

Masseter Muscle Haemangioma with Calcifications Showing Turkey Wattle Sign: A Case Report with Review of Literature

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ABSTRACT

Haemangiomas are unusual benign vascular tumours affecting skeletal muscle (masseter) in less than 1% of cases in the head and neck region. An intramuscular haemangioma of the masseter muscle is often mistaken for a parotid swelling. This case report emphasises the importance of investigations for accurate preoperative diagnosis, along with a literature review. A 43-year-old male patient presented with a chief complaint of swelling on the lower right side of the face for the past six years. Examination revealed a diffuse swelling on the right lower third of the face, extending anteroposteriorly from the right corner of the mouth to the right tragus of the ear. Superoinferiorly, the lesion extended from the right ala tragus line to the inferior border of the mandible. On palpation, the swelling was non-tender, soft, and less compressible, with no pulsations felt. Furthermore, the swelling appeared more prominent during contraction of the masseter muscle, which is termed as the "Turkey Wattle Sign." Doppler ultrasound imaging showed a relatively well-defined mixed reflective mass measuring 28×10 mm, with internal hypoechoic cystic and solid areas in the intramuscular compartment of the right masseter muscle. Magnetic Resonance Imaging (MRI) of the neck revealed a well-defined, non-enhancing, lobulated, heterogeneous lesion measuring about 2.1×1.4×2.4 cm in the right masseter muscle. Surgical excision was performed using a right submandibular approach under general anaesthesia. Histopathological examination of the specimen showed loose fibro-collagenous tissue and dilated vascular channels between skeletal muscle fibers, with a few calcifications suggestive of an intramuscular haemangioma. The turkey wattle sign, combined with appropriate radiographic interventions, can correctly diagnose intramuscular haemangiomas with calcifications.

Keywords: Diagnostic, Intramuscular haemangioma, Magnetic resonance imaging, Ultrasound

CASE REPORT

A 43-year-old male patient reported with a chief complaint of swelling on the lower right side of the face. The swelling had been present for six years, initially small in size but gradually increasing to its present size. The patient noted no history of difficulty with mastication, pain, trauma, fever, or discharge related to the swelling. His past medical and family histories were unremarkable, and he had no adverse habits.

Clinical examination revealed a diffuse swelling on the right lower third of the face, extending anteroposteriorly from the right corner of the mouth to the right tragus of the ear. Superoinferiorly, the lesion extended from the right ala tragus line to the inferior border of the mandible [Table/Fig-1]. On palpation, the swelling was non-tender,

soft, and less compressible with no pulsations felt. Further, the swelling appeared more prominent upon contraction of the masseter muscle during teeth clenching, demonstrating the "Turkey Wattle Sign" [Table/Fig-2].

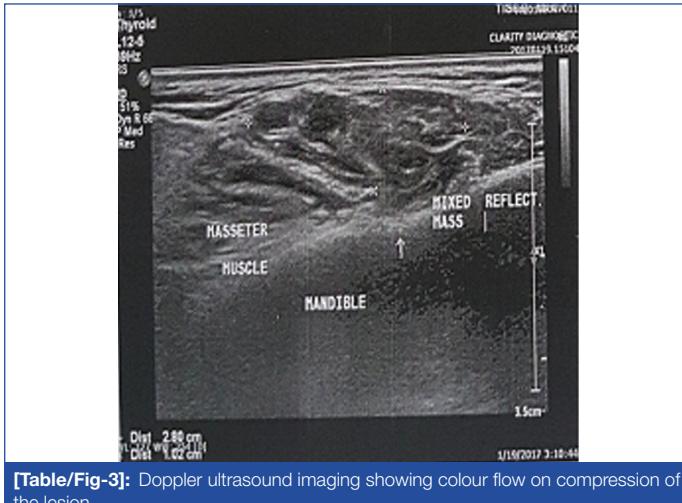


[Table/Fig-1]: A diffuse swelling on the right lower one third of the face.



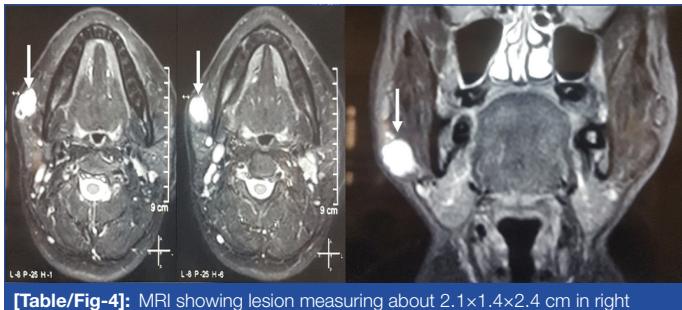
[Table/Fig-2]: Swelling accentuated on clenching teeth showing turkey wattle sign.

A provisional diagnosis of a vascular lesion in the right masseter was established. Differential diagnoses of parotid gland tumour and lymphangioma were also considered. Doppler ultrasound imaging revealed a relatively well-defined mixed reflective mass measuring 28×10 mm, with internal hypoechoic cystic and solid areas in the intramuscular compartment of the right masseter muscle, demonstrating color flow on compression [Table/Fig-3]. This suggested a differential diagnosis of intramuscular haemangioma or arteriovenous malformation.



