Intramuscular Hemangioma of Masseter Muscle: Case Report of Rare Clinical Entity

Ramanpal Singh Makkad, Gunjan Agarwal[1], Santosh Gupta[2], Ravleen Nagi[3], Akshay Ragit[4], Farhin Jamal[4]

Departments of Oral Medicine and Radiology and [4]Oral and Maxillofacial Surgery, New Horizon Dental College and Research Institute, Bilaspur, [1]Department of Surgical Oncology, JNM Medical College, [2]Department of Pathology, Suyash Hospital, Raipur, Chhattisgarh, [3]Department of Oral Medicine and Radiology, Saveetha Dental College, Chennai, Tamil Nadu, India

Abstract

Rationale: Intramuscular hemangiomas are unique benign vascular tumours of skeletal muscles; involving masseter and trapezius muscles in the majority of cases. The rationale was to emphasize that the diagnosis of asymptomatic swelling in the masseteric region is important as due to their deep anatomic location and unfamiliar presentation, they are often misdiagnosed as a parotid swelling or other muscular pathologies. **Patient Concern:** This report describes a rare case of a 25-year-old healthy male patient who presented with an asymptomatic swelling in the right masseteric region. The patient had cosmetic concerns due to the large size. **Diagnosis:** Colour Doppler ultrasonography was done to assess the vascularity within the lesion. **Treatment:** Complete excision was successfully achieved using combined Risdon's and preauricular approach. **Outcome:** No signs of recurrence were observed after 6 months **Take-away Lessons:** Appropriate selection of diagnostic modalities enables the clinician in making an accurate preoperative diagnosis of progressive swelling in the masseteric region.

Keywords: Diagnosis, hemangioma, muscle, surgical excision, ultrasonography

Introduction

Intramuscular hemangiomas (IMHs) are benign vascular tumours that most frequently occur in the trunk and extremities.[1] They constitute about <1% of all hemangiomas, only 10%–20% of cases have been found to arise in the head-and-neck (H and N) region, and 36% of cases involving masseter muscle.[2,3] IMHs usually present as gradually enlarging soft-tissue mass with or without pain. They are located deep within the muscle, hence seldom exhibit clinical signs or symptoms such as bruits, thrills, pulsation which are suggestive of vascular origin. Due to their rare occurrence, unique location, and diverse clinical presentation they are often confused with other soft-tissue pathologies such as parotid swelling or other muscular lesions.[4-6] We present a rare case of a 25-year-old healthy male patient who presented with an asymptomatic swelling on the right masseteric region diagnosed with IMH by imaging, i.e., colour Doppler ultrasonography (USG) and by histopathological examination.

Case Report

A 25-year-old male patient reported to the Department of Oral Medicine and Radiology with a complaint of incidental finding of swelling with gradual growth on the right lower one-third of the face for 1 year [Figure 1]. The patient denies the previous history of facial trauma. Medical, social, and family history were nonsignificant. General physical examination revealed no remarkable findings. On extraoral examination, well-defined oval-shaped solitary swelling was observed extending from the right body of the mandible to the angle of the mandible anteroposteriorly and from the zygomatic process to the inferior border of mandible supero-inferiorly measuring about 6 cm × 5 cm approx. in its greatest dimensions. The colour of the swelling was normal skin colour with smooth surface and no secondary changes were observed. Surrounding structures appeared to be normal. On palpation, the swelling

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Address for correspondence: Dr. Ramanpal Singh Makkad, Department of Oral Medicine and Radiology, New Horizon Dental College and Research Institute, Bilaspur, Chhattisgarh, India. E-mail: drramanpal@gmail.com

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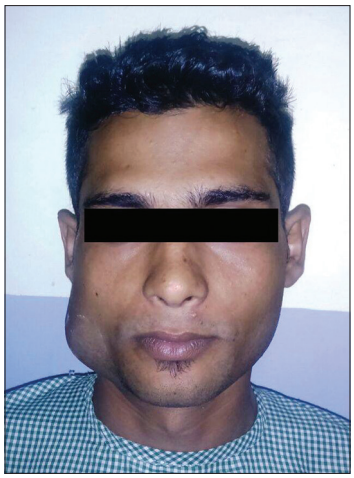

Figure 1: Extraoral photograph showing diffuse solitary swelling on right masseteric region

