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Case Report

Intramuscular haemangioma of the masseter muscle: a case report

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Abstract Intramuscular haemangioma is relatively rare and accounts for less than 1% of all haemangioma. Most of these tumours occur in the trunk, arms and legs. Only 14% of all intramuscular haemangioma arise from the head and neck region, with masseter muscle representing the most common site of involvement, followed by the trapezius and sternocleidomastoid muscles. We present a case of a 60-year-old man with a soft lump in the left cheek. Imaging of the neck showed irregular, dilated enhancing vessels seen in the left masseter muscle suggestive of intramuscular haemangioma. Excisional biopsy was performed, and histopathological findings confirmed the diagnosis.

Keywords: Face; haemangioma; intramuscular; masseter muscle; vascular.

Introduction

Haemangiomas are vascular neoplasm which represent 7% of all benign tumours. It occurs most frequently on cutaneous and mucosal surfaces but rarely observed in the skeletal muscles of head and neck region (Kim *et al.*, 2007). Skeletal muscles haemangiomas or intramuscular haemangiomas only make up 0.8% of all haemangiomas (Narayanan *et al.*, 2009). Patient may present with pain, discomfort and gradual enlargement of the lesion. Clinically, these lesions are seldom correctly diagnosed, mainly because of the rarity, deep seated site and uncommon presentation (Bucci *et al.*, 2008). Thus, intramuscular haemangioma should be considered in the differential diagnosis of any presentation of a nonspecific soft tissue swelling.

Case report

A 60-year-old man was referred with a painless and progressively enlarging lump in the left cheek region for the past 3 years. On clinical examination, a smooth, oval swelling with size of 3 cm in diameter was noted in the left cheek region (Fig. 1). It was well-circumscribed, soft, non-tender and mobile which becomes prominent on clenching. There was no compressibility and the overlying skin was normal. Intraoral examination revealed no abnormality and there was no facial nerve involvement. Provisional diagnosis of the cheek cystic mass was made with differential diagnosis of other soft tissue lesions like lipoma, dermoid cyst, vascular malformation or benign tumour of parotid gland.

A contrast-enhanced CT scan revealed an irregular serpinginious dilated enhancing vessels seen in the left masseter muscle measuring approximately 2.9 cm x 2.6 cm x 1.8 cm. Some of the vessels appear to arise from the branch of left internal jugular vein, however the arterial component was not seen. Two foci of calcifications were observed adjacent to this lesion (Fig. 2). Both the parotid and submandibular glands are normal with no focal enhancing lesion. No significant cervical lymph nodes were

bilaterally observed. Based on the imaging studies, the diagnosis of haemangioma of left masseter muscle was made.

Excisional biopsy of the lesion was planned under general anaesthesia. Incision was made over the lesion. Skin flaps were raised, and dissection was performed through the masseter muscle to expose the well encapsulated lesion. Once the lesion is exposed, a blunt dissection was done around the lesion and cauterization was done for any bleeding vessels. The mass was completely removed with a margin of normal surrounding muscles to prevent recurrence. Primary closure was done. Blood loss during the procedure was minimal. The whole exposed specimen was sent for histopathological examination. Post-surgically, he was prescribed with antibiotics and analgesics for a week.

Histological examination revealed fibrous tissue composed of numerous dilated vascular spaces containing blood clots and occasional thrombi (Fig. 3). There was also proliferation of small and capillary sized of blood vessels in between. These blood vessels were lined by single layer of endothelial cells. There was no evidence of malignancy. The findings were consistent with the features of a haemangioma.

Post-surgical excision showed complete absence of the swelling in the left submandibular region and no recurrence was noted during the 6-month follow-up.

