Clinical Case Reports

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Temporalis Muscle Cavernous Hemangioma in a Pediatric Patient

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Background: Temporalis muscle hemangiomas are an uncommon entity, even among the rare incidences of intramuscular hemangiomas. They are frequently identified as round, painless, soft lesions beneath the skin that have been growing slowly over time. A preliminary diagnosis can be made based on magnetic resonance imaging, but it is confirmed by pathology of the surgically excised lesion. The safest practice is to resect all of the hemangioma plus a safe margin of normal muscle so as to prevent recurrence as much as possible. Watchful waiting is also a reasonable alternative when surgery is not indicated.

Case presentation: Because of the apparent paucity of this lesion, especially in the pediatric population, we report this case of a temporalis muscle cavernous hemangioma in a male under 18 years of age who presented for a growing, painful temporalis lesion 4 years after being managed conservatively.

Conclusion: Temporalis hemangiomas are among the rarest intramuscular hemangiomas (IMHs) reported in the pediatrics population. They are often misdiagnosed, with MRIs being the preferred imaging for preoperative diagnosis, and a CT can determine bony involvement. The superior method of treatment when not simply observing its progress is wide surgical excision with normal muscle borders preceded by embolization for better bleeding control. IMHs do not have very high recurrence rates, but re-operation is feasible and none was shown to metastasize.

Keywords: temporalis, hemangioma, cavernous hemangioma, intramuscular hemangioma, pediatric

INTRODUCTION

Hemangiomas in general are the most common benign tumors of infants and children. [1] Of these, intramuscular hemangiomas (IMHs) constitute about 0.8% of all benign vascular tumors. [2] Signs that point to their vascular nature include discoloration, pulsation and bruits. They are rare and have poor characteristic clinical findings, which is why they are often confused with other muscular tumors, aneurysms or neurofibromas, leading to a misdiagnosis rate above 90% prior to surgery. [2,3] As such, it is possible such cases are being underreported. Most IMHs are present in the extremities and trunk, less often in the head and neck. Even among these, the masseter and trapezius muscles are more often the site of an IMH. The temporalis muscle is an uncommon site for IMHs to occur. [2] Up to this point, only 41 cases of IMHs of the temporalis muscle have been reported across all age groups, with our case of a temporalis cavernous hemangioma being the ninth to be reported in the pediatric population.

CASE REPORT

Our patient is an otherwise healthy male in his early teenage years who was presented for a right temporal hard, painful lesion noticed 4 years prior to presentation and treated conservatively. He reported recurrence of his pain and a growing lesion. A computed tomography (CT) scan of the head and neck showed a mixed-density, well-circumscribed subgaleal structure overlying the right temporal bone with no underlying skull fracture or skull changes. (figures 1 & 2) He underwent an operation for resection of this lesion under general anesthesia. A lazy S incision (figure 3) was made in the right temporal area behind the hairline to cover the lesion and the skin dissected from the underlying temporalis fascia. The temporalis fascia was opened within which the lesion was apparent as a friable, easily bleeding area. It was resected and sent for pathology, which later revealed dilated vascular channels of variable sizes, lined by flattened endothelial cells and separated by variable amounts of fibrous tissue and lymphocytic infiltrates, and surrounded by normal skeletal

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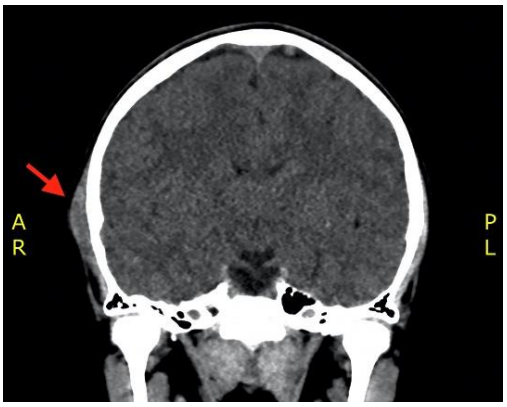


Figure 1 - CT scan of the head, coronal view, soft window, showing a right temporal mixed-density lesion (red arrow) within the temporalis muscle

