CASE REPORT

Cavernous haemangioma of the temporalis muscle: case report and review of the literature

Angioma cavernoso del muscolo temporale: caso clinico e revisione della letteratura

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SUMMARY

Haemangiomas are benign vascular neoplasms characterized by an abnormal proliferation of blood vessels. They may occur in any vascularized tissue including skin, subcutaneous tissue muscle and bone. These tumours are common in infancy and childhood and commonly involve subcutaneous or mucosal tissues. Intramuscular haemangiomas, a distinctive type of haemangioma occurring within skeletal muscle, account for less than 1% of all haemangiomas. They occur more often in trunk and extremity muscles, whereas involvement of the temporal muscle is extremely rare. Herein, the case is reported of a 38-year-old male who presented with a round, painless mass in the left temporal fossa, which was interpreted as an intramuscular haemangioma after a magnetic resonance imaging scan. In this report, clinico-pathological findings are described in an additional case of haemangioma involving the temporal muscle, and a review is made of the international literature on this subject.

KEY WORDS: Temporalis muscle • Benign tumour • Haemangioma

RIASSUNTO

Gli angiomi sono neoplasie benigne di origine vascolare caratterizzati da un'anormale proliferazione dei vasi sanguigni. Possono interessare qualsiasi tessuto vascolarizzato come cute, tessuto sottocutaneo, tessuto muscolare ed osso. Questi tumori sono comuni nell'infanzia e nell'età giovanile e si localizzano principalmente nel tessuto sottocutaneo o a livello delle mucose. Gli angiomi intramuscolari, un tipo differente di angioma localizzato all'interno della muscolatura scheletrica, rappresentano meno del 1% di tutti gli emangiomi. Questi si localizzano più spesso a livello dei muscoli del tronco e delle estremità, mentre l'interessamento del muscolo temporale è estremamente raro. Presentiamo un caso clinico di un paziente di 38 anni, giunto alla nostra osservazione perché presentava una neoformazione rotondeggiante, non dolente in corrispondenza della fossa infratemporale di sinistra, che fu interpretata come un angioma intramuscolare dopo esame RMN. Questo articolo tratta le caratteristiche cliniche ed istopatologiche di un caso di angioma del muscolo temporale ed una revisione della letteratura in merito.

PAROLE CHIAVE: *Muscolo temporale • Tumori benigni • Emangioma*

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Introduction

Haemangiomas are benign vascular neoplasms that most often occur in the skin or subcutaneous tissue. Less than 1% of all haemangiomas of the body occur in muscles, and only 14% of these originate in the musculature of the head and neck [1 2]. The masseter is the muscle most frequently affected, whereas intra-muscular haemangiomas of the temporalis muscle are extremely rare and, to the best of our knowledge, only 23 cases have been reported in the literature (Table I) [2-20]. It has been estimated that 90% of intra-muscular haemangiomas occur before the third decade of life. with no sex predilection [1].

Intramuscular haemangiomas usually present as an intra-muscular mass with distinct margins characterized by painless and slow growth. Because of their rarity and vague presentations, > 90% of cases are misdiagnosed before surgery [1]. An additional case of intra-muscular haemangioma is herewith reported, which presented as a painless mass in the temporal region.

Case report

A 38-year-old man presented in our Department, in 2005, with a history of a mass in the left temporal fossa which had been gradually increasing in size over the last 6 months, causing deformity of the region. He also reported excision of a lipoma, in the left temporal region, many years before.

Physical examination showed a 3 x 2 cm firm, round and painless mass, situated above the zygomatic arch. The overlying skin was normal and no adenopathy was noted. Subsequently, a magnetic resonance imaging (MRI) scan was performed which revealed a 2.5 cm ovoid mass within the left temporal muscle, just above the zygomatic arch (Fig. 1). The tumour had isointense signals on T1 weighted sequences and a high signal of T2 weighted sequences. Fine needle aspiration biopsy (FNAB) of the lesion was non diagnostic on account of the bloody specimen. These findings led to a strong suspicion of haemangioma. Surgical excision was performed through a hemi-coronal flap (Fig. 2A). The mass was found within the temporal muscle and was excised with a margin of normal muscle around it (Fig. 2B). Grossly, a 2.7 cm nodule, relatively circumscribed, with red cut surface, was observed. Microscopically, it showed a vascular tumour with intra-muscular growth composed of a mixture of vascular channels with prevalent large sized vessels lined with flattened endothelium, occasionally thickened by fibrosis of the avventitia (Fig. 3). A rim of muscle tissue was seen peripherally. The definitive diagnosis was consistent with intra-muscular cavernous haemangioma. The post-operative clinical course was uncomplicated. Two years after the operation, no sign of local recurrence was observed.

