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RESEARCH ARTICLE

AN ABERRANT CASE OF INTRAMASSETRIC HEMANGIOMA: A CASE REPORT AND LITERATURE REVIEW

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Abstract

Introduction: Intramassetric hemangioma (IMH), a rare variant of intramuscular hemangioma, is benign tumor of vascular origin affecting the masseter muscle and accounts for less than 1% of all hemangiomas. When occurring in the head and neck, the most common site of occurrence is the masseter muscle followed by the trapezius and sternocleidomastoid muscle. Most often it produces a diffuse swelling at the cheek that becomes prominent on clenching the teeth, which can be often mistaken for masseteric hypertrophy. CT scanning of most hemangiomas show the presence of calcified thrombi called phleboliths which are also seen in the masseterian variety.

Case Report: A 19-year-old female presented with the complaint of swelling in the left cheek region since 1 year which was slowing increasing in size over time. CT showed heterogeneously enhanced mass lesion of the left masseter with the presence of phleboliths. The mass was excised under general anaesthesia and verified to be intramassetric hemangioma of the left masseter muscle. Conclusion: intramassetric hemangiomas are a rare entity which can be easily mistaken for a more commonly occurring massetric hypertrophy. Careful radiographic and histopathological examination will help to attain a definite diagnosis.

Introduction:-

Intramassetric hemangioma is a form of hemangioma involving the skeletal muscle and comprising 0.8% of all hemangioma [1]. Hemangiomas affects mainly the trunk and extremities where the muscle volume is larger but when occurring in the head and neck, it arises most commonly from the masseter muscle [1,2,3]. Other locations include trapezius, periorbital muscle, sternocleidomastoid, temporalis, orbicularis oris, geniohyoid, pterygoid, mentalis, buccinator etc [1,2,4].

Although intramuscular hemangioma has a slight female predominance, the greater part of masseterian hemangiomas are diagnosed in men [4,5,6]. In 90% of cases the diagnosis is made before the age of 30 years [5]. The trigger factors known to cause intramassetric hemangioma include hormonal change, infection or trauma [1,4,5]. Often misdiagnosed (over 90%) due to rarity and unfamiliar presentation, intramassetric hemangioma can be mistaken for lymphoma, lymphangioma, parotid tumour, cysts, masseter hypertrophy among others [1].

Two interesting findings regarding intramassetric hemangioma are Wattle's sign and phleboliths. The Wattle's sign is the unusual pathognomonic manifestation of intramassetric hemangioma and intra parotid hemangioma where

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there is enlargement of the lesion with teeth clenching/with dependent head positioning [3,6]. Although rare in the oral region, calcified thrombi or phlebolith when present can suggest an IMH (3-50%), which is easily diagnosed with CT [3].

Treatment approaches to such a condition can extend from complete surgical extirpation, non-surgical modalities to periodic observation, based on rate of growth, size, cosmetics, pain, muscle compression etc [1,2,7]. The present article reports a case of Intramassetric hemangioma in a 19-year old female and a brief review of the literature

Case Report:
A 19-year-old female reported to the Department of Oral and Maxillofacial surgery with swelling of left side of face, which she stated as having been present since 1 year. The swelling was said to be of acute onset, gradual asymptomatic progression without any history of trauma or any aggravating or relieving factors. The patient was otherwise in good health. Extra oral examination revealed a solitary swelling of 4*4cm that was firm, non-tender, non-fluctuant and non-fixed on the left mandibular angle region. The skin above the swelling was normal, without any rise in temperature. The swelling became conspicuous while clenching the teeth. No palpable cervical lymphadenopathy was evident. Intraoral examination revealed no abnormalities.

Complete haematological work up was undertaken, followed by CECT of the head and neck. CT showed heterogeneously enhanced mass lesion of the left masseter of 30*18mm with two calcified foci, the larger of which was as big as 6mm. A few non-enhancing areas were evident along with necrosis, without extension into the neighbouring mass. This gave the impression of intramassetric hemangioma with phleboliths for which excisional biopsy under general anaesthesia was planned.

Under standard aseptic protocol, a complete excision of the lesion was done under general anaesthesia. Risdon's incision was made (figure 1) following which the masseter muscle and the vascular lesion was exposed by blunt dissection (figure 2). The bleeder vessel was identified and ligated (figure 3) followed by the complete excision of the vascular lesion (figure 4). Once haemostasis was achieved, wound closure was done in layers.