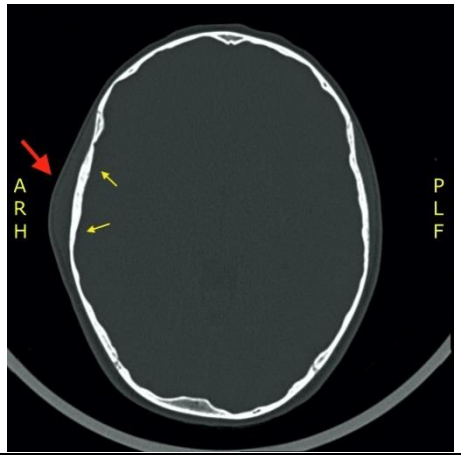


Figure 2 - CT scan of the head, axial view, bone window, showing the area of the right temporalis lesion (red arrow) and absence of bony invasion or erosions (yellow arrows)

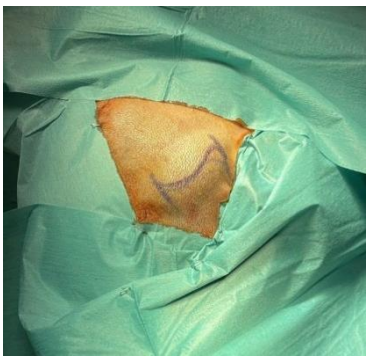
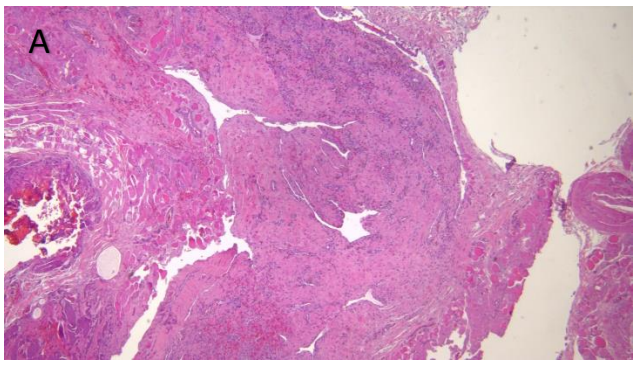


Figure 3 - Intraoperative view of the right temporal hemangioma and the skin incision required for resection

muscle tissue (figure 4). Hemostasis was ensured and the fascia was closed followed by the galea and skin. The patient had no postoperative complications and was discharged on a short course of antibiotics and painkillers. On follow-up a month later, he was doing fine and his sutures were removed with no signs of wound infection, bleeding or hematoma.



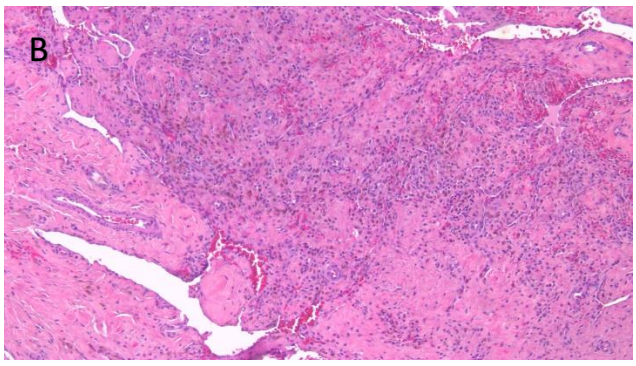


Figure 4A,4B - hematoxylin and eosin staining of the specimen showing dilated vascular channels of variable sizes, lined by flattened endothelial cells and separated by variable amounts of fibrous tissue and lymphocytic infiltrates, and surrounded by normal skeletal muscle tissue; no signs of atypia.

DISCUSSION

Between 80 and 90% of IMHs reportedly occur before the age of 40 and with no gender predominance. They do not involve the skin and subcutaneous tissue. [2] According to Scott et al. (1957), IMHs may be the result of congenital abnormal embryonic sequestration with a role for minor trauma in about 20% of cases or excessive contraction, whereby blood flow in the pre-existing tumor is stimulated [4] Hormones were also suggested to play a role in IMHs pathophysiology as some lesions were observed to remain stable or regress in postmenopausal women. [7]

According to the Allen and Enzinger classification published in 1972, IMHs can be classified into 3 types: capillary (68%), cavernous (26%) and mixed (6%). Their presentation in the head and neck region is estimated at 30%, 19% and 6% for each type respectively. Both the capillary and the cavernous types tend to present in the third decade of life with the mixed type reported to present a bit earlier in the second to third decades of life. The capillary types tend to occur in the trunk and upper limbs and are usually small (< 140 µm diameter), with a short clinical history to presentation, whereas the cavernous types tend to occur in the lower extremities and are usually larger (> 140 µm in diameter) and with a longer history to presentation. The mixed types are often found in the trunk and have a clinical course similar to that of the cavernous types. [4,9,10] In the pediatric population, these numbers differ. Only 8 cases have been previously reported in this age group, accounting to less than 20% of total cases reported in all age groups. No study in the literature discussed their statistics such as site distribution and others separately from other age groups. From what we gathered, only 4 of the reported cases were under the age of 8, with the rest being teenagers. [8] The new distribution of incidences of temporalis

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hemangiomas in pediatrics, with this case included, is around 56% for cavernous, 22% for venous and 11% for each of the capillary and mixed types.

