RCS
ADVANCING SURGICAL STANDARDS

ONLINE CASE REPORT

Ann R Coll Surg Engl 2014; **96:** e15–e17
doi 10.1308/003588414X13946184903126

Intramuscular haemangioma of the tongue

D Babu, R Bhamre, R Katna, P Pai

Tata Memorial Hospital, Mumbai, India

ABSTRACT

Haemangiomas are one of the most common benign tumours. Clinicians come across haemangiomas of different subtypes at different locations in the body. They are often faced with the question of whether to treat them or leave it to the natural history of the disease. We present a case of the intramuscular variety of haemangioma found in the unusual location of the tongue in a 60-year-old woman. Fine needle aspiration was inconclusive and on magnetic resonance imaging, it mimicked a malignancy, which prompted treatment. We also review the unique pathology of this variety of haemangioma, which defines their treatment. The radiological attributes of the disease and recurrence rates of surgery are also discussed.

KEYWORDS

Intramuscular haemangioma – Pathology – Surgical excision

Accepted 4 April 2014; published online XXX

CORRESPONDENCE TO

Donald Babu, E: donaldbabu@hotmail.com

Case History

A 60-year-old woman presented to us with swelling in the dorsum of the tongue that she had had for 5 years (Fig 1). The swelling was well defined, 4cm × 3cm in diameter and soft to firm in consistency, non-compressible, non-mobile with no restrictions to the mobility of the tongue. Fine needle aspiration (FNA) of the swelling revealed only acellular fluid. Magnetic resonance imaging (MRI) showed a lesion that was hypointense on T1 weighted sequences and hyperintense on T2 weighted sequences with postcontrast enhancement. The lesion had irregular margins but there was no invasion of the base of the tongue or floor of the mouth and it did not cross the midline (Fig 2). However, the MRI did not give a definitive diagnosis.

Owing to the diagnostic dilemma, we opted for surgical excision. The routine investigations (including haemoglobin, differential blood counts, platelets and coagulation profile) were normal. The lesion being small and accessible, the patient was operated in a day-case setting under general anaesthesia with nasal intubation. Since MRI did not reveal any proximity to vessels and the bleeding profile was normal, bleeding was unexpected. Surgical excision was performed with wide margins and the cavity was closed primarily with absorbable sutures along the long axis of the tongue.

The procedure was uneventful, and the patient was discharged and called for follow-up visits on the third and tenth postoperative days. Histopathology revealed non-keratinised stratified squamous epithelium of the tongue and a nest of cells termed von Ebner's serous glands of the tongue. A magnified view of the staining results showed the 'pseudoinfiltrative' pattern of the intramuscular haemangioma (Fig 3).

Discussion

Haemangiomas of skeletal muscle represent 0.8% of all benign vascular neoplasms.[1] Of these, 13.8% occur in the head and neck region, with the masseter muscle being the most common site, followed by the trapezius and sternocleidomastoid muscles respectively.[2] A variety of tumours can be confused clinically with an intramuscular haemangioma (IMH), and the differential diagnoses include salivary neoplasms, cysts, lymphangiomas, rhabdomyosarcomas and schwannomas.

Haemangiomas can have varied histology with capillary and cavernous haemangiomas being the most common. They are never encapsulated. An IMH shows capillaries in a loose fibrous stroma interspersed between striated muscle bundles in a pseudoinfiltrative fashion that may mimic malignancy. Vessel lumens are usually well developed in this tumour but occasional cases show a more solidly cellular appearance. Mitotic activity is usually not pronounced and intraluminal tufts of endothelial cells may be seen to project into vessel lumens. Capillaries may be seen to proliferate in perineural sheaths.[3] However, these tumours are clearly benign. They can be distinguished from angiosarcomas by the presence of a two-cell layer (endothelial and perithelial) and by formation of round to elongated vascular spaces without anastomosis.[4]

There are three subtypes of IMH described in the Allen and Enzinger classification, and they vary in the clinical presentation, histopathological findings and recurrence rate.[5] In the IMH capillary variant, accounting for 68% of lesions, there is often a brief clinical history of localised, painless swelling, usually smaller than 10cm, with a histological pattern of small vessel vascularisation and a recurrence rate of