Histopathological analysis of the specimen revealed muscle fibre intermingled with adipose tissue. The muscle and adjacent connective tissues showed numerous blood vessels of varying sizes (figure 5), thus giving a diagnosis of intramassetric hemangioma.

Post-surgical healing was uneventful without any functional or aesthetic impairment (figure 6). At 1 year after the surgery, the patient was well and without evidence of recurrence.

Discussion:-

Comprising only of 0.8% of all hemangiomas, intramuscular hemangioma is a relatively rare condition affecting the skeletal muscles of more commonly the trunk and extremities where the muscle volume is larger [1,3].

When occurring in the head and neck region (10%-15%), it can affect the masseter, sternocleidomastoid, trapezius, temporalis, mylohyoid, mentalis, buccinator, lip, tongue, geniohyoid and medial pterygoid [4,6]. Approximately 36% of head and intramuscular hemangiomas occur in the masseter [7], followed by trapezius and sternocleidomastoid [4]. Most often they present before the age of 30 [4], although cases of elderly with intramassetric hemangioma have been reported. Intramuscular hemangiomas are known to have slight female predilection, nonetheless intramassetric variants have more predilection for the male gender [4].

Intramassetric hemangioma in the masseter presents as an indistinct slowly enlarging relatively firm, mobile mass with variable size [2,5,7]. Pain can be a variable symptom depending on the rate of growth, adjacent vital structures and thrombosis [7]. Being a deep seated lesion, intramassetric hemangioma rarely cause any change in colour of skin or mucosa nor does it manifests pulsation or bruits, due to the sluggish blood flow [7]. There have been reports of abrupt onset facial palsy resulting from an enlarged lesion pressing on the facial nerve [2].

An unusual pathognomonic manifestation of intramassetric and intra-parotid hemangiomas is the presence of wattle sign [3]. The turkey wattle is a red vascular structure at the neck of the male turkey that can increase in size when its engorged with blood [3,7]. Similarly, the lesion enlarges in size while clenching or during dependent head

positioning [3]. The phenomenon is due to vascular engorgement within the lesion that resists venous return from the head to the superior vena cava [3].

Another striking feature about intramassetric hemangioma is the presence of phleboliths. They are seen in about 3-50% of cases of IMH. Phleboliths are calcified thrombi usually seen in vascular abnormalities [7]. The tortuous vascular channels in the intramassetric hemangioma cause stasis that produce thrombi, that eventually undergoes calcification resulting in phlebolith formation [7]. Phlebolith may be in close proximity to the parotid gland and duct, confusing it for a sialolith. Sialolith are usually single, ovoid structures that are contoured by the wall of the Stensen's duct with concentric ring pattern while phleboliths may be smaller multiple without any signs or symptoms [7].

There are no known racial factors for occurrence of intramassetric hemangioma [6]. Muscle contraction and trauma seem to be of an important aetiology; hormonal factors also play a role with clear increase in volume in relation to menarche, pregnancy and menstrual cycles [5].

Due to the rarity of the lesion along with their deep location and unfamiliar presentation, over 90% of all intramassetric hemangioma are misdiagnosed. A variety of other conditions can be clinically confused with intramassetric hemangioma including masseteric hypertrophy, parotid tumours, lymphangioma, lymphoma, rhabdomyosarcoma, schwannoma and cysts [1, 4].

Allen and Enzinger classified them histologically as capillary, cavernous and mixed types. Capillary variety have vessels smaller than 140 micrometre in diameter and while there are larger than 140 micrometre in diameter in the cavernous types. The mixed type of hemangioma has both small and large vessels. The capillary varieties are known to be highly cellular, making them firm and lacking the clinical signs to suggest a lesion of vascular origin. They present with a shorter history unlike the cavernous variety. The later generally present with a longer history and tend to be larger and painful, with an occurrence of only 19% in the head and neck [4].

Diagnosis of an intramassetric hemangioma is best accomplished when the clinical picture of a neoplastic-like swelling is factored in with the result of imaging particularly with CT and MRI [8]. Although CT is an excellent imaging approach for diagnosis of phleboliths, it does not allow for good tissue differentiation between the lesion and surrounding tissues [2,4,7]. Without the phleboliths the CT scan may not be diagnostic even if contrast shows an enhanced mass lesion in view of its vascularity [8]. MRI represents the best imaging modality for the diagnosis and is considered the gold standard [1,7]. A T2 weighted shows intramassetric hemangioma to have an intense signal when compared to the surrounding structure, as there is abundant free water in the stagnant blood in the larger vessels, which is seen particularly in the proliferative phase of the lesion with more homogeneity [1,2,7]. During the involution phase of the lesion, a T2- weighted image will be more heterogeneous owing to areas of fat replacement [1]. Unlike arterio-venous malformations, angiography do not have very high value in the diagnosis of a intramassetric hemangioma although it might be of use for detecting a larger feeding artery [1,7]. Fine needle aspiration is not recommended as it poses a threat of haemorrhage [1].