As much as 98% of IMHs present as a slow-growing, firm, mobile mass with obvious margins, often painless (pain in 30-60%, as is with this case) and with no vascular signs (pulsations/ thrills/ bruits). [11] Most IMHs occur in the lower extremities (45%), especially the thighs, followed by 27% in the upper extremities then 14% in the trunk and around 14% in the head and neck region, the masseter being the most common location in that area (36%), followed by the trapezius (24%) and then the sternocleidomastoid. [2,4,9] The temporalis muscle is an uncommon site for IMHs. They are usually slow-growing and painless, with occasional temporomandibular joint tenderness. If the lesion is cavernous in nature, it can change size with head positioning or the Valsalva maneuver. [5]

Rarely, IMHs may display microscopic features like mitotic activity, intraluminal papillary tufting and perineural infiltration by capillary vessels, but they are still benign in nature and have never been shown to metastasize. [10]

In this case, only a CT was done to localize the growing lesion for resection. X-rays of the skull, ultrasounds, CTs and angiography may not be specific enough to identify the lesion. A CT alone may show the form, size and anatomic relations of the lesion, but is not enough to differentiate it from surrounding tissue and define its vascular nature. It is helpful, though, in determining bony erosion and intracranial extension. [2,4] An angiography can show an early venous runoff and feeders for later preoperative embolization, but they may be missed if too small. Aspiration cytology and open biopsies are not recommended as the first will yield a very bloody sample with possibly no identifiable tissue and the latter carries the risk of overt bleeding. [4] Ideally, magnetic resonance imaging (MRI) is of choice to best recognize an IMH. On T1-weighted imaging, IMHs are usually isointense and may appear as lesions with variable high signal intensity due to adipose tissue, thromboses and hemorrhages when present. On T2-wighted imaging, they appear markedly hyperintense with tubular structures with blood flow characteristics. [5] Features suggesting a hemangioma as stated by Buetow et al in 1990 included: a high intensity signal on T2W imaging, endothelium-lined vascular channels separated by fibrous and/ or fatty linear tissue in lesions more than 2 cm in diameter and areas of thrombosis, fibrosis, hemosiderin deposition and/ or calcification. [4,9] Phleboliths made of calcium carbonate and calcium phosphate were detected in 15-25% of IMHs. They are thought to organize, mineralize and then calcify on an existing thrombus formed when the peripheral blood flow slows. [12] Gadhia et al. and Cui et al. disclosed findings of phleboliths in their cases. [12,13]

The lesion in this case was reported as a well-circumscribed subgaleal mass of mixed density and no bone involvement. Unfortunately, no MR imaging was available to further define it. Based on these non-specific findings, the pre-operative differentials included a subacute subgaleal hematoma, with other possibilities such as a lipoma, neurofibroma or epidermoid or dermoid lesion that cannot be excluded. Despite a more accurate diagnosis by soft tissue visualization on MRI, the surgeon went with the clinical picture and CT imaging already done alone as clinical suspicion was very high and considering the financial constraints of the patient's family. Many authors listed common differentials for each of the lesions in their case reports. These included our own listed differentials in addition to aneurysms, enlarged lymph nodes, soft tissue sarcomas, myositis ossificans, temporal arteritis, lymphangiomas and schwannomas [9,14]

There has been a consensus since the earliest publications on the topic that surgery for excision of an IMH is the optimal choice of treatment since they tend to be infiltrative. The excision should be wide with resection of surrounding muscle beyond the borders of the tumor. [6] Intraoperative hemorrhage can be minimalized by preoperative embolization using autogenous clots, muscle or synthetic materials like gelatin foam. [1] Early resection is advocated to preserve the muscle structure and decrease rates of complications. [8] Proper excision results in lower rates of recurrence. But care must be taken not to injure the temporal and auricular branches of the facial nerve. [3,4,5]

Injection of a sclerosing agent and the use of corticosteroids or beta-blockers is questionable at best, and embolization or irradiation alone are inadequate. [7] Sclerotherapy did not result in postoperative scarring or functional impairment, but it may allow tumor regrowth and cause renal damage. [14] Irradiation is not curative and only serves to suppress growth. Plus, it is not helpful for large, slow-growing lesions and carries risks of radiation exposure in children and of malignant transformation. [6]

