VIA MEDICA

CASE REPORT

Acta Angiol.
Vol. 13, No. 1, pp. 42–46

ISSN 1234–950X
www.angiologia.pl

Intramuscular haemangioma of the masseter muscle in a 9-year-old girl*

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*This article does not have any support for the work in the form of grants, equipments or drugs

Abstract

Intramuscular haemangioma are rare benign neoplasms which are usually asymptomatic until a growth spurt occurs in the second or third decade of life. The case of intramuscular haemangioma occurring in the masseter muscle of a 9-year-old girl is presented here. Magnetic resonance imaging and colour Doppler ultrasonography revealed the vascular nature of the lesion, and fine needle aspiration was strongly suggestive of the diagnosis and excluded other soft tissue tumours. Treatment options were reviewed and a "wait and watch" policy was adopted until her adolescence for excision in case of more invasive growth causing cosmetic problems and severe pain.

Key words: haemangioma-intramuscular-masseter muscle

Introduction

Vascular birthmarks are classified by Mulliken and Glowacki [1] as vascular malformations and haemangioma, based on their clinical appearance, histopathologic features and biological behaviour. Vascular malformations represent a lifelong disturbed vessel architecture with normal endothelial cell turnover, while haemangioma show areas of endothelial hyperplasia which is usually followed by spontaneous regression. Differential diagnosis is usually made according to clinical appearance and course, although this is a challenge, either because of an atypical presentation or because of difficulties of classification [2–4].

Intramuscular haemangioma are rare benign congenital neoplasms which account for less than 1% of all haemangioma, and less than 20% of these are found in the head and neck area. The masseter muscle is the most frequently involved site in the head and neck area accounting for 5% of all intramuscular haemangioma [5–7]. Unlike infantile cutaneous haemangioma, they do not regress spontaneously, and their deep location and unfamiliar presentation may require sonography, magnetic resonance imaging and sometimes angiography for accurate diagnosis [5, 6].

Because intramuscular haemangioma usually start to grow and are detected in the second or third decade of life; we found it interesting to report the intramuscular haemangioma of the masseter muscle in a 9-year-old girl [6, 7].

Case report

A 9-year-old girl who was otherwise healthy presented with a few bluish macules on the right inferior eyelid and the right zygomatic area. She underwent medical examination due to mild pain on her right cheek for the last one year. The mother said that the bluish macules on the right inferior eyelid and the right zygomatic area had first been noticed at the age of one week,

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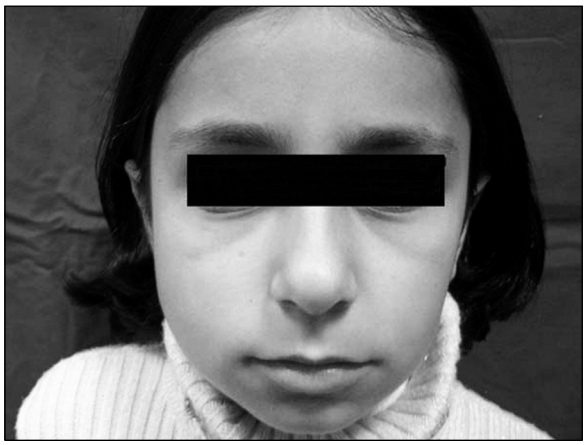

Figure 1. Mild asymmetry is seen between the cheeks

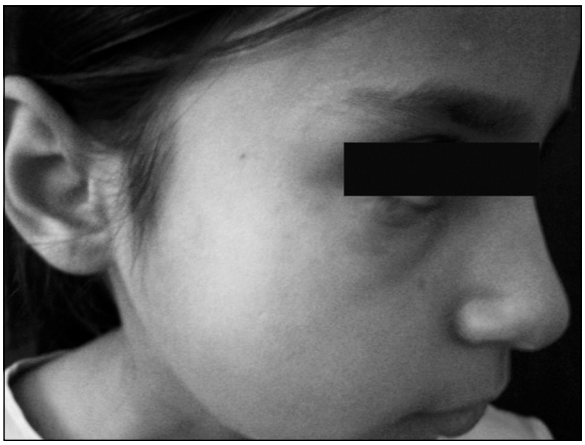

Figure 3. Bluish macules which belong to involuted cutaneous haemangioma are seen on the right inferior eyelid and the right zygomatic area

