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A rare cause of pediatric nasal obstruction and epistaxis: Nasal septal mixed hemangioma

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SUMMARY

Hemangiomas are rare benign vascular endothelial tumors mostly arising in the head and neck area. The infantile cases are the most frequently seen pediatric tumors with an incidence of 5%. Usually, congenital lesions are located on the facial skin or oral mucosa, the nasal cavity is not a common site for hemangiomas. Histologically, there are three types including capillary, cavernous and mixed hemangiomas. Epistaxis and nasal obstruction are the most common presentations for nasal involvement. In this article we present the first reported case of pediatric capillary-cavernous hemangioma of the nasal cavity in the literature with a review of current theories regarding the etiology, diagnosis, and treatment of this rare type of hemangioma.

1. Introduction

Hemangioma is a benign neoplasm of vascular phenotype [1]. According to the predominant type of vascular channel these lesions are classified as lobular capillary, cavernous and mixed (Capillary-cavernous hemangioma) [1–3].

Lobular capillary hemangioma (LCH) was first defined as "human botryomycosis" and afterward called as pyogenic granuloma, granuloma pedunculatum, infected granuloma and telangiectatic granuloma [1–6]. They are pedunculated, benign and very fast growing vascular lesions that usually involve the skin or oral mucosa, frequently encountered by dermatologists and otolaryngologist [7]. Although not a rare lesion of the head and neck region, nasal cavity is extremely rare areas for LCHs in children [3,8]. Nasal LCHs generally originate from the entrance of the nasal cavity [8]. Currently despite the etiology of LCH remains unclear, there is some evidence to support both trauma and hormonal influences. LCH may occur in all age groups, but it is more frequent in women at the third and fourth decades of life. In pediatric age group male occurrence is prominent [1,7]. The presence of a unilateral rapidly growing intranasal mass in children is an alarming clinical sign, especially when associated with bleeding [6].

Cavernous hemangioma of the nasal cavity is rare [9]. Mean age at presentation of cavernous hemangioma of the nasal cavity is 40 years and it is seen in both genders with equal frequency [10–12]. This tumor, when symptomatic, produces recurrent epistaxis or hemoptysis and nasal obstruction. Histologically, cavernous hemangiomas have a microscopic appearance of large blood-filled canals lined with a single layer of endothelial cells and separated by scant connective tissue stroma [13].

In this article, the first pediatric case of nasal septal mixed (capillary-cavernous) hemangioma, is presented.

2. Case report

A 6-year-old female, was admitted to the clinic with complaints of recurrent epistaxis and nasal obstruction from left nostril for 3 months' duration. There was no history of facial trauma, headache, facial pain, prior surgery or nasal packing for epistaxis. She did not require any hospital admission for epistaxis that was managed by her parents with local finger compression of anterior nasal vestibule or wash out the nose with cold tap water. No other rhinologic, otologic or dental symptoms were present. Endoscopic examination revealed pale reddish, hemorrhagic, 2 cm mass attached to the antero-superior aspect of the left nasal septum by a pedicle, and extended 5 mm to the proximal caudal septum (Fig. 1). The left nasal mucosa was slightly edematous, and minimal purulent secretion in the nasal passage was noted. The mass was sensitive to touch and bled easily on contact.

The right nasal cavity, right lateral nasal wall, and right side of the nasal septum were normal. Rest of the examination revealed

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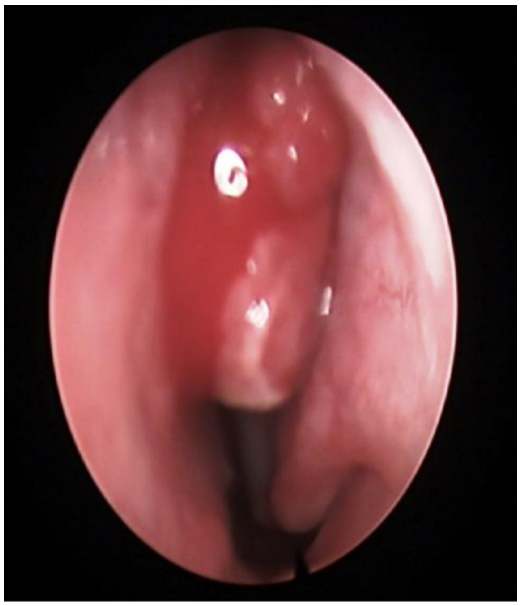

Fig. 1. Preoperative image of an hemangioma arising from the antero-superior aspect of the left nasal septum.

