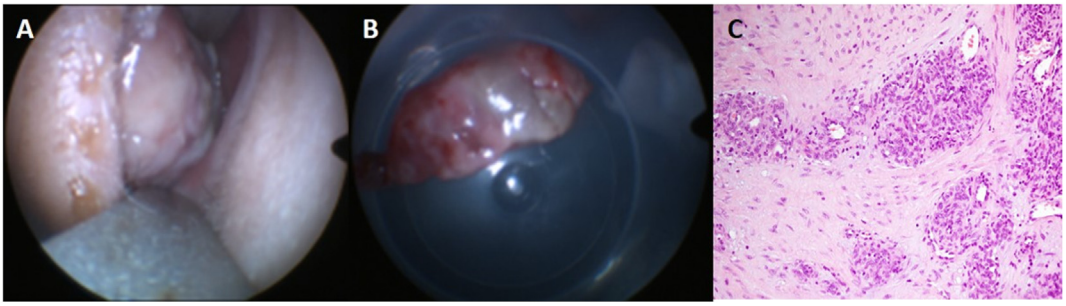

Fig. 1. Case 1. (A) Examination of right nasal cavity with a Killian speculum. (B) Gross appearance of the nasal mass after excision. (C) LCH composed of small capillaries lined by plump endothelial cells (H&E staining).

attached to the inferior turbinate.

Plain and contrast enhanced computed tomography (CT) of the paranasal sinuses revealed an intensely enhancing big vascular lesion with liquid content, well-circumscribed, in the right nasal cavity (anterior area of middle and inferior meatus) with no obvious bony remodeling or destruction (Fig. 2).

The differential diagnosis included a turbinate abscess, hemangioperycitoma, a turbinal hematoma, and juvenile nasopharyngeal angiofibroma which are more common in this population.

The lesion was completely resected endoscopically using cold dissection and bipolar coagulation, with no complications. Histological analysis confirmed the diagnosis of LCH with surgical margins free of disease (Fig. 1C).

The patient made a successful recovery and remains on follow-up with no recurrence two years after surgery.

2.2. Case 2

A 12-year-old female came to our institution with a 4-months history of right-sided nasal epistaxis. She had no other medical history and was not on medication. There was no family history of note. Anterior rhinoscopic examination showed a bilobuled dark-red mass arising from the anterior septum of the right nasal cavity which bled easily when the lesion was touched by a telescope.

CT scan showed a well-defined soft tissue density lesion in the anterior aspect of left nasal cavity without bony erosions (Fig. 3).

Endoscopic excision with cold dissection and bipolar coagulation was done under general anesthesia and the specimen was sent for histopathological examination, which confirmed the diagnosis of LCH.

The patient presented no complications and remains asymptomatic on follow-up with no recurrence one year after surgery.

3. Discussion

Capillary hemangiomas constitute 7% of all benign head and neck tumors in children. Nearly 75% of cases seen in children occur in this region with the gingiva, lips, and tongue being the most common sites [3]. The nasal cavity is a rare location for LCH, mostly seen in women between 3rd and 5th decade of life [4,5].

Pathogenesis of LHC remains unclear, though some evidence support nasal trauma [1], endocrine disorders [1,2], viral oncogenes, arteriovenous malformations, and angiogenic growth factors [6].

The relative frequent location of LCH at the anterior nasal septum (Kiesselbach's area) in recurrent nose pickers or patients with a history of nasal packing lends to belief that local trauma may precede the genesis of LCH [7]. In our first case, localization of the mass and previous history of nasal trauma would support this theory. However, a retrospective study of 112 patients by Pagliai and Cohen found a history of trauma in only 4% of patients with clinically diagnosed LCH [8].

On the other hand, increased levels of estrogen and progesterone have been associated to the pathogenesis of a specific form of LCH of the mucosa, called “the pregnancy tumor,” which occurs during pregnancy more commonly on the gingiva and, less commonly, in the nasal cavity [9]. These lesions generally regress