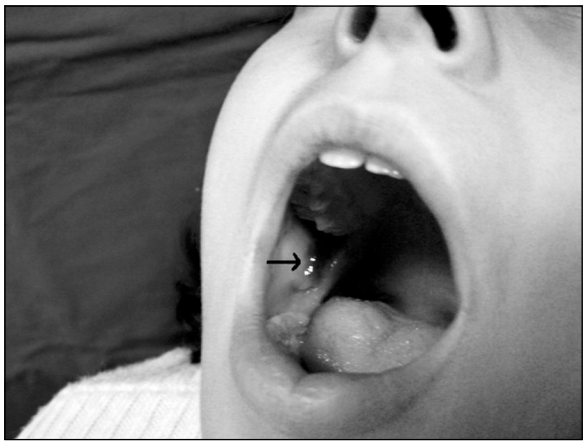

Figure 2. Oral photograph showing the reddish-blue plaque on the right posterior buccal mucosa which belongs to mucosal haemangioma indicated with the arrow

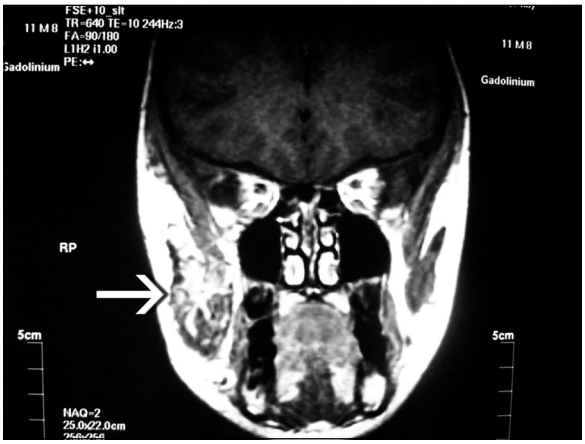

Figure 4. Contrast-enhanced sagittal T1-weighted MRI showing isointense signals in the right masseter muscle (indicated with the arrow)

at which time they had been more elevated and reddish. They had become flat and bluish one year ago. Physical examination showed a mild asymmetry between the cheeks with some firmness on the right side (Figure 1), and a reddish-blue plaque between the superior and inferior molar teeth on the right posterior buccal mucosa (Figure 2), as well as the bluish macules on the right inferior eyelid and the right zygomatic area (Figure 3). The lesions on the overlying skin of the right inferior eyelid and the right zygomatic area were diagnosed as involuted cutaneous haemangioma, and the lesion on the right posterior buccal mucosa was clinically diagnosed as a mucosal haemangioma.

The maxillofacial magnetic resonance imaging (MRI) with contrast revealed a 5 × 4 × 3 cm sized mass lesion, which was isointense with the muscle on T1 weighted series, in the anterolateral part of the right masseter muscle extending to the premaxillar distance, and showing continuity from the neighbourhood of the superior alveolar rim to the outer part of the masseter muscle (Figure 4). On T2 weighted postcontrast series, the mass lesion had inhomogeneous signals which were hyperintense compared to the surrounding masseter muscle (Figure 5A). Other similar hyperintense signals were noticed in the neighbourhood of the medial part of the mass lesion in the right masseter lesion with deep extension from the level of the ramus mandibula to the oropharynx and tonsil lodge and right infraorbital subcutaneous area (Figure 5B). These findings were strongly suggestive of an intramuscular haemangioma of the masseter muscle as well as another two haemangioma,