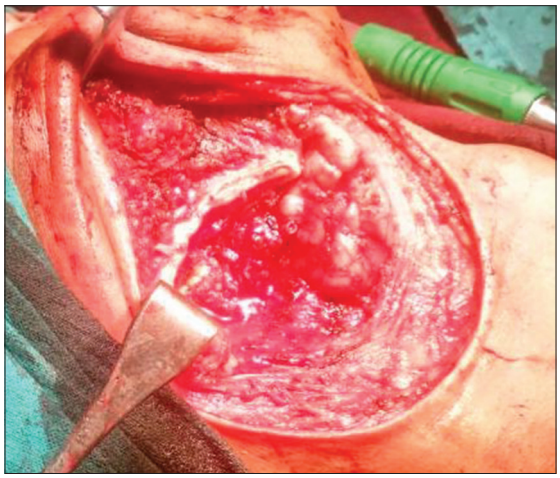

Figure 3: Intraoperative view showing hemangioma along with masseter muscle on the right side

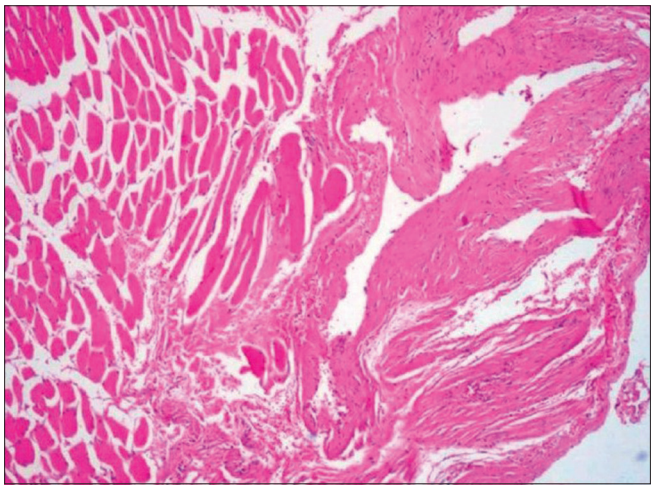

Figure 5: Microphotograph showing abundant of vascular spaces lined by endothelial cells interspersed with skeletal muscle bundles. (H and E stain × 40)

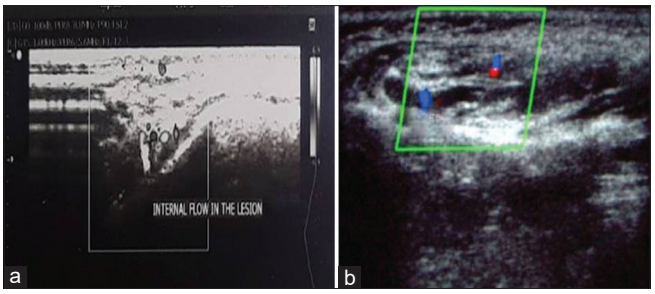

Figure 2: (a and b) Colour Doppler ultrasonography showing enhanced vascularity within the lesion

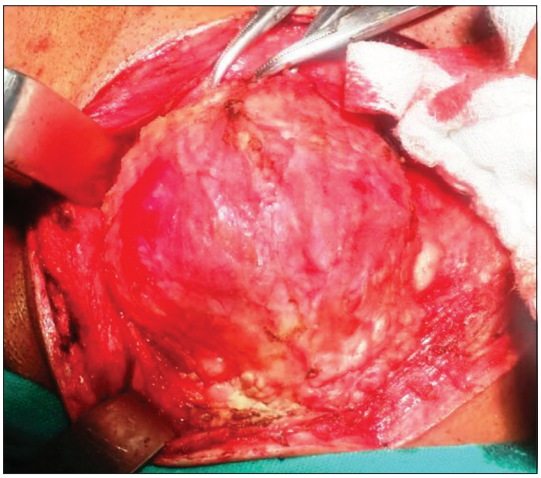

Figure 4: Intraoperative view after excision of hemangioma by preservation of facial nerve

was nontender, afebrile, firm, noncompressible, nonfluctuant, nonpulsatile [Figure 1]. On intraoral examination, no abnormality was seen with the hard and soft tissues related to the extraoral swelling. Parotid duct orifice was noted to be patent and no signs of xerostomia were evident. Based on history and clinical examination, provisional diagnosis of right masseteric hypertrophy was made and differential diagnosis included fibroma, lipoma, and parotid tumour.

Patients concern

Patient had cosmetic concern due to large size swelling with gradual growth on right masseteric region for 1 year.