According to the size, extend, location, rate of growth, accessibility of the lesion, age of the patient and the condition of the surrounding structures, the treatment should be planned and individualised that provides the best prognosis [2,4,7]. Treatment is indicated if there is an increase in growth, loss of function of the adjacent anatomical structures, pain, cosmetic concern or necrosis of overlying tissues [7]. Medical and surgical modes of treatment have been tried and tested over the years ranging from cryotherapy, sclerosing agents, radiation therapy, steroid administration, lasers, ligation of feeding vessel, embolization, carbon dioxide snow to surgical excision [2,4,7]. As the lesion is vascular, without a definite capsule and poorly circumscribed there are high chances of incomplete surgical excision of the lesion and therefore the complete excision of the masseter muscle is justified [2,4,7]. Preoperative arterial embolization and injection of sclerosing agent presurgery is advised before the surgical excision of a larger lesion to prevent the chances of severe haemorrhage, along with the ligation of the feeding blood vessel or the ligation of masseteric branch of the facial artery during the surgery [2]. The lesion can be accessed intraorally if it is located anterior in the masseter muscle and close to the oral mucosa via mucosal incision anterior to the Stensen's duct thus avoiding a visible scar and possible facial nerve injury. If not preauricular incision followed by superficial parotidectomy will provide a good exposure but the same poses a threat of facial nerve damage after facial nerve dissection [2]. Minor feeding vessels and residual tumour can be a cause of recurrence which occurs in 18% of cases, with 7% recurring more than once [1,2]. Regional and distant metastasis or

spontaneous regression has not been reported. Periodic observation and follow up is mandatory to diagnose recurrence, if any, relatively early [4].

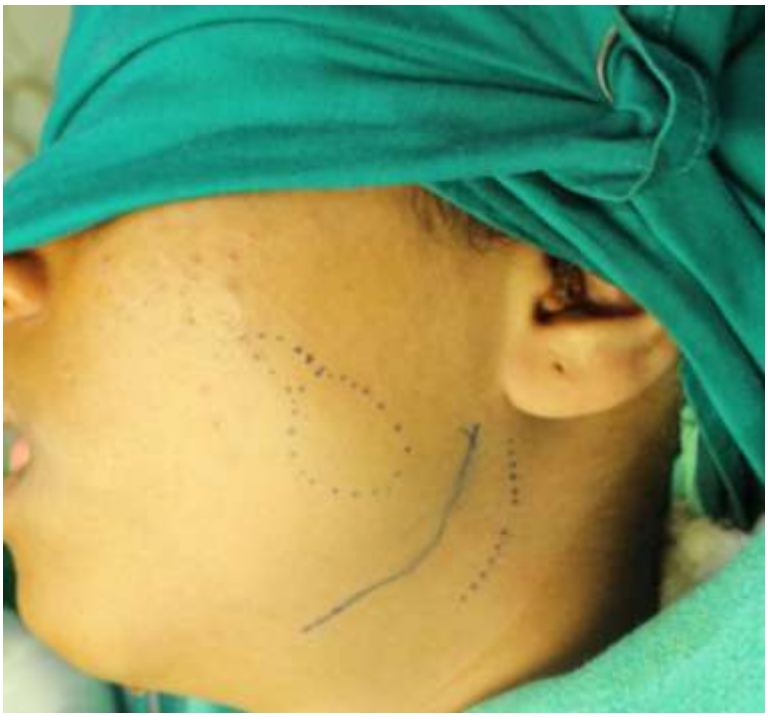

Figure 1:- Skin marking before placing incision.