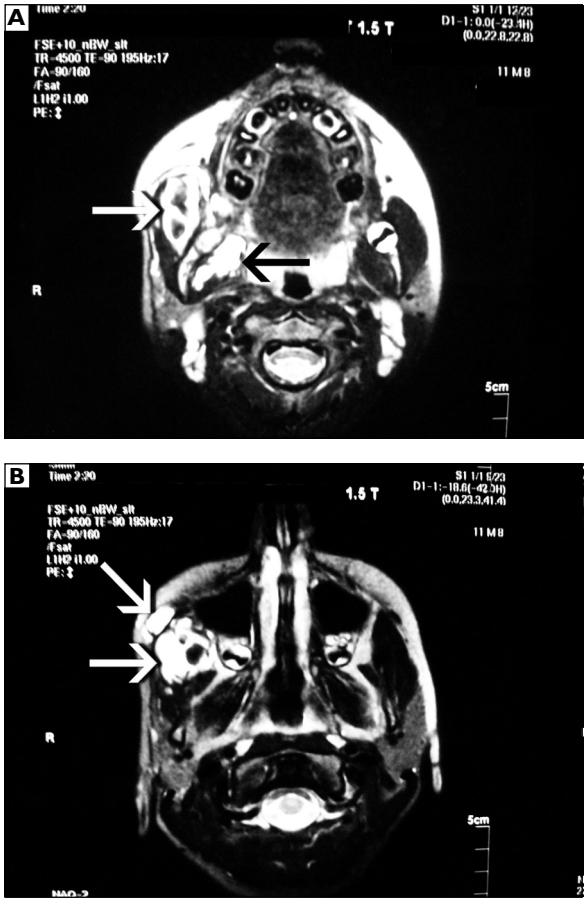

Figure 5A. Contrast-enhanced axial T2-weighted MRI shows hyperintense masses in the right masseter muscle and in the neighbourhood of the medial part of the mass lesion in the masseter muscle with deep extension from the level of the ramus mandibula to the oropharynx and tonsil lodge (indicated with the arrows). **B.** Contrast-enhanced axial T2-weighted MRI showing hyperintense masses in the right masseter muscle and right infraorbital subcutaneous area (indicated with the arrows)

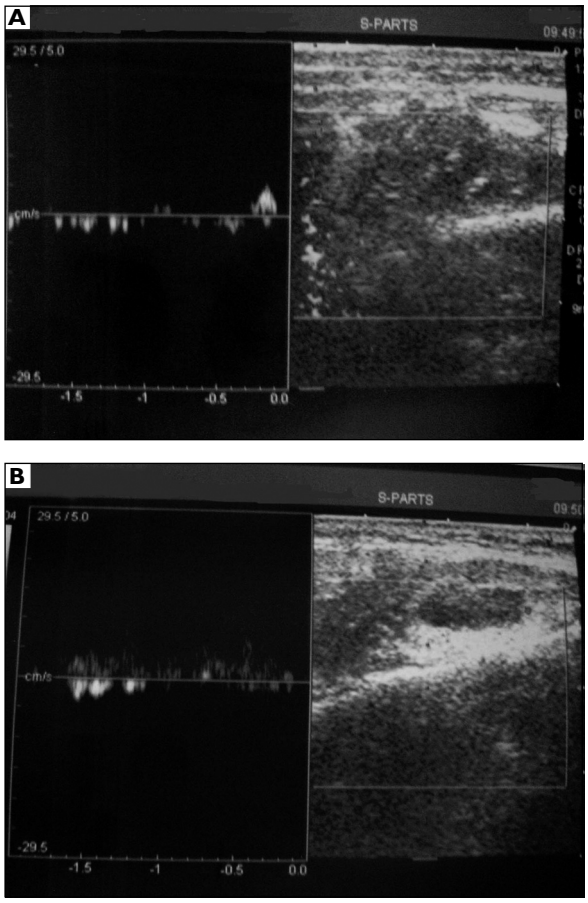

Figure 6A. Colour Doppler sonography of the right masseter muscle showing slow-flow venous Doppler signals. **B.** Colour Doppler sonography of the right masseter muscle showing some arterialized flow with low resistance

one in the inner side of the ramus mandibula which showed extension to the tonsil lodge and oropharynx, and one in the right infraorbital subcutaneous area.

Four months later a colour Doppler ultrasonography was performed, and hypoechoic solid mass lesions with lobular contours were observed in the right subcutaneous infraorbital area, right buccal area and right masseter muscle. Slow-flow venous Doppler signals compatible with haemangiomatous tissue features were seen in the mass of the lesion in the right masseter muscle (Figure 6A) as well as some arterialized flow with low resistance (Figure 6B).