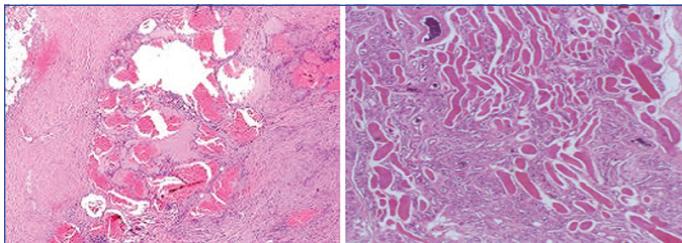
[Table/Fig-3]: Doppler ultrasound imaging showing colour flow on compression of the lesion.

The MRI of the neck revealed a well-defined, non-enhancing, lobulated, heterogeneous lesion measuring about 2.1×1.4×2.4 cm in the right masseter muscle, with two to three hypointense foci [Table/Fig-4] representing phleboliths, suggestive of an intramuscular haemangioma of the right masseter with calcifications.



[Table/Fig-4]: MRI showing lesion measuring about 2.1×1.4×2.4 cm in right masseter muscle with two to three hypointense foci.

Surgical excision was performed using the right submandibular approach under general anesthesia. Histopathological examination of the specimen showed loose fibro-collagenous tissue and skeletal muscle fibers with dilated vascular channels within the muscular layer, along with a few calcifications [Table/Fig-5]. These features were suggestive of intramuscular haemangioma. Furthermore, follow-up over seven years showed no recurrence [Table/Fig-6].



[Table/Fig-5]: Histopathological sections showed skeletal muscle fibres with dilated vascular channels within muscular layer and few calcifications (Haematoxylin and Eosin (H&E) staining, X20 magnification).

DISCUSSION

Haemangiomas are uncommon benign vascular tumours affecting skeletal muscle, occurring in less than 1% of cases in the head and neck region. The masseter is the most commonly involved muscle,



[Table/Fig-6]: Follow-up of 7 years showed no recurrence.

followed by the trapezius and sternocleidomastoid muscles. An intramuscular haemangioma of the head and neck region involving the masseter muscle is often mistaken for parotid swelling [1,2].

Most haemangiomas can be diagnosed through clinical examination; however, imaging is required in cases of deep haemangiomas with normal overlying skin or in cases of clinically apparent soft-tissue masses. Conventional radiographs can help identify phleboliths and calcifications, but they may not be specific. Ultrasonography and MRI are the commonly used modalities of choice. In intramuscular haemangiomas, Colour Doppler sonography can evaluate pathological changes like fibrosis and detect calcifications by visualising the vascular structures in and around the muscle [3]. Therefore, this case report emphasises the importance of investigations for accurate preoperative diagnosis, along with a literature review.

Intramuscular haemangiomas were initially described by Liston in 1843. According to the size of the vessel, Allen and Enzinger categorised them in 1972. Three types of intramuscular haemangiomas are recognised: capillary, cavernous, and venous [4]. Intramuscular haemangiomas are gradually enlarging lesions that lead to cosmetic problems, and some are associated with pain. They predominantly occur before the age of 30 years without gender predominance; however, they are more common in males [1].

In the present case, the swelling was quite prominent, especially with enlargement upon clenching the teeth, indicating the turkey wattle sign. An uncommon pathognomonic indicator of intramasseter and intraparotid haemangiomas is the turkey wattle sign. This sign describes the expansion of the lesion with dependent head positioning or teeth clenching. Vascular engorgement within the lesion may cause this sign, which prevents blood from returning from the head to the superior vena cava. The male turkey's neck contains a red vascular structure that can enlarge when filled with blood, hence the name "turkey wattle sign" [5].

Lesions of the masseter are often mistaken for parotid swelling. Differential diagnoses include salivary gland tumours, masseter muscle hypertrophy, lymphangioma, angiosarcoma, haemangiopericytoma, rhabdomyosarcoma, and myositis ossificans [6]. A tentative diagnosis of haemangioma was made in this case due to the deep-seated position and the rarity of signs indicating a vascular etiology.

Preoperative diagnostic accuracy is only 8%; therefore, Computed Tomography (CT), MRI, and ultrasound examinations are crucial. The CT scan plays an important role in identifying the size and shape of the tumour and the surrounding tissues. An ultrasonogram coupled with Colour Doppler can distinguish haemangioma from

vascular malformation and is often used to guide sclerotherapy. MRI is also essential for diagnosis, as it characterises the type of lesion, extent of the lesion, involvement of tissue layers, and flow pattern to guide treatment towards transarterial or percutaneous embolisation [7,8]. Phlebography is significant in identifying the venous drainage pattern, which can be cavitary, spongy, or dysmorphic. On average, phlebolith development occurs in 15% to 25% of intramuscular haemangiomas. Phleboliths are primarily composed of calcium phosphate and carbonate salts. It is thought that the pathophysiology of phleboliths involves thrombi formed by reduced peripheral blood flow, which subsequently organise and mineralise to create calcified thrombi [4].

