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CLINICAL REPORT

Lobular Capillary Hemangioma of the Ethmoid Sinus: A Report of Two Cases

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Abstract Soft-tissue tumors are rare in the paranasal region. Lobular capillary hemangioma (LCH) is a distinctive vascular lesion most commonly seen on the skin and oral mucosa in the fourth and fifth decades of life. Forasmuch as the nasal cavity and paranasal sinuses are rare locations for LCH, it is not well known by many ENT surgeons. Here we present two cases of LCH of the ethmoid sinus. These cases are being reported owing to their scarcity as per the literature published global.

Keywords Hemangioma · Ethmoid sinus · Pathology · Iran

Introduction

A vast type of processes can produce space-occupying lesions in and around the orbit and paranasal sinuses. These include benign tumors, malignant neoplasms, vascular lesions, inflammatory processes, congenital lesions, and infection. Lobular capillary hemangiomas (LCHs) are benign neoplasms and despite their high incidence of head and neck, those located in the paranasal sinuses are scarce [1]. They commonly present as a sinonasal mass with nasal obstruction, intermittent epistaxis and occasionally as a proptosis. LCH of the ethmoid sinus is a rare entity.LCH of the ethmoid sinus should be considered in the differential diagnosis of a sinonasal mass since surgery or biopsy can lead to an unexpected loss of large amount of blood. Careful clinicopathological assay assisted by different imaging procedures and a high index of suspicion are needed for a correct diagnosis and well-timed intervention to lessen the morbidity to the patient [2]. Here we describe two cases of LCH of the ethmoidal sinus with a review of literature. To the best of our knowledge, there has been only one report of LCH originating from the ethmoid sinus [3].

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Cases Report

Case 1

A 43 year-old male presented to the ENT out patient clinic with a 2-months history of headache, left eye purulent discharche and left-sided nasal obstruction without epistaxis. He had no history of trauma or nasal packing. No other rhinologic, otologic or dental symptoms were present. Physical examination revealed a middle-aged man, not pale and afebrile. There was left sided proptosis and the left nasal cavity was obstructed by a relatively large mass. The mentioned mass was sensitive to touch and bled easily on contact. Left eye movement was restricted and painful. Mucosa was normal on the right side. The postnasal space was clear and rest of the head and neck evaluation did not reveal any abnormal findings. His laboratory parameters were within normal limits. A CT scan of the paranasal sinuses revealed a heterogenous expansile soft tissue opacity (Fig. 1) in the left paranasal sinuses, originates

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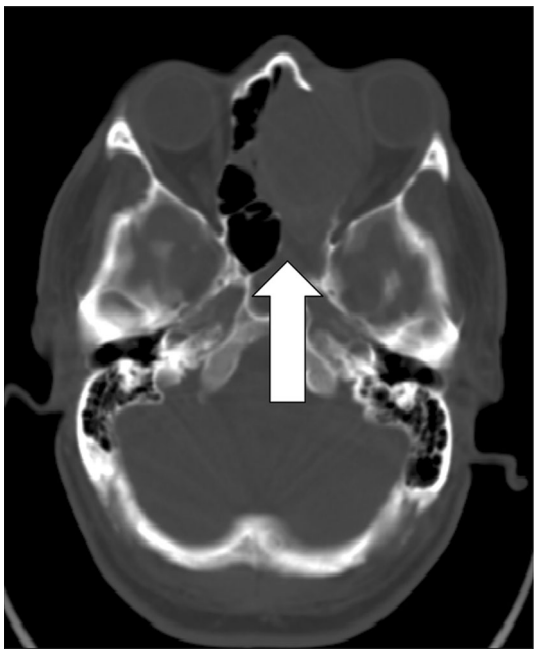

Fig. 1 Reveals a heterogenous expansile mass originates from the left ethmoidal sinus with bone erosion and extension to maxillary, frontal, sphenoidal and nasal cavity (*arrow*)

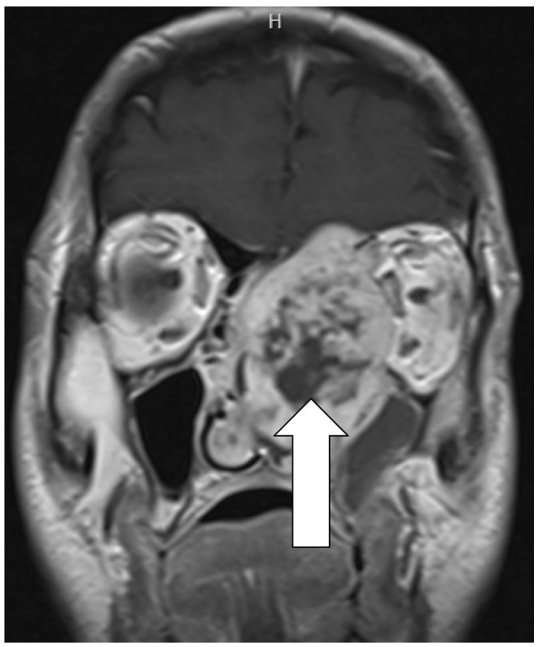

Fig. 2 Shows mass extension into the anterior cranial fossa with destruction of left cribriform plate (*arrow*)