She was reviewed two months later and fine-needle aspiration (FNA) was performed. An extensive amount of pure blood, which was strongly suggestive of haemangioma, came out.

Because she was 9-years-old, and had only mild pain in the cheek area, and there were no gross cosmetic problems, we decided in favour of clinical and radiological follow-up until adolescence, and will consider excision if the haemangioma causes cosmetic problems or severe pain due to more invasive growth.

Discussion

Haemangioma are benign proliferative vascular lesions characterized by increased endothelial cell turnover. These tumours usually appear after birth, grow rapidly, and involute over the years [8]. Within the spectrum of vascular lesions, intramuscular haemangioma are very rare, accounting for less than 1% of all haemangioma, and the masseter muscle is the most frequent muscle, accounting for 5% of all intramuscular haemangioma [6, 7]. The trapezius, periorbital, sternocleidomastoid, and temporalis muscles follow the masseter muscle in frequency [7]. The tongue, extraocular, and posterior neck muscles have also been reported to be in-

volved with haemangioma with less frequency [9–12]. Intramuscular haemangioma are non-metastasizing benign congenital tumours that, after remaining unrecognized for long periods, may suddenly start to grow in the second or third decade of life [6, 7]. A possible hormonal role in the growth of intramuscular haemangioma was speculated, but no specific data was available to irrefutably prove this hypothesis [6]. They are usually asymptomatic until a growth spurt occurs, at which time pain occurs in about 50% of cases. A palpable, fluctuant or firm mass is present in up to 98% of cases [5].

The diagnosis of intramuscular haemangioma requires a high index of suspicion. Whenever a mass of soft-tissue density is encountered in the region of skeletal muscle in a young adult, haemangioma should be considered in the differential diagnosis [13].

Haemangioma differ from vascular malformations by their appearance and course [2, 3]. Arteriovenous malformations (AVMs) are high-flow anomalies consisting of multiple abnormal communications between arteries and veins. Arteriovenous malformations are usually initially detected in infancy and remain quiescent during childhood. Puberty, pregnancy, hormonal treatment or trauma often precipitate evolution. Arteriovenous shunts bypass the capillary bed and result in cutaneous ischemia, ulceration and haemorrhage. The lesions may be disfiguring, impair function or result in increased cardiac output and congestive heart failure [14]. Venous malformations are present from birth and initially appear as either a faint blue patch or soft mass. They often enlarge slowly during childhood and adolescence and to a lesser degree during adulthood. Episodic thromboses occur, and large venous malformations may be associated with chronic consumption of fibrinogen and release of fibrin split products [15]. Lymphatic malformations are present from birth and are usually detected by the age of 2 years. They may be macrocystic, microcystic or combined [16]. Macrocystic lymphatic malformations are cool, soft, smooth, translucent masses that occur beneath normal or bluish skin. Microcystic lymphatic malformations permeate the skin and muscles and are often associated with tiny, clear, cutaneous vesicles. Recurrent infection and intralesional haemorrhage are frequent complications [15].

Most haemangioma are recognized clinically and do not require any investigation or any treatment as they will subside spontaneously. However, imaging is needed in cases of deep haemangioma with normal overlying skin, cases of clinically atypical soft-tissue masses, when the evaluation of extension of obvious haemangioma is necessary, cases of alarming haemangioma and for guiding therapy [4]. On plain X-rays and computerized tomography scans, phleboliths and calcifications can sometimes be identified, but they may not be specific [7, 17–20]. Sonography is the first-line imaging procedure for patients with soft tissue swellings [5]. Colour Doppler sonography is especially useful to demonstrate the vascular structures in and around the masseter muscle, and has the potential of being used to evaluate the pathological changes [21]. Haemangioma could be distinguished from other soft tissue lesions by the features of abundant vascularity and high blood flow velocity [4, 22]. When performing sonography on soft tissue masses in the head and neck, the presence of a colour Doppler signal in a well defined hypoechoic mass with heterogeneous echotexture should raise the possibility of haemangioma [23]. Haemangioma with arterial flow can be distinguished from arteriovenous malformations (AVM) by the presence of solid parenchymal tissue [15].