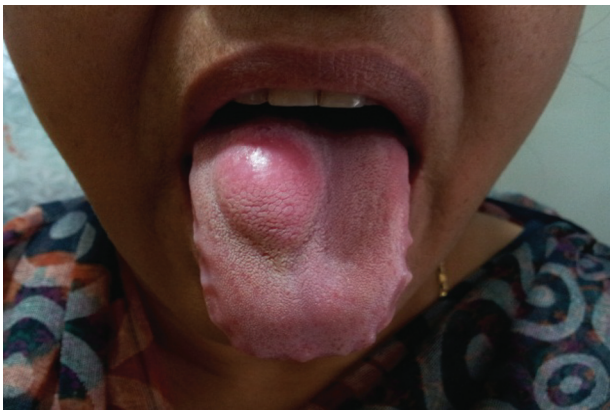

Figure 1 Intramuscular haemangioma in the central part of the tongue

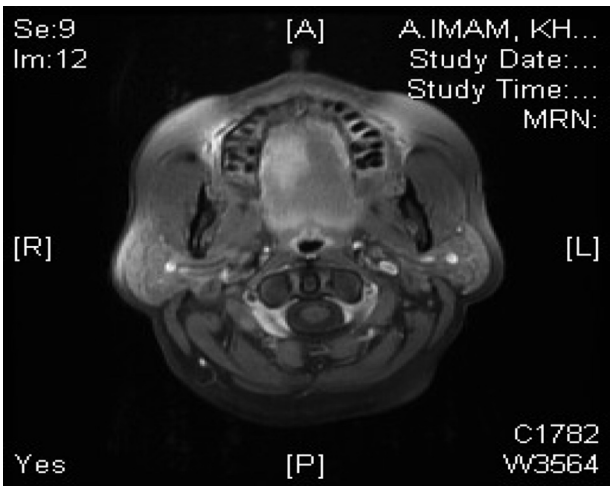

Figure 2 T2 weighted magnetic resonance imaging showing intramuscular haemangioma of the tongue. In the image it is seen as a triangular hyperechoic lesion as compared with the surrounding hypoechoic tongue tissue. The lesion shows fluid content without any solid component (hyperechoic on T2 imaging).

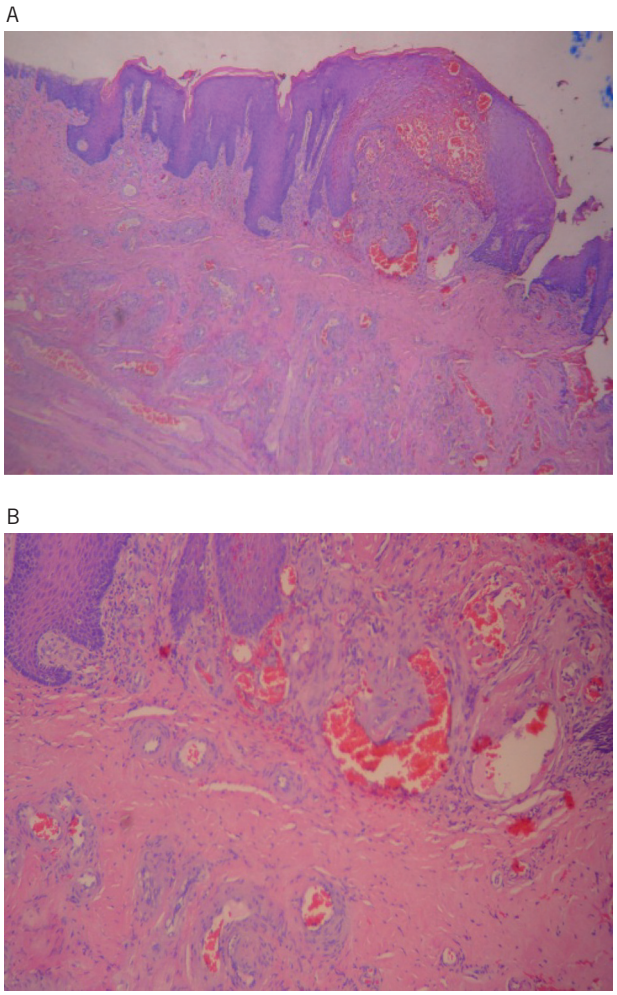

Figure 3 Haematoxylin and eosin staining of the tongue: The non-keratinised stratified squamous epithelium of the tongue is seen in violet (A). Below this layer are the intrinsic muscles of the tongue in pink, which contain a nest of cells in lavender. These are von Ebner's serous glands of the tongue. The prominent largest cavity in red is the haemangioma. The magnified view of the haemangioma (B) shows capillaries separated out in the muscular tissue (small vessels around the main vessels). This is the 'pseudoinfiltrative' pattern of the intramuscular haemangioma.

about 20%. Recurrence rate is often related to incomplete surgical excision.[6] In the cavernous type (26%), symptoms usually last longer, the lesion is bigger in size, the vessels have larger diameters and the recurrence rate is less than 9%. In the mixed type (6%), there is a combination of capillary and large vessels, and the incidence of recurrence is the highest reported in about 28% of cases.

