

INTRAMASSETRIC HAEMANGIOMA ASSOCIATED WITH A PHLEBOLITH

Karthik Rajaram Mohan, Saramma Mathew Fenn, Ravikumar Pethagounder Thangavelu and Kumar Appusamy

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Abstract

Intramuscular hemangiomas are rare, accounting for less than 1% of all hemangiomas, and occur normally in the trunk and extremities. Intramassetric haemangioma constitute approximately about 10-20% in the head and neck region, most often in the masseter muscles. The typical clinical characteristic is a painless soft tissue mass resulting in unaesthetic appearance without cutaneous changes. Such a case of intramassetric Haemangioma in a 21-year-old male within the right masseter muscle associated with a phlebolith is presented here.

Keywords: Intramuscular haemangioma, Masseter, MRI, Phlebolith, Ultrasonography

Introduction

The Intramuscular Haemangiomas occurring within the massetric muscle accounts for about 10-20% of all Haemangiomas occurring in the Head and neck region. (Alami *et al.*, 2015).

Such haemangiomas within the masseter muscle are present from birth, usually unilateral but become clinically apparent only after pubertal age and usually do not recur spontaneously. The only complaint was that of an unaesthetic appearance of the face. They appear as painless compressible swellings and becomes more apparent clinically during clenching of the teeth. Since they are located deep, they neither clinically present as reddish or purplish coloured swellings nor do they present as any involution in the form of telangiectasia on the external surface of the skin.

Case Report

A 21-year-old-male reported to our outpatient department of oral medicine and radiology with a chief complaint of unaesthetic appearance due to the presence of swelling on the right side of the face. (Figure 1). Patient was afebrile and his vitals were stable. No history of similar swelling was present among his family members. Patient did not have any deleterious habits of chewing or smoking tobacco. History revealed the swelling started as a small one from the time of his birth and has progressed in size to the present state. Extraoral examination revealed a swelling seen on the right angle just above the lower border of mandible which measured approximately 4.5×2.5cm near the lower border of the angle of the mandible, extending superiorly 2cm

Karthik Rajaram mohan, Vinayaka Mission's Sankarachariyar Dental College, Vinayaka Mission's Research Foundation (Deemed to be University), Ariyanur, Salem-636308, Tamilnadu, India. E-mail: drkarthik@vmsdc.edu.in

* Corresponding author

away from right ala-tragal plane, inferiorly extends 0.5 cm below the right-side angle of the mandible, anteriorly the swelling extends 3 cm away from right corner of vermillion border of lip, posteriorly it extends 0.5 cm in front of tragus of right ear. All the inspectory findings such as number, site, size, shape, surface and extent were confirmed on palpation. The swelling was firm in consistency and non-tender, Compressible and reducible. There was no warmth felt on palpation of the swelling and was negative to transillumination. No other secondary changes like sinus, punctum, erythema or ulcer were seen over the surface of the swelling except a visible scar. A scar was seen on the surface of the skin near the right masseter muscle region just above the angle of mandible. The scar was because, he was operated for similar swelling within the right masseter muscle region, which occurred 1 year back. He underwent microvascular surgery for similar swelling within the right masseter muscle region 1 year back. The swelling becomes more prominent during clenching of his teeth, suggesting that the swelling arises deep within the muscle planes. (Figure 2).



Figure 1. Extraoral profile



Figure 2. Swelling becomes prominent during clenching and a surgical scar

Correlating the history, clinical findings a provisional diagnosis of massetric Hypertrophy is made. Haemangiomas affecting the masseter muscle are usually painless on Palpation. Massetric