from the ethmoid sinus with bone erosion and extension to maxillary, frontal, sphenoidal sinuses and nasal cavity. Nasal septum was eroded and deviated to the right side. Lamina papiracea was destructed due to mass extension into the orbital extraconal fat, but medial rectus muscle appeared intact. Also proptosis and glob displacement were evident. Mass extension into the anterior cranial fossa with destruction of left cribriform plate was seen, caused mass effect on the left frontal lobe without brain paranchymal invasion. After contrast injection (Fig. 2) the lesion showed heterogeneous enhancement with central necrosis. A biopsy from the mass was done. The specimen was sent for histopathological examination. Microscopic examination showed a cellular proliferation of capillary-sized vessels, many of which had inconspicuous lumina, and it was associated with an inflammatory infiltrate (Fig. 3). These morphologic findings were suggestive of LCH. The mass was excised completely under general anesthesia using an endoscopic surgery technique in another center. Final diagnosis was LCH. The patient had an uneventful post-operative course, and there is no recurrence till date.

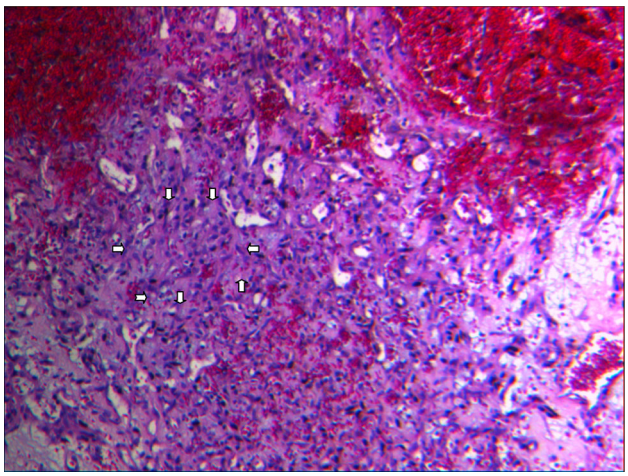

Fig. 3 Reveals cellular proliferation of capillary-sized vessels, many of which had inconspicuous lumina (*arrows*), and it is associated with an edematous stroma (H&E stain ×10)

Case 2

An 11-year-old boy presented to the ENT out patient clinic with intermittent bleeding, nasal obstruction and recently onset left eye proptosis. His symptoms had begun since 5 months ago. There was no known history of trauma, nasal packing or irritation. Physical examination and nasal endoscopy revealed a mass in the left nasal cavity. The mass bled easily with light touch and had an irregular surface. The postnasal space was clear and the rest of the otorhinolaryngological examination was normal. Examination of the nose and post-nasal space under general anaesthesia confirmed the presence of an irregular necrotic mass in the left nasal cavity. CT scan of the paranasal sinuses without contrast showed a lobulated soft tissue opacity occupying the left ethmoid sinus with extension to nasal cavity, maxillary and sphenoid sinuses (Fig. 4). The nasal septum was deviated to the right side. Also the lesion

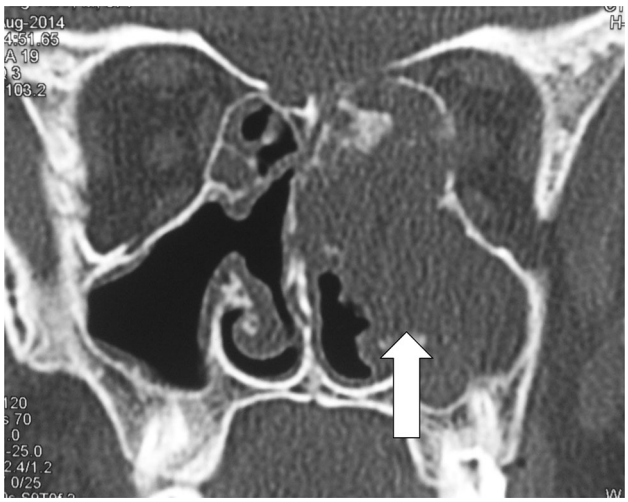

Fig. 4 Shows a lobuated soft tissue opacity occupying the left ethmoid sinus with extension to nasal cavity, maxillary and sphenoidal sinuses (*arrow*)

caused erosion of the medial wall of the maxillary sinus. Lamina papiracea was displaced laterally and protruded into the orbit with erosion. No intra orbital extension of the lesion was seen. A biopsy from the mass was done which was reported as LCH. The mass was excised in another center. The patient was reviewed 6 months later and there was no recurrence.