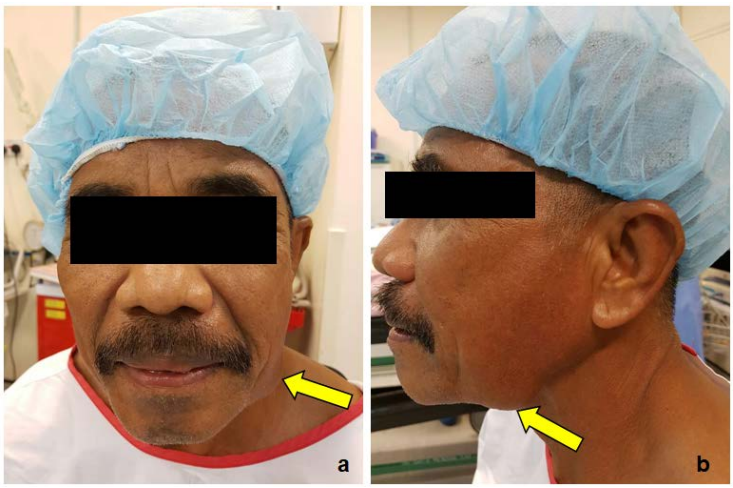

Fig. 1 Cystic mass located at the lower part of left cheek, seen from anterior view (1a) and lateral view (1b).

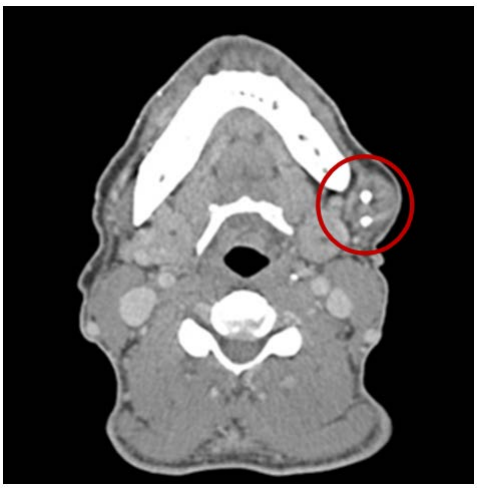

Fig. 2 Contrast CT revealed some irregular dilated enhancing vessels seen in the left masseter muscle (red circle).

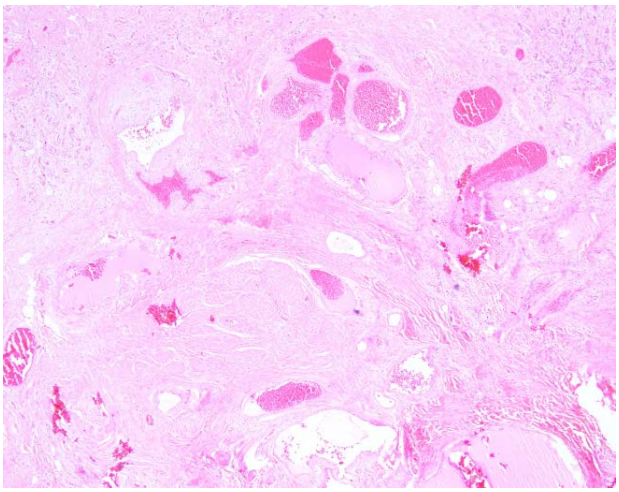

Fig. 3 Numerous dilated blood vessels containing blood with some small capillary-sized blood vessels (H&E, 4x/0.10).

Discussion

Intramuscular haemangiomas were first described by Liston in 1843. This benign vascular lesion of skeletal muscles makes up for less than 1% of all haemangiomas (Kim *et al.*, 2007). Intramuscular haemangioma is commonly present in early childhood or early adult life, although there were some studies which reported such cases in elderly patients (Welsh and Hengerer, 1980). Generally, intramuscular hemangioma was shown to have an equal sex distribution except for the involvement of the masseter muscle which has a definite male predominance with ratio of 3 to 1 (Hoehn *et al.*, 1970).

Histologically, haemangioma is classified as capillary (vessels smaller than 140 micrometres in diameter), cavernous (vessels larger than 140 micrometres in diameter) and mixed (consisting of both small and large vessels) (Allen and Enzinger, 1972).