hypertrophy usually occur bilaterally and affects both the right and left masseter muscles. whereas cavernous haemangiomas usually seen unilaterally in relation to masseter muscle. Radiologically the differential diagnosis of calcifications include parotid sialolith or calcified parotid lymphnode. Parotid sialolith occur mostly singly mostly ipsilaterally whereas phleboliths usually appear in multiple areas and contralateral locations. Ultrasonography revealed areas of calcification suggestive of Phlebolith in relation to the right masseter muscle. (Hwang *et al.*, 2019) stated that Intramassetric schwannomas can be misdiagnosed as intramassetric haemangiomas. Colour Doppler ultrasonography helps to identify whether there is high flow or low flow within the lesion. MRI is the gold standard imaging technique for diagnosing intramassetric Haemangiomas. Sagittal section of MRI revealed a large unencapsulated, well defined, lobulated, smooth marginated soft tissue lesion in the right masticator space deep to right masseter muscle. (Figure 3). The lesion is uniformly hypointense when compared to the adjacent tissue planes in T1 weighted (Figure 5 and Figure 7) and hyperintense in T2 weighted images. There were multiple hypodense (dark) areas seen within the lesion suggestive of phleboliths. The lesion within the right massetric region measuring approximately 4cm anteroposteriorly $\times$ 2.6cm mediolaterally and 4.3cm craniocaudally appears bright in T1 weighted (Figure 4 and Figure 6) and low in T2 weighted image (Figure 5 and Figure 7) with no erosion of underlying bone, suggestive of intramassetric cavernous haemangioma. The diagnosis in our case was confirmed only by magnetic resonance imaging and patient was not willing for treatment as it was recurrent in nature.



Figure 3. MRI-T1 sagittal section showing hypointense signal within masseter muscle region

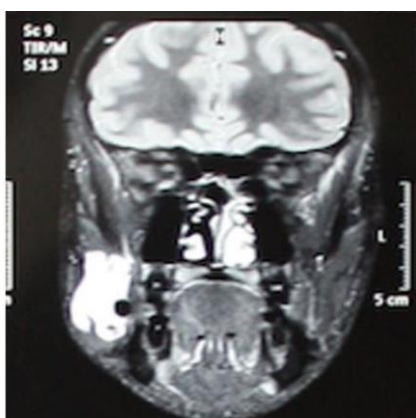


Figure 4. MRI-Coronal T1-showing hypointense phlebolith and hyperintense signal in the right masseter muscle region

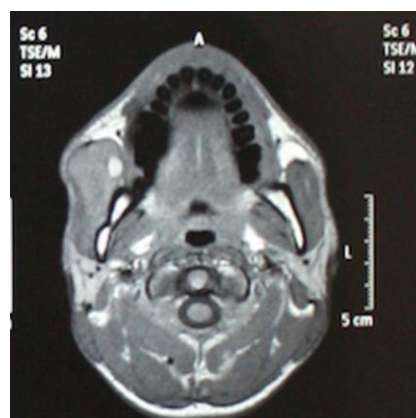


Figure 7. MRI-T2-Axial image showing hyperintense Phlebolith

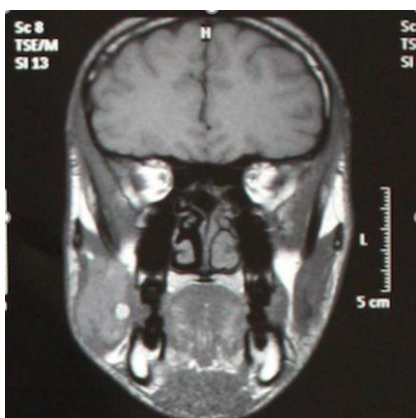


Figure 5. MRI-Coronal T2-showing hyperintense phlebolith and hypointense signal in the right masseter muscle region

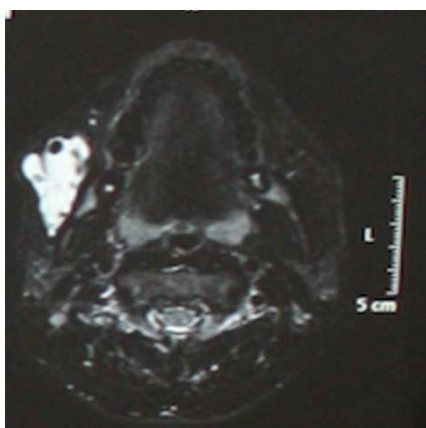


Figure 6. MRI-T1-Axial section showing hypointense phlebolith and hyperintense signal within the right masseter muscle