MRI is more reliable in detecting and delineating deep situated and large intramuscular haemangioma, and it gives the most diagnostic information [5, 24]. The MRI findings of an intramuscular haemangioma consist of an intermediate signal on T1 weighted images and an intense signal on T2 weighted images [25]. But it should be noted that not all intramuscular haemangioma will give a high-intensity signal on T2 weighted MRI [26]. Arteriography is diagnostic in most cases, but may fail to show the feeding arteries of an intramuscular haemangioma if it is supplied by small arteries with slow blood flow [5, 6, 7]. It must be performed if surgical intervention is considered to supply definitive information about the nature of the tumour. Preoperative embolisation can also be performed during preoperative angiography to minimize blood loss [7].

Although some authors believe that FNA is commonly non-diagnostic, showing the presence of a purely bloody specimen may be considered strongly suggestive for intramuscular haemangioma as seen in our case [27–29]. FNA should also be performed to aid in the exclusion of other soft tissue tumours [26].

Management of intramuscular haemangioma should be individualized according to the size and anatomic accessibility of the tumour, its growth rate, age of the patient and cosmetic and functional considerations [5, 28]. If indicated, complete surgical resection is preferred, but local recurrence rates have been reported as 18% and 19% in two different studies [6, 30]. Alternatives to surgical excision of intramuscular haemangioma are embolisation and radiotherapy, with the treatment of choice being decided by the site of the lesion. But it has been reported that modalities such as embolisation, injection of sclerosants and radiotherapy have had limited success [31]. Unlike cutaneous haemangioma, the intramu-

scular haemangioma do not regress spontaneously, and they do not seem to be sensitive to systemic corticosteroid therapy [11, 32]. In view of the clinical appearance and the age of the patient, we decided to wait and watch her until adolescence and consider excision if the tumour shows more invasive growth and causes cosmetic problems and severe pain.

Conclusion

Intramuscular haemangioma may start to grow in childhood and should be considered in the differential diagnosis of isolated muscle enlargement. MRI and colour Doppler sonography are very helpful in diagnostic workup and the treatment of choice should be individualized in view of the clinical status of the patient.

