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Huge Cavernous Hemangioma of the Infratemporal Fossa

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Abstract: Hemangiomas are benign vascular tumors. We reported a case of a 17-year-old patient had been present for six years with a swelling of the right hemiface, in whom the clinical examination showed a mass of firm consistency, painless on palpation, measuring 13 cm in its major axis without facial paralysis. Computed tomography and facial magnetic resonance imaging revealed a cavernous hemangioma of the right infratemporal fossa. The surgical excision in one piece was carried out in dual temporal and vestibular approach twenty-four hours after embolization. After one year of follow-up there is no recurrence. The infratemporal location of the cavernous hemangiomas is exceptional, magnetic resonance imaging is considered the reference examination, and complete surgical excision represents the treatment of choice.
Keywords: cavernous hemangioma, infratemporal fossa, Benign tumour.

INTRODUCTION

Hemangiomas are benign vascular malformations that originate in mesodermal nets of vasoformative tissue. Synonyms for hemangioma are benign mesenchymoma, infiltrating angiolipoma, angiofibrolipoma, hemartoma and cavernous hemangioma. Depending on their histology, hemangiomas are classified as capillary, cavernous and mixed-type. In adults, the most common type is cavernous hemangioma [1]. They are frequently seen on the trunk and extremities, but up to 20% of hemangiomas are located in the head and neck region [2] but they are exceedingly rare as primary tumors of the infratemporal fossa [1].

CASE REPORT

A 17-year-old boy, with no notable pathological history, who had swelling of the right hemiface gradually evolving for six years, the clinical examination found a right temporomandibular cheek mass of firm consistency, painless to the palpation, measuring 13cm in its major axis, adherent to deep plans and respect to the cutaneous plane, without limitation of mouth opening, nor facial paralysis (Figure 1).

Computed tomography (CT) showed a poorly encapsulated tumor process, with lobed, multi-lobed contours, little enhancement after contrast injection, containing calcifications, occupying the right infra-temporal fossa and pushing the zygomatic arch without bone lysis (Figure 2).

Magnetic resonance imaging (MRI) revealed a right temporo-jugal lesion process, heterogeneous, multi-lobed, that contained cystic areas, with T1 hyposignal, T2 hypersignal, and heterogeneously enhanced after gadolinium injection. Measured: 122x43x41 mm.

The tumor occupied the right infra-temporal fossa, where it drove Bichat's fat ball and maxillary sinus without invading it, encompassed the tendon of the temporal muscle, and came into contact with the lateral pterygoid muscle that it drove downwards and backwards. The tumor spread downward to the masticatory space and drove the right masseter muscle out and up, extending to the right outer temporal fossa. (Figure 3)

Surgery was performed 24 hours after preoperative embolization of the internal maxillary artery, using a dual approach:

- Hemi-coronal right: which allowed the dissection of temporal portion of the tumor until the zygomatic arch
- Upper right vestibular, ranging from 11 to 18: which allowed the release of the jugal portion.
- Then the release of the infra-temporal portion, passing the tumor mass under the zygomatic arch, which was pushed back and forth by the tumor, widening the infra-temporal fossa and allowing the extraction of the tumor, by the endobuccal approach in one piece (Figure 4).

No postoperative complication such as hematoma, infection or facial paralysis was noted. The anatomopathological study of the operative specimen confirmed the diagnosis.

After a 1-year follow-up, the patient kept a projection in front of and outside the right zygomatic arch (Figure 5) which will be treated later by a repositioning osteotomy.

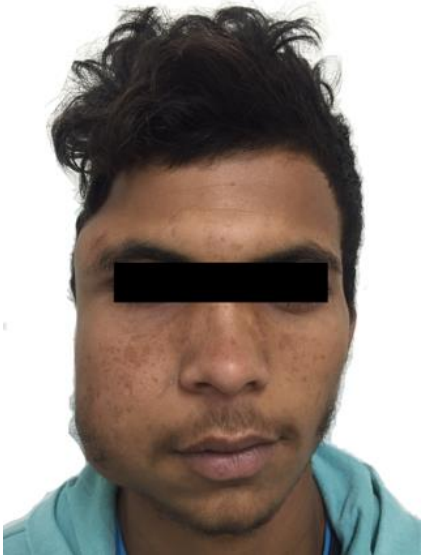

Fig-1: a right temporomandibular cheek mass

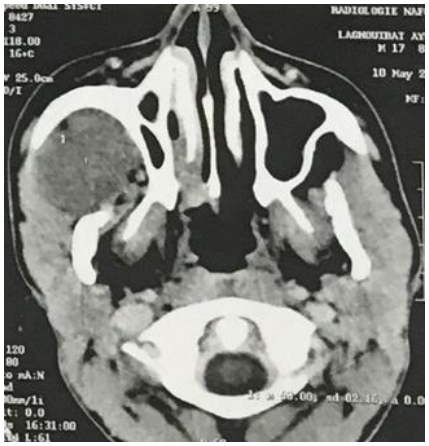

Fig-2: CT: a poorly encapsulated tumor process.

