

Intramuscular Hemangioma of Orbicularis Oris

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Abstract

Case Report

Intramuscular hemangiomas are the infrequent benign congenital neoplasms which account for less than 1% of all hemangiomas and less than 20% of these are found in cervicofacial region. Masseter muscle is the often involved muscle in head and neck region accounting for 5% of all hemangiomas. Most commonly present in first three decades of life and unlike cutaneous hemangiomas, they show no signs of regression and are deep seated within the muscles which are slow growing [1]. Intramuscular hemangiomas scarcely display any clinical signs and symptoms that reveal their vascular nature. So these are most commonly misdiagnosed entity [2]. We report a rare case of intramuscular hemangioma of orbicularis oris in a 32 year old male.

Keywords: intramuscular hemangioma, hemangioma, orbicularis oris.

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CASE REPORT

A 32-year old male patient reported to Department of Maxillofacial Surgery with the complaint of swelling in the lower lip and chin region of 10 years duration. The medical history was insignificant but he gave a history of attempted surgical excision 7 yrs back and due to profuse bleeding the procedure was abandoned. Patient also reported an incident of blunt trauma to the chin region during his

childhood. Physical examination was unremarkable and blood investigations were within normal limits.

Examination revealed a non-tender swelling measuring 3x3 cm in the lower lip extending from one commissure to other which was mildly pulsatile (Figure.1). Overlying skin had 'peau de orange' like appearance and there was no paraesthesia of involve region. Intra oral examination revealed an exophytic proliferative gingival growth in relation to the central incisors which were mobile.



Fig-1: Swelling measuring about 3x3cm with peau de orange like appearance.



Fig-2: Contrast enhanced CT angiography with 3D-reconstruction

Orthopantomogram showed no relevant findings. Fine needle aspiration obtained blood. 3D colour Doppler ultrasound showed well-defined hypochoic soft tissue mass with feeders. Contrast enhanced CT angiography with 3D - reconstruction showed tangled masses of blood vessels which were fed by bilateral facial arteries (Figure.2). Considering it to be a vascular lesion a differential diagnosis of hemangioma, hematoma and low flow venous malformation was made.

The patient was treated with surgical excision of the mass with ligation of feeder vessels under

hypotensive anesthesia. Submandibular incisions were placed bilaterally with a submental extension. Skin, subcutaneous tissue, superficial fascia, platysma and deep fascia are reflected and facial arteries were identified and ligated bilaterally. Swelling was approached both intraorally and extraorally. On the contrary to the expectation, we did not encounter any blood filled tissue except, a mass of connective tissue which was excised with a margin of normal muscle (Figure.3). The wound was closed in layers and post-operative course was uneventful.



Fig-3: excision of lesion along with normal muscle

Histopathological examination revealed diffuse distribution of capillaries lined by flat endothelial lining infiltrating deep into muscle layers, sometimes organized into a branching network, surrounded by perivascular myxoid tissue with chronic inflammatory changes. These features were in consistent with infiltrating hemangioma into surrounding fat & muscle tissue suggestive of intramuscular hemangioma.

DISCUSSION

Intramuscular hemangioma is an uncommon vascular malformation accounting for less than 1% of all hemangiomas. It affects most commonly in trunk and extremities where muscle volume is larger. Approximately 13% of these lesions present in head and neck region. In the head and neck region masseter is the most frequently involved muscle [3]. Rarer sites

include orbicularis oris, depressor angular oris and orbicularis oculi [4]. Our case is the intramuscular hemangioma involving orbicularis oris in lower lip.

Intramuscular hemangioma presents as progressively enlarging, painful swelling and are in deep location. Due to their fibro-muscular nature intramuscular hemangiomas have a rubbery, firm texture and they scarcely display any clinical signs of vascular lesions like pulsations, bruit and thrill [5]. The absence of characteristic pathognomic findings and rare incidence of these lesions in head and neck region make accurate preoperative diagnosis highly impossible. A peculiar finding that can be observed in intramuscular hemangiomas is the presence of Wattle sign i.e., an enlargement of clinical vascular swelling when the head is in dependant position [4] vascular return from the head to superior vena cava is impeded by gravity. Vascular engorgement is also observed in Valsalva

maneuver [6]. This unusual pathognomic sign is also called as “turkey wattle sign“, termed for the red vascular structure in the neck of male turkey, which has ability to become engorged with blood and to swell [4]. Imaging modalities that are helpful in diagnosing the intramuscular hemangiomas are ultrasound, MRI, CT angiography and digital subtraction angiography. Ultrasound is useful in differentiating a low flow from a high flow tumor [7]. An angiogram may define the vascular extent tumor. Occasionally these tumors are suitable for pre – operative arterial embolisation under radiologic control which may reduce intra operative bleeding.

In our case we have opted 3D reconstructed CT – angiogram to identify the vascular mass in the lower lip (orbicularis oris) and the vessels feeding were identified as bilateral facial arteries. Various treatment options for management of intramuscular hemangiomas are steroid therapy injections, radiation, and injection of sclerosing agents. However the optimal management is surgical resection with wide margins of surrounding normal muscle because of infiltrative nature of intramuscular hemangiomas [8]. Intramuscular hemangiomas have tendency to recur even after complete excision. In our case after 18 months follow up the patient was well with no evidence of recurrence [9].

CONCLUSION

Intramuscular haemangiomas are unusual, benign tumours in the head and neck region. When evaluating intramuscular lesions, a wide range of differential diagnoses including both, benign and malignant lesions, must be considered. The preoperative diagnosis is often difficult in these cases due to their variable size, consistency, deep location and unfamiliar clinical and radiological presentation. In order to avoid misdiagnosis and to ensure appropriate management, it is necessary that the surgeon who deals with these tumours is aware of their distinctive histopathology and biological behaviour.

Optimal management requires good clinical judgement, surgical planning and complete excision together with a cuff of normal tissue. Postoperative cosmetic and functional disability after excision of these lesions has been minimal, even with significant removal of surrounding muscle. Incomplete excision or other forms of treatment are associated with an unacceptably high rate of local recurrence and are to be discouraged.

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