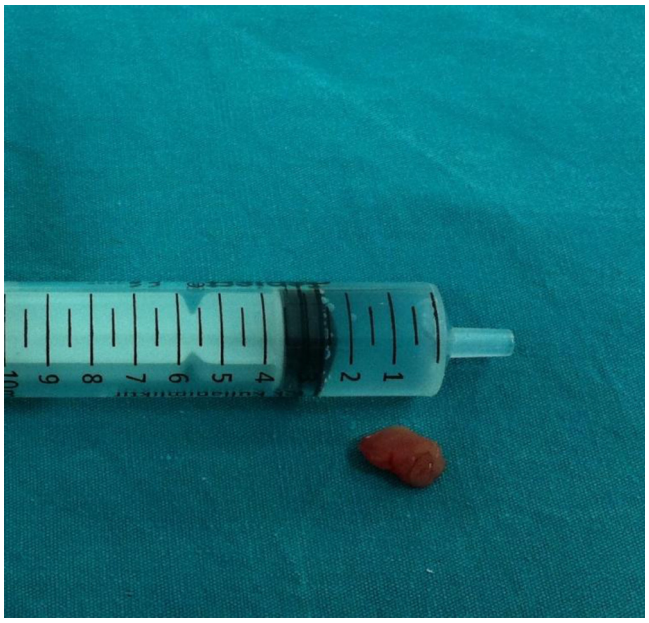

Fig. 3. Haemangioma after extirpation.

essentially normal findings. The patient was scheduled for an operation under local anesthesia. Intraoperatively by using a 0 degree Hopkins telescope it was seen that the mass had a thin peduncle that originated from the anterosuperior left nasal septum. The 20 mm × 10 mm × 8 mm sized mass was totally excised subperichondrially with normal mucosal margins under endoscopic view (Figs. 2 and 3). There was no destruction or erosion in the septal cartilage. Following excision, a tampon soaked with antibiotics was placed into the left nasal cavity. The postoperative course was uneventful. The tampon was removed two days after the surgery. The pathology report described polypoidal fragment of tissue surrounded by stratified squamous epithelium with focal ulceration. The underlying loose, oedematous stroma was composed of proliferating thin walled capillary and cavernous vascular channels arranged in lobular fashion. The stroma was infiltrated by mononuclear inflammatory cells (Fig. 4a). Immunohistochemically, capillary and cavernous vascular channels lined by flattened endothelial cells were highlighted by CD 34 (Fig. 4b). The cells were not reactive by EMA. The definitive diagnosis was capillary-cavernous (mixed) hemangioma. Neither recurrence nor any residual disease was seen in endoscopic examination in the postoperative fifth month.

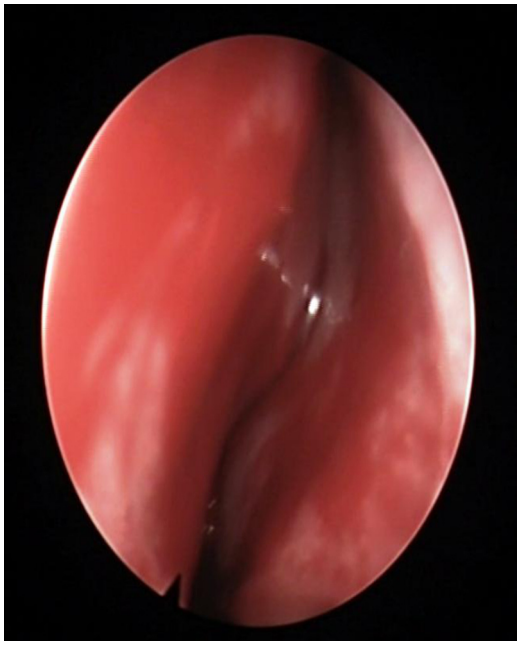

Fig. 2. Image of nasal septum after excision of an hemangioma.

3. Discussion

Haemangiomas are benign vascular tumors, which originate in the skin, mucosae and deep structures such as glands, bones and muscles [1,3]. The classic histological classification as capillary, cavernous and mixed hemangiomas has no clinical relevance [1–3,8].

LCH is a common, benign, acquired vascular tumor and occurs in the skin and mucous membranes [8]. It was first described by Poncet and Dor in 1897 as "human botryomycosis" [4,5]. In 1980, Mills et al. [6], noting the paucity of evidence to support an infectious origin, proposed the term "lobular capillary hemangioma" derived from the characteristic microscopic features of this tumor [1,8]. The gingiva, lips, tongue and buccal mucosa are the most common sites of mucosal lobular capillary hemangioma, but intranasal localization is very rare [1,14]. The anterior portion of the septal mucosa and the tip of the inferior turbinates are the most involved areas in the nasal cavity [1].

Although LCH may appear in all ages, it is more common in the 3rd decade and in females [4]. It is considerably rare in children; only a few cases have been reported [1–3]. In a clinicopathological research performed on 178 children with LCH, it was reported that 62% of the lesions were located in the head and neck area, 21.8% were present in mucosal membranes such as the oral cavity and conjunctiva, with only one case of LCH arose in the nasal mucosa [15].

Cavernous hemangiomas have a microscopic appearance of large blood-filled canals lined with a single layer of endothelial cells and separated by scant connective tissue stroma [13]. Cavernous haemangiomas with a location at the nasal cavity or paranasal sinuses are uncommon. They have been described arising from vomer, the inferior turbinate, maxillary sinuses and