In the present case, MRI scans revealed two to three hypointense foci within the lesion in the right masseter muscle, representing phleboliths, which were suggestive of intramuscular haemangioma of the right masseter with calcifications. Histologically, intramuscular

recurrence rate has ranged from 9% to 28%. Medical options, such as corticosteroids, interferon, and vincristine, have been effective for massive and life-threatening cases [1,7].

This paper also reviewed cases of masseter muscle haemangioma from 2009 to present. The English-language literature was surveyed on cases of haemangiomas in the masseter muscle, and all findings were tabulated in [Table/Fig-7] [1-4,6-17]. Seventeen cases have been reported in the masseter muscle, including the present case [1-4,6-17]. Among all the reported cases, ages ranged from seven to 66 years, with a predominance in females. In most cases, Doppler, MRI, CT, and angiography have been utilised to diagnose the lesion. Histopathological examination revealed capillary, cavernous, venous, or mixed vessel type haemangiomas. Three cases showed phleboliths, including this case [4,14]. In almost all reported cases, surgery was performed, and follow-up showed no recurrence.

S. No.	Author	Year	Age/Gender	Investigations	Histopathological diagnosis	Treatment
1.	Present case	2025	43 y/male	Doppler and MRI	Intramuscular haemangioma	Surgery with follow-up of 7 yrs showed no recurrence
2.	Shah R et al., [4]	2024	19 y/male	Ultrasonography Panoramic radiograph and Computed Tomography (CT) angiography	Intramuscular cavernous haemangioma with phleboliths	Surgery with 1 month follow-up - No recurrence
3.	Marion OLH et al., [10]	2023	30 y/female	Doppler ultrasonography MRI	-	Not done
4.	Komatsu M et al., [11]	2022	56 y/female	Contrast-enhanced Computed Tomography (CT) and Magnetic Resonance Imaging (MRI)	Intramuscular haemangioma	Surgery with 3 years of follow-up - No recurrence
5.	Park D et al., [12]	2021	66 y/female	Doppler ultrasonography	Mixed vessel type intramuscular haemangioma	Surgery
6.	Makkad RS et al., [13]	2021	25 y/male	Colour Doppler ultrasonography	Intramuscular capillary haemangioma	Surgery with follow-up of 6 months - No recurrence
7.	Kumar ES et al., [7]	2020	20 y/male	Magnetic Resonance Imaging (MRI) neck	Intramuscular haemangioma	Surgery with 6 months follow-up - No recurrence
8.	Shetty UA et al., [9]	2018	26 y/female	Magnetic Resonance Imaging (MRI)	Intramuscular mixed capillary and cavernous haemangioma (venous haemangioma)	Surgery with telephonic conversation - No recurrence
9.	Kim I et al., [8]	2017	48 y/male	Magnetic Resonance Imaging (MRI), External Carotid Angiography (ECA)	Intramuscular capillary haemangioma	Surgery with 8 months follow-up - No recurrence
10.	Elhariti L et al., [6]	2017	16 y/female	CT and MRI	Not mentioned	Surgery after arterial embolisation
11.	Kumar LKS et al., [1]	2016	35 y/male	MRI and angiogram	Intramuscular cavernous haemangioma with calcifications	Surgery
12.	Khaladkar SM et al., [2]	2016	7 y/female	Ultrasonography and MRI	-	Not done
13.	Alami B et al., [14]	2015	34 y/male	Maxillofacial Magnetic Resonance Imaging (MRI)	Phleboliths	Not done
14.	Lakshmi KC et al., [3]	2014	23 y/female	Colour Doppler ultrasound, MRI	Venous haemangioma	Surgery with follow-up of 3 years - No recurrence
15.	Jain V et al., [15]	2011	8 y/female	Colour Doppler sonography and MRI	Cavernous haemangioma	Surgery
16.	Kim HW et al., [16]	2010	56 y/male	Magnetic Resonance Imaging (MRI)	Intramuscular haemangioma	Surgery
17.	Narayanan CD et al., [17]	2009	17 y/female	Contrast CT	Capillary haemangioma	Surgery

[Table/Fig-7]: Review of literature showing 17 cases of masseter muscle haemangioma including present case [1-4,6-17].

haemangiomas are characterised by large and small proliferating vessels embedded within muscle tissue in the deep layer, exhibiting somewhat different characteristics from other types of haemangiomas [9].

There are two hypotheses for intramuscular haemangiomas: the first is hereditary, while the second is trauma-related. Intramuscular haemangiomas predominantly occur in childhood, though they are often not discovered until a sudden enlargement, discomfort, or cosmetic asymmetry develops in adulthood. Trigger factors include hormonal changes, infection, or trauma [8].

Medical and surgical options are available for the treatment of intramuscular haemangiomas. The proximity to the facial nerve and the postoperative flattening after masseter muscle removal make the intra-masseteric position particularly problematic. The

CONCLUSION(S)

The turkey wattle sign, along with radiographic interventions, can correctly diagnose intramuscular haemangioma. Although intramuscular haemangioma of the masseter muscle is a very rare occurrence, it should still be included in the differential diagnosis of tumours in the parotid region.

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