Burgos-Sosa et al. pointed out that endovascular treatment may restrict direct vision and interfere with proper vascular control. [8] Kishimoto et al. argued for improved aesthetic outcomes, greater safety, fewer surgical complications and less damage to surrounding tissue with endoscopy-assisted surgery since it offered an enhanced view of the surgical field for identification of anatomical structures. [3]

Some authors considered observation a valid option in certain cases where the lesions were not growing or causing the patients discomfort. [3,4,5] Some IMHs of the temporalis may remain stable and tolerable and can be followed up clinically and radiologically without ever requiring operation as Alqahtani pointed out with the two studies by Heckl et al. and Gadhia et al. [11] In all cited literature, there has only

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been one account of a venous hemangioma of the temporalis muscle that disappeared on its own. [15] Indications for surgery discussed in previous reports dealt with age, repeatedly severe hemorrhage, site and size, depth of invasion, rate of growth, intractable pain, cosmetic deformity and suspicion of malignancy [9,12] In our case, the patient was successfully treated conservatively with clinical and radiological follow-up and pain medications for 4 years until the pain recurred and the lesion was found to have grown, warranting surgery for lesion progression, cosmetic reasons and patient quality of life.

Rarely have surgical complications after removal of a temporalis IMH been reported in the available literature. Eryilmaz et al. discussed a facial nerve branch paralysis when their patient was not able to lift his left eyebrow directly after surgery, but it resolved on its own in 2 months. [4] The mixed types of IMHs tend to recur the most at a rate of 28%, followed by the capillary (20%) and cavernous types (9%). [11] Recurrence is usually attributed to incomplete excision, regardless of the type. [5] Around 18% of IMHs recur due to remaining minor feeders, which can be optimally controlled by preoperative visualization of the feeders, occlusion of the ipsilateral external carotid artery to decrease blood flow to the lesion by about 50% and wide dissection to include the temporalis fascia superficially and subperiosteal deep to the muscle. [2]

There is no official protocol, but an apparent consensus in the literature on the follow-up for temporalis muscle hemangiomas to last a period of 2 years at least, with the option of re-operation in case of progression. [9,14] Our case has only been recently operated on, with no signs of recurrence at 6 months postoperatively. We will continue to monitor our patient for a total of 2 years with repeat imaging as needed. Any further, the patient and his family will be educated about signs and symptoms of recurrence to report back to our clinic for further workup in case they occur.

CONCLUSION

Temporalis muscle hemangiomas rarely occur. They usually present as slowly growing, painless soft masses with well-defined borders and no skin abnormalities in young adults with no gender predilection. They are rare and have poor characteristic clinical findings, which is why they are misdiagnosed in more than 90% of cases. An MRI is the preferred imaging for preoperative diagnosis whereby the hemangioma appears isointense on T1W and hyperintense on T2W images. A CT can determine involvement of bones and adjacent structures. The superior method of treatment when not simply observing the progress of the lesion is wide surgical excision with normal muscle borders preceded by embolization if warranted for better bleeding control. It also offers the least complication rates compared to other treatment options. IMHs do not have very high recurrence rates, but re-operation is feasible and none was shown to metastasize.

DISCLOSURES

Ethical approval

This study was performed in line with the principles of the Declaration of Helsinki.

Consent to participate

The patients gave consent to use their information and images for research purposes. *Consent for publication*

The patient gave consent to use his information and images for publication.

Conflict of interest

The authors report no conflict of interest concerning the materials or methods used in this study or the findings specified in this paper."

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Artificial intelligence

The authors affirm that no artificial intelligence tools were used in the writing, editing, or content generation of this manuscript. All work was conducted manually, based on thorough research and academic expertise.

CONTRIBUTIONS

-**Asmaa Ibrahim Kebbe**: Data curation, Investigation, Project administration, Resources, Supervision, Validation, Visualization, Writing – original draft, Writing – review & editing
-**Mohammad-Nabih Mohammad-Walid El Houshiemy**: Conceptualization, Project administration, Resources, Supervision, Writing – review & editing
-**Khaled Haitham Sidani**: Data curation, Investigation, Resources, Writing – original draft
-Sarah Ahmad Kawtharani: Project administration, Supervision, Validation, Writing – review & editing
-Marwan Wahib Najjar: Data curation, Project administration, Supervision, Validation, Writing – review & editing
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