References

1. Mulliken JB, Glowacki J (1982) Hemangiomas and vascular malformations in infants and children: a classification based on endothelial characteristics. Plast Reconstr Surg, 69: 412–422.
2. Enjolras O, Mulliken JB (1993) The current management of vascular birthmarks. Pediatr Dermatol, 10: 311–313.
3. Meyer JS, Hoffer FA, Barnes PD, Mulliken JB (1991) Biological classification of soft tissue vascular anomalies: MR correlation. AJR Am J Roentgenol, 157: 559–564.
4. Dubois J, Garel L (1999) Imaging and therapeutic approach of hemangiomas and vascular malformations in the pediatric age group. Pediatr Radiol, 29: 879–893.
5. Terezhalmy GT, Riley CK, Moore WS (2000) Intramuscular hemangiomas. Quintessence Int, 31: 142–143.
6. Wolf GT, Daniel F, Krause CJ, Arbor A, Kaufman RS (1985) Intramuscular hemangiomas of the head and neck. Laryngoscope, 95: 210–213.
7. Rossiter JL, Hendrix RA, Tom LW, Potsic WP (1993) Intramuscular hemangiomas of the head and neck. Otolaryngol Head Neck Surg, 108: 18–26.
8. Gampper TJ, Morgan RF (2002) Vascular anomalies: hemangiomas. Plast Reconstr Surg, 110: 572–585.
9. Gilett D, Fahmy F, Eveson JW, Shotton JC (2003) Intramuscular capillary hamartoma of the tongue. J Laryngol Otol, 117: 734–735.
10. Christensen SR, Borgesen SE, Heegaard S, Prause JU (2002) Orbital intramuscular haemangioma. Acta Ophtalmol Scand, 80: 336–339.
11. Kiratli H, Bilgic S, Caglar M, Soylemezoglu F (2003) Intramuscular hemangiomas of extraocular muscles. Ophtalmology, 110: 564–568.
12. Makeieff M, Maurice N, Mondain M, Crampette L, Guerrier B (2001) Intramuscular hemangioma of posterior neck muscles. Eur Arch Otorhinolaryngol, 258: 28–30.
13. Wild AT, Raab P, Krauspe R (2000) Hemangioma of skeletal muscle. Arch Orthop Trauma Surg, 120: 139–143.
14. Kohout MP, Hansen M, Pribaz JJ, Mulliken JB (1998) Arterivenous malformations of the head and neck: natural history and management. Plast Reconstr Surg, 102: 643–654.
15. Paltiel HJ, Burrows PE, Kozakewich HPW, Zurakowski D, Mulliken JB (2000) Soft-tissue vascular anomalies: utility of US for diagnosis. Radiology, 214: 747–754.
16. Burrows PE, Laor T, Paltiel H, Robertson RL (1998) Diagnostic imaging in the evaluation of vascular birthmarks. Dermatol Clin, 16: 455–488.
17. Buetow PC, Kransdorf MJ, Moser RP Jr, Jelinek JS, Berrey BH (1990) Radiologic appearance of intramuscular hemangioma with emphasis on MR imaging. AJR Am J Roentgenol, 154: 563–567.
18. Chan MJ, McLean NR, Soames JV (1992) Intramuscular hemangioma of the orbicularis oris muscle. Br J Oral Maxillofac Surg, 30: 192–194.
19. Sharma BS, Chari PS, Joshi K, Rajvanshi A (1991) Hemangioma of the temporalis muscle. Ann Otol Rhinol Laryngol, 100: 76–78.
20. Stofman GM, Reiter D, Feldman MD (1989) Invasive intramuscular hemangiomas of the head and neck. Ear Nose Throat J, 68: 612–616.
21. Ariji Y, Kimura Y, Gotoh M , Sakuma S, Zhao YP, Ariji E (2001) Blood flow in and around the masseter muscle: normal and pathologic features demonstrated by color Doppler sonography. Oral Surg Oral Med Oral Pathol Oral Radiol Endod, 91: 472–482.
22. Dubois J, Patriquin HB, Garel L et al (1998) Soft-tissue hemangiomas in infants and children: diagnosis using Doppler sonography. AJR Am J Roentgenol, 17: 247–252.
23. Yang WT, Ahuja A, Metreweli C (1997) Sonographic features of head and neck hemangiomas and vascular malformations: review of 23 patients. J Ultrasound Med, 16: 39–44.
24. Kudawara I, Yoshikawa H, Araki N, Ueda T (2001) Intramuscular haemangioma adjacent to the bone surface with periosteal reaction. Report of three cases and review of the literature. J Bone Joint Surg Br, 83: 659–662.
25. Hawnaur JM, Whitehouse RW, Jenkins JP, Isherwood I (1990) Musculoskeletal hemangiomas: comparison of MRI with CT. Skeletal Radiol, 19: 251–258.
26. Clement WA, Graham I, Ablett M, Rawlings D, Dempster JH (2002) Intramuscular hemangioma of the posterior belly of the digastric muscle failing to highlight on magnetic resonance imaging. Ann Otol Rhinol Laryngol, 111: 1050–1053.
27. Odabasi AO, Metin KK, Mutlu C, Basak S, Erpek G (1999) Intramuscular hemangioma of the masseter muscle. Eur Arch Otorhinolaryngol, 256: 366–369.
28. Heckl S, Ascoff A, Kunze S (2002) Cavernous hemangioma of the temporal muscle. Neurosurg Rev, 25: 63–67.
29. Giudice M, Piazzo C, Bolzoni A, Peretti G (2003) Head and neck intramuscular hemangioma: report of two cases with unusual localization. Eur Arch Otorhinolaryngol, 260: 498–501.
30. Tang P, Hornicek FJ, Gebhardt MC, Cates J, Mankin HJ (2002) Surgical treatment of hemangiomas of soft tissue. Clin Orthop, 399: 205–210.
31. Quinn PS, Siemnarine K, Lawrence-Brown M, Tan P (2001) Intramuscular hemangiomas: hookwire localization prior to surgical excision: report of four cases. ANZ J Surg, 71: 62–66.
32. Edgerton MT (1976) The treatment of hemangiomas: with special reference to the role of steroid therapy. Ann Surg, 183: 517–532.