

Hemangioma of the buccal fat pad

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Abstract

Hemangiomas are benign vascular neoplasms characterized by an abnormal proliferation of blood vessels. Buccal fat pad (BFP) is a rare place for hemangioma. In this report, clinical, radiographic, and histopathological findings are described in a rare case of hemangioma with phleboliths involving the BFP, and a review is made of the international literature on this subject.

Keywords: Buccal fat pad, hemangioma, vascular malformation

Introduction

Vascular lesions are the most common congenital abnormality.^[1,2] In 1982, Mulliken *et al.* divided vascular lesions in the maxillofacial area into two groups: Hemangioma and vascular malformations.

The term “hemangioma” generally covers a series of hereditary vascular abnormalities.^[3] Hemangiomas are benign vascular tumors. The most common sites for their incidence are the head and neck. They are more common in women than men (3:1).^[4]

About 60% of hemangioma is located in the head and neck.^[5,6] Normally, 80% of hemangioma lesions can occur as a single lesion.^[7] Lips, tongue, and buccal mucosa are the most common sites of involvement. However, it is more likely to occur in the gingiva, mandible, palate, floor of the mouth, and parotid gland. There are few reports on the incidence of phlebolithiasis in these areas.^[8-10] Hemangioma is often present at birth and extends during growth. However,

many vascular lesions regress spontaneously before or during puberty.^[10] This report introduces a rare incidence of hemangioma in the buccal fat pad (BFP) along with phlebolithiasis.

Case Report

The patient was a 28-year-old woman who referred with the chief complaint of a swelling and stiffness in the left cheek. From a clinical perspective, there was a slight swelling in the masseter muscle. On palpation, a moving mass with a stiff area was felt; and in intraoral examination, its position was felt in the anterior ramus. The patient expressed no specific history of systemic disease. There was also no history of trauma incidence in the area. The only point in the patient's history was a course of laser therapy for skin rejuvenation in the left cheek and several other areas in her face.

In the medical history of the lesion, there were 3 times of triamcinolone injection in the area during the last 3 years to treat the lesion by another physician. The patient said that reduction in tumor size was seen for a while after these injections.

Aspiration was performed for the patient through intraoral approach, whose result was negative. Magnetic resonance imaging (MRI) radiography (with and without contrast media) was also prepared.

Magnetic resonance imaging revealed a solid heterogeneous mass in the pterygopalatine fossa area with penetration and extension towards both buccal and masticator spaces on the left. The submandibular area and carotid space were normal and no abnormality was seen in the nasopharynx area. Paranasal sinuses were also completely normal [Figure 1a and b].

According to the results of clinical and radiographic examinations of the treatment plan, the excisional biopsy of the studied mass was selected under complete anesthesia.

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Under general anesthesia, the patient underwent a surgery with intraoral access through a cut in the upper area of anterior ramus. After dissection in the upper-side direction, vascular lesion was seen in buccal extension of the BFP [Figure 2]. The BFP capsule was intact and the mass had offended no soft/hard adjacent tissue. A single firm nodule (probably calcified) was also seen in the lesion.

Given two points, first, “the possibility of a vascular lesion” and second, “for the purpose of liposuction for cosmetic goal and remove of the swelling on the patient’s cheek,” the vascular mass was removed along with anterior lobe, as excisional biopsy through intraoral approach and hence that we did not enter into the vascular lesion [Figure 3]. The specimen was sent to a pathologist for histopathologic examination. The clinical swelling of the cheek was removed immediately after the surgery. In the macroscopic viewpoint,

the lesion was a yellow and dark purple mass measuring 2 cm × 3 cm × 4 cm along with a hard nodule-like area. Microscopic results represented a vascular lesion composed of large amounts of small to large vascular structures covered with endothelial cells. Hemorrhage areas were also observed. Fatty tissues composed of lipocytes were seen in other areas. There was no sign of Malignancy [Figure 4].

Discussion

Vascular lesions are the most common congenital abnormality.^[1,2] Vascular lesions are generally divided into two categories: Hemangioma and vascular malformations.

Hemangiomas usually appear within a few weeks after birth and have a growth rate that exceeds the growth rate of children. In this growth phase, hemangioma will have its own characteristics: Endothelial cells getting fatter along with frequent mitotic division, increased number of mast cells, and multilayer basement membrane. Following this stage, flat and inactive endothelial cells are located in a context called fibrous fatty tissue with a normal view.^[11,12]

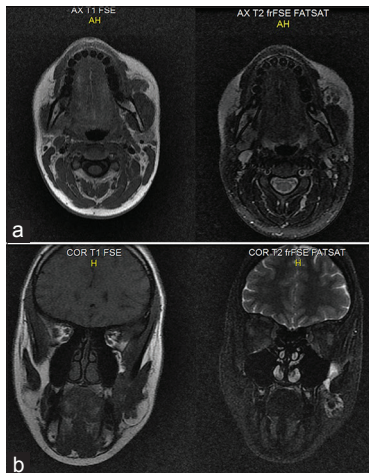


Figure 1: Magnetic resonance imaging (MRI) of the lesion. MRI revealed a solid heterogeneous mass in the pterygopalatine fossa area with penetration and extension toward both buccal and masticator spaces on the left. (a) Axial view T1 and T2. (b) Coronal view of T1 and T2



Figure 2: Intraoperative view of the lesion. Note the buccal fat pad with hemangioma