FNA is inconclusive in arriving at a diagnosis as it yields only a bloody aspirate. MRI of haemangiomas is characterised by a combination of large vessels with slow flow and non-vascular tissues, which are predominantly fat and fibrous elements. Typical MRI features are isointensity on T1 and heterogeneously increased signal intensity on T2 weighted sequences related to the skeletal muscle. The margins are poorly delineated on T1 but are well defined on T2 imaging. On contrast enhanced T1 weighted imaging, marked enhancement is almost always noted. On T2 weighted MRI, there are also rounded foci of low signal intensity areas that are suggested as highly specific for haemangiomas and may be representing fibrofatty septa seen in cross-section, smooth muscle components, calcification, ossification, or hyalinised or thrombosed vascular channels.

As the imaging characteristics of non-vascular elements in the lesion and the extension of the mass are well demonstrated on MRI, it has been proposed as the examination of choice in the evaluation of deep soft tissue haemangiomas. Computed tomography (CT) can show the mass frequently isodense to the skeletal muscle with decreased attenuated areas representing fat and it also demonstrates clearly the calcification or ossification if present. Nevertheless, the margins of the lesion are generally poorly delineated on CT.[7–9]

Many treatment modalities like cryotherapy, radiation, steroids and embolisation have been advocated but the main treatment at present remains surgical excision.[1,2] Owing to lack of encapsulation and the infiltrative nature of IMH, surgical excision including an adequate rim of surrounding healthy tissue is the treatment of choice for the management of such lesions despite local recurrences ranging from 9% to 28%, even after wide resection of a cuff of normal muscle around the tumour.[2]

In a study by Bella *et al* of 110 patients treated for IMH with various modalities over 25 years, Kaplan–Meier analysis showed 76% of patients managed without surgery were disease free at 2 years and 66% at 5 years.[10] For patients treated with surgery, 86% and 73% were recurrence free at two and five years respectively. There were substantial differences in local recurrence when stratified by margin: 93% of patients were recurrence free at five years when the excision was marginal and wide, 65% when intralesional without any gross remaining tumour and 33% when intralesional with gross remaining tumour. Surgical margins and tumour size were the only identified risk factors for recurrence.

Conclusions

Head and neck IMH is a rare, unencapsulated, vascular neoplasm characterised frequently by a slow and painless growth. This presents difficulties in correct preoperative diagnosis and treatment planning. The recurrence rate is high, even after wide surgical excision, owing to its microscopically infiltrative pattern of diffusion into the surrounding muscular tissue along the planes of least resistance. Nevertheless, surgical resection well beyond the gross limits of the tumour is generally considered the most appropriate therapeutic strategy. Long-term clinical and radiological follow-up is strongly recommended for early diagnosis and to treat further recurrences.

References

1. Watson WL, McCarthy WD. Blood and lymph vessel tumors; a report of 1056 cases. *Surg Gynecol Obstet* 1940; **71**: 569–588.
2. Wolf GT, Daniel F, Krause CJ, Kaufman RS. Intramuscular hemangioma of the head and neck. *Laryngoscope* 1985; **95**: 210–213.
3. Rossiter JL, Hendrix RA, Tom LW, Potsic WP. Intramuscular hemangioma of the head and neck. *Otolaryngol Head Neck Surg* 1993; **108**: 18–26.
4. Mills SE. *Sternberg's Diagnostic Surgical Pathology*. 5th edn. Baltimore: Lippincott Williams & Wilkins; 2010.
5. Allen PW, Enzinger FM. Hemangioma of skeletal muscle. An analysis of 89 cases. *Cancer* 1972; **29**: 8–22.
6. Beham A, Fletcher CD. Intramuscular angioma: a clinicopathological analysis of 74 cases. *Histopathology* 1991; **18**: 53–59.
7. Teo EL, Strouse PJ, Hernandez RJ. MR imaging differentiation of soft-tissue hemagiomas from malignant soft-tissue masses. *Am J Roentgenol* 2000; **174**: 1,623–1,628.
8. Greenspan A, McGahan JP, Vogelsang P, Szabo RM. Imaging strategies in the evaluation of soft-tissue hemangiomas of the extremities: correlation of the findings of plain radiography, angiography, CT, MRI, and ultrasonography in 12 histologically proven cases. *Skeletal Radiol* 1992; **21**: 11–18.
9. Buetow PC, Kransdorf MJ, Moser RP *et al*. Radiologic appearance of intramuscular hemangioma with emphasis on MR imaging. *Am J Roentgenol* 1990; **154**: 563–567.
10. Bella GP, Manivel JC, Thompson RC *et al*. Intramuscular hemangioma: recurrence risk related to surgical margins. *J Clin Orthop Relat Res* 2007; **459**: 186–191.