Diagnostic aids

Routine hematological investigations revealed all the parameters within normal ranges. USG of the right masseteric region revealed lobulated, hypoechoic mass in the right masseter muscle. In Colour Doppler USG, vascularity was visible within the mass [Figure 2a and b]. On the basis of above radiological investigations, final diagnosis of intramuscular hemangioma of right masseter muscle was made.

Treatment

After obtaining patients' consent, IMH was excised by combined Risdon's and preauricular approaches. For the surgical procedure, incision below and behind the angle of

the mandible and preauricular skin incision were made on the right side and skin flaps were raised A parotid gland appeared normal and the underlying masseter showed a diffuse bulge but with no surface abnormality. The branches of the facial nerve were identified and a superficial parotidectomy was performed. The nerve fibers were subsequently dissected from its masseteric bed. The underneath masseter muscle exhibited prominent bulge with hemangioma, the facial nerve branches were gently lifted and then the masseter was mobilized from the mandible after severing its attachment with the zygomatic arch. Through the same incision, the external carotid artery was ligated and vascular control was achieved. Anteriorly, the masseteric fascia was preserved along with the fine communications of the facial nerve. Thereafter, complete excision of the masseter muscle along with hemangioma was successfully achieved [Figures 3 and 4].

Specimen excised was sent for histopathological examination which revealed connective tissue stroma with numerous skeletal muscle bundles. Large dilated vascular spaces lined by endothelial cells, interspersed between the skeletal muscle bands were observed which is [Figure 5] suggestive of intramuscular capillary hemangioma.

Outcome and follow-up

No recurrence was observed after 6 months of follow-up.

Discussion

IMHs are benign hamartomatous vascular tumours involving masseter muscle most commonly, are deeply seated, therefore the characteristic features such as bluish translucent hue, pulsations, bruits, thrills, and the sign of emptying are seldom seen and due to their location and proximity with adjacent anatomical structures, they are often misdiagnosed as muscle hypertrophy, muscle neoplasms, parotid tumours, fibroma, deep-seated lipoma, and congenital cyst.[7-11] Imaging plays a key role in achieving a proper diagnosis of IMH, conventional radiography has a limited role and only helps in detecting calcifications in hemangiomas.[8,9] Colour Doppler USG is the most convenient and economical imaging modality and precisely detects the feeder vessels which further helps the surgeon to treat the patient more appropriately and conservatively.[8-12] In this case, colour Doppler USG revealed enhanced vascularity within the muscle tissue suggestive of IMH of the masseter muscle.

Allen and Enzinger classified hemangiomas as large vessel (>140 mm in diameter), small vessel (<140 mm in diameter) and mixed vessel types, clinically corresponding to cavernous, capillary, and mixed types, respectively.[1,2] IMH develops feeder vessels deep in the muscle tissue which makes it different from the other types of hemangiomas. On reviewing the pertinent literature, limited cases of IMH have been documented, Alami *et al*.[1] observed space-occupying lesion in the left masseter region, hyperintense signal on T2-weighted magnetic resonance imaging (MRI) images containing nodular hypointense foci. In another case, Surej *et al*.[12] observed well-enhanced circumscribed intramuscular mass on MRI images, and findings were suggestive of vascular lesion.

The selection of treatment depends on the factors such as tumour morphology, anatomy, growth rate, depth of invasion, age of the patient, and cosmetic and functional consideration.[13] Various treatment modalities such as cryotherapy, intralesional sclerosing agents, electrocoagulation, and radiation therapy, have shown optimal success rates with minimal recurrence but surgical excision with wide margins remains the treatment of choice.[14-16] The recurrence rate of 9%–28% has been reported after wide surgical excision, therefore, to reduce relapse complete excision of the muscle is recommended.[16] In our case, this was more of a cosmetic concern and depending on the size and anatomy of the lesion, the mass was successfully excised.

The present case was a diagnostic challenge due to its asymptomatic gradually progressive nature of the swelling, which was soft but nonpulsatile on physical examination. To exclude other possible pathologies such as masseteric hypertrophy and benign parotid tumour that may arise in the masseteric region, we confirmed our presumptive differential diagnosis by colour Doppler USG that suggested intramuscular mass to be of vascular origin. Although MRI has been proven to have high diagnostic efficiency in accurate detection of neurovascular structures, colour Doppler USG was advised to our patient due to its cost-effectiveness in comparison to MRI.[1,12]

Conclusion

Adequate knowledge of likely differential diagnosis of IMH with the selection of appropriate diagnostic modalities enables the clinician in making an accurate preoperative diagnosis of this rare entity.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published and due efforts will be made to conceal their identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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