Discussion

Rossiter *et al.* (1993) Intramasseteric haemangiomas are uncommon, about 36% occur in the masseteric region. They are probably congenital but not usually recognized until second or third decade. (Hwang *et al.*, 2019) stated that Intramasseteric Haemangiomas clinically appear as painless, slow growing, firm or compressible swellings. They are unlikely to be recognized clinically unless if superficial, the overlying skin is purplish red and warmth to touch. Intramasseteric Haemangiomas do not appear to regress spontaneously. They are best delineated by MRI Scans. Angiography also helps to diagnose it but indicated only when large vascular connections are suspected. The recurrence rate after Surgical excision is usually high 61%. Newer modalities like laser or cryotherapy can be indicated as treatment of choice for intramasseteric haemangiomas. (Scott, 1957) in a study of 393 cases on Haemangiomas, only 4.89% occurred in the masseter muscle. (Hart and Schwartz, 1995) Intramasseteric Haemangioma is a benign congenital hamartomatous tumour without racial or genetic determinism. 90% of the cases were diagnosed before the age of 30. Some important etiological factors considered in Intramasseteric haemangioma include Excessive muscle contraction or trauma; hormonal factors also been found to play a role in intramasseteric haemangioma, because of its occurrence correlated to menarche, pregnancy and menstrual cycle accounts for the slight female predominance. (Meziane *et al.*, 2010) stated that, In males, about 70% of the cases are seen localised within the masseter muscle. Intramasseteric

Haemangioma presents clinically as a single firm mass within the masseter muscle. More than a half of cases rarely have associated intermittent pain, due to muscle compression. The deep location and surrounding fibrosis of intramasseteric haemangioma masks the classical clinical description of hemangioma as a blue, soft, compressible tumor. Bruit or thrill are exceptional on auscultation, which usually gets masked by the thickness of the superficial muscular layer. Lowering the head increases the chance of occurrence of Bruit or thrill-the Wattle sign, which are found only in superficial and large intramasseteric Haemangioma. (Agarwal *et al.*, 2016) described the clinico-radiological spectrum of imaging of masseteric haemangioma. (Alami *et al.*, 2015) Conventional radiographs may detect Phleboliths but needs to be distinguished from intraparotid sialoliths or calcified intraparotid lymphnode. Echo-Doppler ultrasonography aids in assessing the vascular nature of the intramasseteric haemangioma, but cannot precisely determine its boundaries, which can be determined only with contrast-enhancement on MRI. Fine needle aspiration is contraindicated because of the risk of severe haemorrhage that can be encountered in such lesions. (Righini *et al.*, 2014) Non-surgical treatment modalities for the treatment of intramasseteric haemangioma include arterial ligation of the feeding vessels and embolization by the use of sclerosing agents like polyvinyl alcohol or polidoconal solutions. (Capote *et al.*, 2008) suggested infratemporal-preauricular-cervical approach for resection of cavernous hemangioma. (Cheng and Lai, 2016) described Transcervical excision as a treatment of choice for Intramasseteric cavernous Haemangioma. It is very much essential to preserve facial nerve during such surgery which are in close proximity to the masseter muscle. (Azma *et al.*, 2018) stated that Intense pulsed light lasers are effective in the treatment of cavernous haemangiomas. The aim of laser therapy for hemangioma is the selective destruction of tumoral vessels through the absorption of laser photons by hemoglobin molecules in red blood cells. Thermal energy of laser photons scatters radially within the blood vessel inducing selective microvascular hazardous effects on the endothelium lined blood vessels within the masseter muscle, through photocoagulation and causes mechanical injury. (Bella *et al.*, 2007) The recurrence rate of intramasseteric Haemangioma ranges from 18% to 61%.

Conclusions

Unilateral enlargement of swelling within the masseter muscle must not be overlooked as a lymph node enlargement and warrants other specialized imaging technique like colour doppler ultrasound and MRI. MRI is the gold standard for diagnosis of such intramasseteric haemangiomas which clinically present as a painless, slowly enlarging, compressible, reducible, non-transilluminant mass within the masseter muscle without any other clinical signs and symptoms. Patient affected by intramasseteric haemangioma visit a dentist or a general physician only for unaesthetic facial appearance. A patient with facial asymmetry must not be overlooked by clinician and must be thoroughly examined by proper history taking, clinical and radiological investigations that aids in diagnosis of such a rare entity such as intramasseteric haemangioma. A thorough clinical history and clinical examination and radiological investigation facilitates the dentist in their treatment planning.

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Statement of Ethics: written informed consent was obtained from the patient for publication of this case report and accompanying images.

Conflict of Interest Statement: The Authors have no conflicts of interests to declare.

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