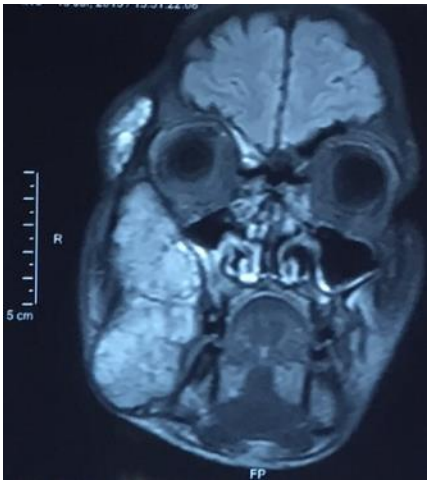

Fig-3: MRI: a right temporo-jugal lesion process

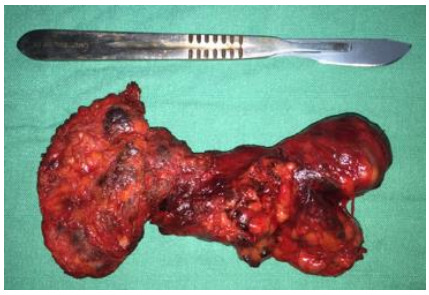

Fig-4: the tumor after extraction in one piece

Fig-5: After a 1-year follow-up

DISCUSSION

Hemangiomas are benign vascular tumors that likely occur due to abnormal development of embryonic vascular structures [3]. They are most commonly found on the trunk and extremities. Up to 20% of hemangiomas are found in the head and neck region, they may remain undetected for a long time. These tumors are likely to show spontaneous growth during the second or third decade of life. Almost 50% of cases remain silent until the mass grows and then pain suddenly occurs [2].

Nearly all tumors seen in the infratemporal fossa are either metastatic or extension of intramuscular or osseous lesions. Primary tumors of the infratemporal fossa are rare, and primary hemangiomas of the infratemporal fossa are exceedingly rare, as only a few cases have been reported in the literature [1].

Haemangiomas are readily distinguished from other soft-tissue tumours by computerized tomography (CT), MR I, and arteriography. CT is useful for defining the form, size and anatomic relationship of the tumour but MRI is the method of choice in defining the vascular nature of the tumour. On T1 weighted imaging, haemangiomas are iso-intense or hypo-intense to muscle. With T2 imaging, the lesions are hyper-intense on account of the volume of stagnant blood, clearly differentiated from the normal muscle and fibro fatty septa. Arteriography is helpful in delineating major vascular feeders for pre-operative embolization [4].

Treatment options for hemangioma include simple observation, irradiation, and injection of sclerosing agents, corticosteroid treatment, embolization and surgical excision. Although, based on the clinical history and physical examination, some authors argue that the cavernous type can be differentiated from the capillary type and recommend simple follow-up of these lesions unless they cause cosmetic, neurological or functional deficits, many others claim that surgery is the best way to exclude malignancy and report good outcomes and low recurrence rates with surgical excision [5].

A preoperative angiography can be helpful in the operative planning and embolization of the tumor, but embolization alone has been reported as an inadequate treatment if not followed by surgery [6]. Irradiation is not recommended as the amount of radiation needed is very high and has severe potential complications especially in children [7]. Indications for surgical treatment include: patient age, repeated severe hemorrhagic episodes, site and size of the tumor, depth of invasion, rate of growth, intractable pain, cosmetic deformity and suspicion of malignancy [4].

The complete removal of an infratemporal fossa lesion can be difficult because of the anatomic complexity and the inaccessibility of some masses. Additionally, complete excision raises concerns with regard to major postoperative complications. Both lateral and anterior approaches have been used to gain access to the infratemporal fossa. The lateral approaches include the direct, the transparotid, and the transotic procedures. The major complications of these operations are facial palsy, trismus, enophthalmus, malunion, and hearing loss. The most common anterior approaches are the transmandibular, the transoral, and the transmaxillary procedures, all of which require an osteotomy. Complications of these procedures include malunion, postoperative infection, neural deficits, dysphagia, and facial scarring. In addition, Komfehl *et al*. have described a relatively new transpalatine approach that does not require an osteotomy [1].

In the case reported, the tumor was completely excised, after embolization, by a double Temporal and vestibular approach, facilitated by the widening of the infra-temporal fossa due to the displacement of the zygomatic arch by the tumor. Periodic postoperative clinical and radiological follow-up is performed because of the high risk of recurrence which is 18% and 19% according to Wolf et al. and Tang *et al*. [2].

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