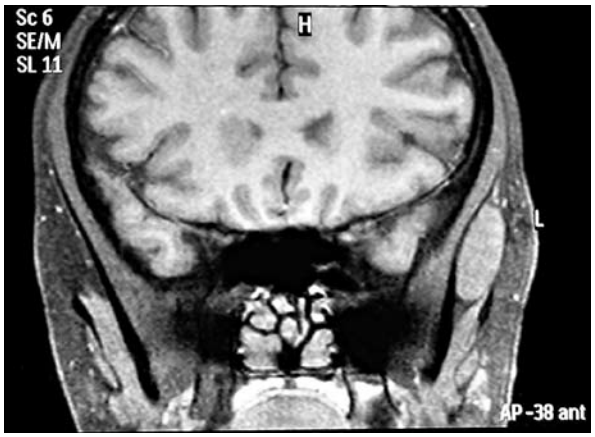

Fig. 1. T2 weighted coronal MRI showing a 2.5 cm ovoid mass within left temporal muscle.

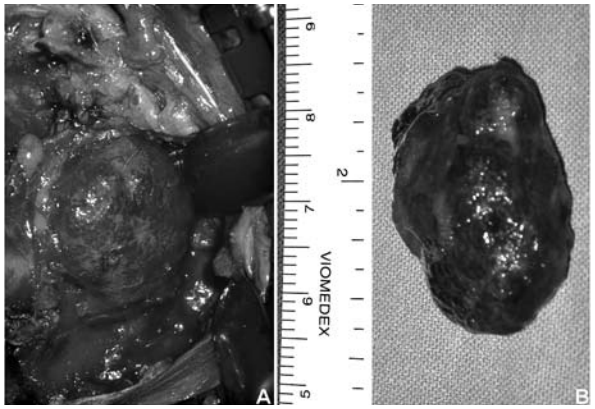

Fig. 2. (A) Intra-operative view of the haemangioma approached through hemicoronal flap. (B) Surgical specimen. Mass was well circumscribed and excised with a margin of normal muscle.

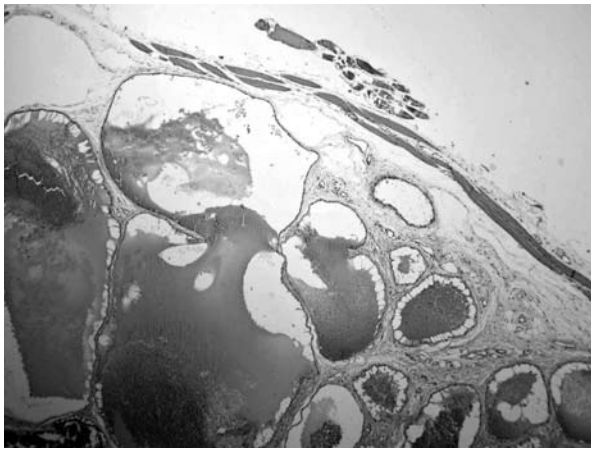

Fig. 3. Microscopy revealed a vascular tumour with intra-muscular growth composed of a mixture of vascular channels with prevalent large size vessels lined by flattened endothelium, occasionally thickened by fibrosis of avventitia. Peripherally, a rim of muscle tissue was visible (Haematoxylin & Eosin x 106).

Discussion

Intramuscular haemangiomas are rare tumours accounting for 0.8% of all haemangiomas and approximately 14% of cases are localized in the musculature of the head and neck [1,2]. The masseter (36%), followed by the trapezious (24%), are the muscles most frequently affected [1], whereas intramuscular haemangiomas of the temporalis muscle are extremely rare, with only 23 cases having been reported in the literature (Table I) [2-20]. Intra-muscular haemangiomas were first described by Liston [21], in 1843, and classified, in 1972, by Allen and Enzinger depending on the vessel size [22]. The capillary variant (small vessels) occurs more frequently and represents 68% of intra-muscular haemangiomas, followed by the cavernous (large vessels) and mixed type (small/large vessels) with an incidence of 26% and 6%, respectively [23]. The aetiology of these lesions is unknown, although trauma or hormonal changes are considered important factors in the proliferation of embryonic vascular tissue. Even a congenital theory has been proposed on account of the high incidence of haemangiomas during the first years of life.