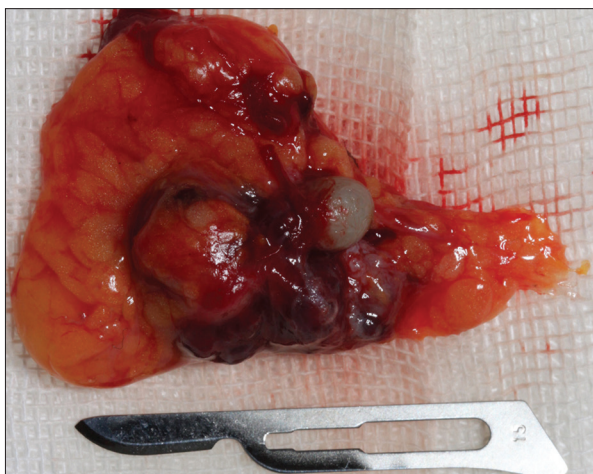


Figure 3: View of the totally excised lesion showing hemangioma with phlebolith

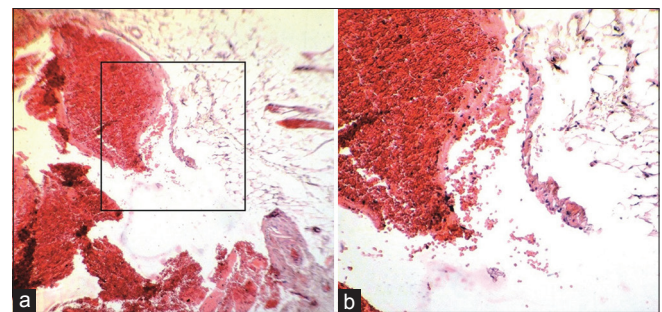


Figure 4: Histopathologic view of the lesion showing vascular lesion composed of large amounts of small to large vascular structures covered with endothelial cells. Hemorrhage areas were also observed. Fatty tissues composed of lipocytes were seen in other

Hemangiomas generally do not affect adjacent bones and they can be shallow or deep. Sometimes, they can even involve all layers of the skin and offend the muscles.^[13]

At the cellular level, hemangiomas are characterized by increased birth and death rate of endothelial cells and proliferation of mastocytes during the postnatal proliferative phase in the lesion. Derived from young proliferating hemangiomas, capillary endothelium is easily grown in cell culture mediums and forms tubes. In accumulated hemangiomas, the number of mastocytes decreases and becomes equal to natural tissues. Hence, a normal hemangioma is an endothelial tumor with a very complex life cycle of cell proliferation and natural regression.

Vascular malformations are the second major group of vascular lesions. In fact, they are an abnormal morphogenesis of blood or lymphatic vessels. Vascular malformations are present at birth. However, their clinical manifestations are not obvious sometimes until late infancy or even childhood.^[14]

The formation of phleboliths typically causes no symptoms. Phleboliths consist of a mixture of calcium carbonate and calcium phosphate salts^[15] and are thought to form when a fibrous component attaches to a developing phlebolith and becomes calcified. Radiologically, they have either a radiolucent or a radiopaque core, and repetition of this calcification causes an onion-like appearance or concentric rings.^[16]

It should be noted that BFP is also a mass composed of fat tissue covered with a thin capsule membrane and is mainly located in the buccal space. It plays an important role in the formation and activity of masticatory muscles. BFP has a rich blood supply and it is a proven fact that BFP has multipotential cells.^[17]

Kirmisson first reported a hemangioma case with phlebolithiasis in 1905. However, there was no report about the incidence of hemangioma in the BFP until 1956. Deighan and Barton first pointed to the incidence of a hemangioma case with phlebolithiasis in the BFP mass in 1956.^[18]

After a review of English literature, only two cases of the incidence of hemangioma in the BFP mass were found, except the above case. Ikegami and Nishijima reported the incidence of hemangioma in the BFP mass in a 23-year-old patient in 1984. In that case, the tumor was enucleated and the presence of a cavernous hemangioma was confirmed. No lesion recurrence was reported.^[19]

The last report on the incidence of hemangioma in the BFP was published by Tanaka *et al.* in 2000. The patient was a 3-year-old boy whose tumor was diagnosed 4 months after birth. The tumor was removed by extraoral access. Histopathology results showed the presence of capillary hemangioma with no evidence of phlebolithiasis.^[10] Unlike the

report of Ikegami and Nishijima in which the lesion surface was irregular, in this case, the lesion surface was reported smooth. However, the surface of the removed lesion was also irregular in this report. Like the case described in this report, no involvement was reported in the BFP mass adjacent areas in the previously reported cases.

When radiographic examinations reveal a radiopaque lesion in the tumor, the differential diagnosis will be easier and there will be two possibilities: Hemangioma or sialolithiasis of the parotid gland.

However, when phlebolithiasis is not seen, preoperative diagnosis of hemangioma will be very difficult.^[19]

In the report by Tanaka *et al.*, the tumor was removed through extraoral access. However, in the extraoral access, the facial nerve is more likely to be damaged and risk of scar is present. Moreover, access is more convenient through intraoral access. However, Tanaka *et al.* suggested that they used extraoral access because they assumed the probability of large extension of the lesion.^[10] This shows the importance of careful radiographic examinations for accurate diagnosis of the lesion limits and the selection of appropriate surgical technique. Therefore, if the tumor does not have too large extension according to clinical and radiographic examinations, then intraoral access is preferred. In such cases, the use of only one single imaging modality cannot provide enough information with the physician about the diagnosis and treatment of vascular lesions. Therefore, the use of MRI and computed tomography (CT) is recommended in these cases. The use of ultrasound/color Doppler will also be very helpful for validating the MRI and CT interpretational results.^[20]

Conclusion

In general, the incidence of hemangioma in the BFP will be very rare, but in cases where this lesion is suspected, precise preoperative clinical and radiographic examinations are recommended.

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