Discussion

LCH was first described by Poncet and Dor in 1897 as "human botryomycosis" [4] Already, this entity was described by Hartzell as "pyogenic granuloma," but it is really neither infectious nor granulomatous in nature. Mills described this disease as "lobular capillary hemangioma" according to the morphologic features [5]. LCH of the paranasal sinuses is a rare enity. Jamal reported the first case of LCH of the right frontal sinus [6]. Boedts described the first case of LCH of the ethmoidal sinus [3]. Results of one study showed that the most common presenting symptoms were epistaxis (75 %), obstruction (36 %), and pain (3 %), with no sex predilection (17/17; M/F), and median age was 39 years [7].Our first case was a 43 years old and the second patient was an 11 years old boy. However, in report of Elsayed and Al-Serhani, there was no child, although two female cases in their series were in their late teens [8]. Our first case sufferred from proptosis while epistaxis with nasal obstruction was chief complaints of the second patient. In a study on 40 cases, unilateral epistaxis and nasal obstruction occurred in 95 and 35 % of patients respectively [9]. The accurate etiology is still a dilemma. However microtrauma and pregnancy are the most commonly proposed etiological agents. Puxeddu et al., in their study containing a large number of patients with LCH, have identified prior trauma and pregnancy as a conceivable cause in only 15 and 5 % of patients respectively. Patrice et al. reported a series of 178 patients with LCH. Seventy-four per cent of their cases had no history of trauma [10].In our patients, none of these potential etiological factors were present, it is consistent with another report [11].It should be noted that contrast enhanced MRI is the preferred method for imaging, although CT scan can also be considered if MRI is not available. Furthermore CT scan may demonstrate bony expansion or erosion [12]. CT features of an LCH lesion consist of an intensely enhancing mass and an iso- or hypoattenuating cap of variable thickness around it [13]. In other words LCH should be considered as a potential diagnosis when a well-defined soft-tissue mass with mild diffuse homogeneous enhancement or marked central enhancement of the mass with a peripheral isodense area on the enhanced CT scans is seen. Lee et al. [13] reported the CT features of six cases of nasal LCH. The reported features were an intensely enhancing mass and an iso- or hypoattenuating cap with changeable thickness around the intensely enhancing mass in 5 (83.3 %) of 6 patients. In the residual cases (16.7 %), CT findings consisted of a lobular, intensely enhancing mass and a hypoattenuating area containing linear and spotty enhancing foci. Some believe that the degree of enhancement of the tumour on the CT scans is not constant, probably due to the variable size of both the tumour and vessels [14]. LCH was not thought to be universally associated with bony erosion though a few studies have found bony erosion to be more common [15]. In one study, bony erosion (50.0 %) and displacement (33.3 %) were seen in 50.0 and 33.3 % respectively. Bone destruction was seen in CT scan of our cases. Some believe that bony erosion is due to compression [16] and true bone invasion is rarely seen in LCH [2]. LCH can be difficult to distinguish from a malignant tumor owing to their rapidly growth and bleeding and also other benign tumors. For example Darren Stubbs et al. presented a case of LCH appearing as a suspicious nasal lesion of the lateral nasal wall with unilateral nasal obstruction and necrotic centre [17]. Perhaps the most clinically important lesions that need to be distinguished from a LCH are nasophaiyngeal angiofibroma, glomangiopericytoma and angiosarcoma. The thick blood vessels and stellate fibroblasts of angiofibroma are significantly different from the small capillary-size vessels with a lobular pattern seen in LCH. The nuclear atypia and infiltrative pattern that characterize angiosarcoma are absent in a LCH. Glomangiopericytomas are larger and more cellular lesions than LCH. They have an attenuated endothelial lining surrounded by a somewhat uniform population of plump to spindled cells [18]. In

children, the possibility of a foreign body should be considered. To date, there have been no reported cases of malignant transformation. The popular treatment of choice for this lesion is complete surgical excision [19]. Promising results have been reported for different modern treatments, such as laser therapy and sclerosing agents [20].

In conclusion lobular capillary haemangioma is a rare neoplasm which should be considered in the differential diagnosis of rapidly enlarging vascular lesions within the paranasal sinuses. These cases are reported due to the rarity of this lesion and its potential for being misdiagnosed. A high index of suspicion is needed to diagnose these cases.

Compliance with Ethical Standards

Conflict of interest Authors declare that they have no conflict of interest.

Ethical approval This article does not contain any studies with human participants or animals performed by any of the authors.

Informed consent Informed consent was obtained from all individual participants included in the study.

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