Intra-muscular haemangiomas usually present as a slow-growing mass with distinct margins, mobile and do not exhibit any of the vascular signs such, discoloration of overlying skin or pulsation. The differential diagnosis includes: neurofibroma, lipoma, dermoid cyst, enlarged lymph nodes, soft-tissue sarcoma, myositis ossificans and temporal arteritis [17,19].

Haemangiomas are readily distinguished from other soft-tissue tumours by computerized tomography (CT), MRI, 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 [24].

Certain radiological features, on MRI, would appear to suggest hemangiomas:

1) high-intensity signal on T2 weighted imaging;

Table I. Intramuscular haemangiomas of temporalis muscle: review of literature.

Case no.	Author/Year	Sex (M/F)/Age (yrs)	Histology	Treatment
1-4	Scott 1957 [3]	Information not available	Information not available	Information not available
5	Joehl et al., 1979 [4]	F/59	Cavernous	Surgery
6	Knox et al., 1990 [5]	M/19	Cavernous	Surgery
7	Sharma et al., 1991 [6]	M/21	Capillary	Surgery
8	Murakami et al., 1991 [7]	M/51	Cavernous	Surgery
9	Hughes & Hutchison, 1993 [8]	M/28	Cavernous	Surgery
10	Couloigner et al., 1996 [9]	F/41	Cavernous	Surgery
11	Tada et al., 1996 [10]	F/14	Cavernous	Surgery
12	Cappabianca et al., 1996 [11]	F/13	Cavernous	Surgery
13	Lopez-Cedrun et al., 1996 [12]	M/41	Cavernous	Surgery
14	Itosaka et al., 1997 [13]	F/12	Cavernous	Surgery
15	Shpitzer et al., 1997 [2]	F/29	Cavernous	Surgery
16	Benateau et al., 1997 [14]	F/61	Capillary	Surgery
17-18	Sharma et al., 2001 [15]	(F/5-M/27)	Cavernous/Capillary	Surgery
19	Sherman & Davies, 2001 [16]	M/1	Cavernous	Surgery
20	To et al., 2001 [17]	F/54	Cavernous	Surgery
21	Heckl et al., 2002 [18]	F/55	Cavernous	Follow-up
22	Bui-Mansfield et al., 2002 [19]	M/44 (bilateral)	Cavernous	Surgery
23	Top & Barcin, 2004 [20]	M/46	Mixed type	Surgery

2) endothelial-lined vascular channels separated by fibrous and/or fatty linear tissue in lesions > 2 cm;
3) presence of areas of thrombus, fibrosis, hemosiderin deposition and/or calcification [24].

Arteriography is helpful in delineating major vascular feeders for pre-operative embolization.

To the best of our knowledge, in the international literature, only 23 cases of temporal muscle haemangiomas have been reported, but only 19 of those cases are well documented (Table I) [2-20]. Cavernous haemangiomas are the most common type seen in the temporalis muscle (15/19 cases). There is a slight sexual predominance (10 females, 9 males) with mean age of 33 years. Surgical excision was the treatment of choice (18/19 cases). Generally, surgical excision including a rim of normal surrounding tissue is the mainstay of treatment, as it permits a histological evaluation of the mass and it reduces the local recurrence rate [17 18]. Indications for surgical treatment include: patient age, repeated severe haemorrhagic episodes, site and size of the tumour, depth of invasion, rate of growth, intractable pain, cosmetic deformity and suspicion of malignacy [1 15 17]. Heckl et al., instead, advocate only a strict clinical and radiological follow-up for at least 2 years and reserve surgical treatment only in the presence of capillary haemangioma due to its more aggressive behaviour (high rate of recurrence, invasive growth), in case of lesion progression and when quality of life is reduced (presence of intractable pain, aesthetic problems and functional impairment) [18].

Local recurrence, related primarily to incomplete resection, is different for the three pathological subtypes. Rates of recurrence of 28%, 20%, and 9% have been reported for mixed type, capillary, and cavernous intra-muscular haemangiomas, respectively [12].

The use of corticosteroids, radiotherapy and sclerosing agents has been proposed, both as alternatives and adjuncts to surgical excision, to reduce tumour bulk in the case of massive organ involvement (i.e., the tongue) [23]. A meticulous and prolonged follow-up, based on clinical and radiological examination, is strongly recommended in order to ensure immediate diagnosis of eventual local recurrence.

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