Capillary haemangioma usually presented with a short history (Ozer and Schuller, 2006). It is highly cellular which explains its solidity and lack of clinical signs to demonstrate the vascular nature. On the other hand, the cavernous hemangioma generally presents with longer history of symptoms and tend to be larger in size with accompanied pain. Cavernous type is most common in the lower extremity with only 19% occurring in the head and neck region (Watson and McCarthy, 1970). Whereas the mixed type is histologically and clinically similar to the cavernous type. We find the cavernous classification most suggestive in the present case study as evidenced by our clinical and histological findings.

Etiopathogenesis of intramuscular haemangioma remains unclear. However, there are several theories which have been proposed to explain its pathogenesis. The most likely explanation is that the intramuscular haemangioma is a congenital mass which is derived by the abnormal embryonic sequestrations. This aetiology is similar to the congenital arteriovenous malformations (Scott, 1957). Trauma have also been suggested and may give rise to the aetiology or growth spurts of the lesion (Welsh and Hengerer, 1980). Hormonal role in the growth of intramuscular haemangioma was postulated, but there is no specific data available to validate this hypothesis (Wolf *et al.*, 1985).

Clinically, these tumours present as a gradually enlarging mass lesion with duration often less than a year. The swelling is normally diffuse in nature, compressible and typically deep within the muscle. However, the mass may not be soft and compressible due to local fibrosis and overlying musculature. The prominent cellularity of capillary type tumours would also conceal the clinical findings. Pulsations, bruits or thrills are unusual but when present, arteriography is suggested to discover any large vessel communications (Wolf *et al.*, 1985). There are usually no skin changes. Clenching the teeth could cause the lesion to become more fixed and prominent (Wolf *et al.*, 1985).

Diagnosis of intramuscular haemangioma is often challenging. Diagnosis by FNAC is usually inconclusive as it only shows blood-tinged aspirate (Terezhalmy *et al.*, 2000). Super selective arteriography with subtraction would clearly demonstrate the changes in vascular pattern and flow dynamics including feeder vessels. Imaging studies such as contrast-enhanced CT may demonstrate vascular nature of the tumour as in the present case where some irregular dilated enhancing vessels was noted. And because of the multiplanar capability, MRI can display good tissue delineation and contrast of the lesion from its adjacent tissue.

Management of intramuscular haemangioma should be categorized according to the tumour location, extension, tumour growth rate, anatomical approachability, patient age and aesthetic concern (Hawnaur *et al.*, 1990). Some of the patients can be kept under observation with the reliability of the MRI especially in young children. Many treatment options like cryotherapy, radiation therapy, steroid administration and embolization, sclerosing agents, carbon dioxide snow and blood vessel ligation have been advocated (Wolf *et al.*, 1985), but the treatment of choice at present remains surgical excision as in the present case.

The indications for surgery include symptomatic tumours, sudden rapid acceleration of tumour growth, gross functional impairment, local skin necrosis, thrombocytopenia, cosmetic deformity and suspicious of malignancy (Terezhalmy *et al.*, 2000). Difficulty in intraoperative localization of the exact extent of the tumour due to its nature and the absence of a definite capsule justifies a complete excision of the muscle. Cosmetic and functional disabilities after excision have been minimal even after significant removal of surrounding normal muscle (Terezhalmy *et al.*, 2000). Local recurrences occur in approximately 18% due to incomplete surgical resection (Wolf *et al.*, 1985). Spontaneous regression does not occur. Regional and distant metastasis has not been reported.

Conclusion

Intramuscular haemangiomas are rare in the head and neck region, and it should be considered in the differential diagnosis of masses in these regions. Its diagnosis is difficult because of its rarity, nonspecific signs and frequent absence of typical signs of haemangioma such as pulsation and noise. The knowledge of the nature and recurrence rate of an intramuscular haemangioma is useful for appropriate management.

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