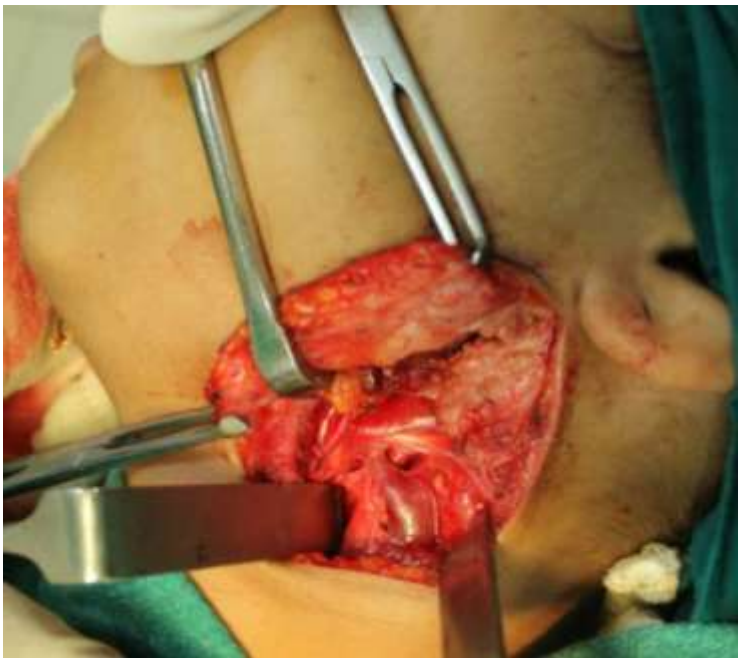

Figure 2:- Dissection to expose the lesion.

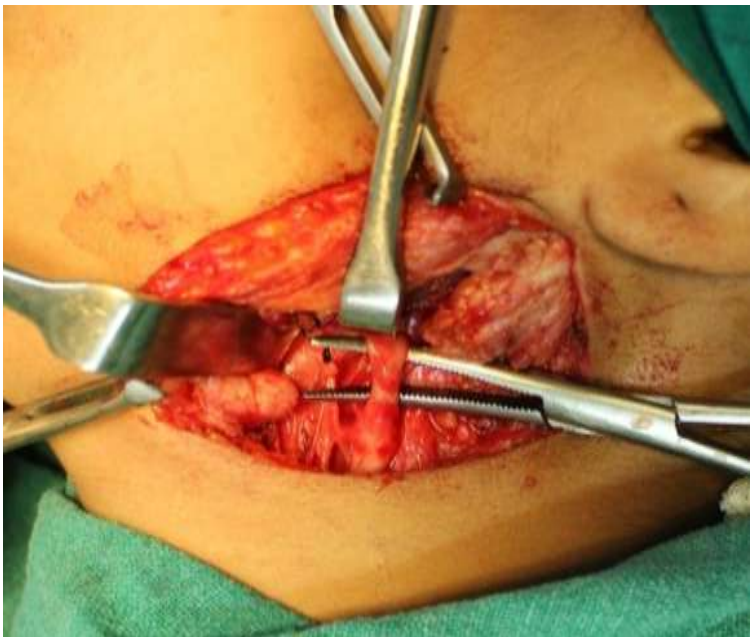

Figure 3:- Feeder vessel ligation.

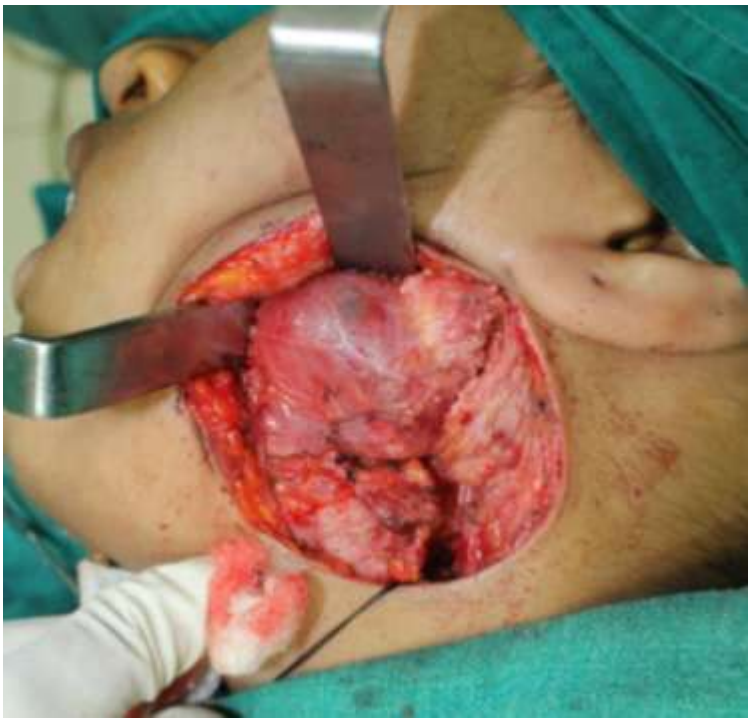

Figure 4:- exposed lesion.

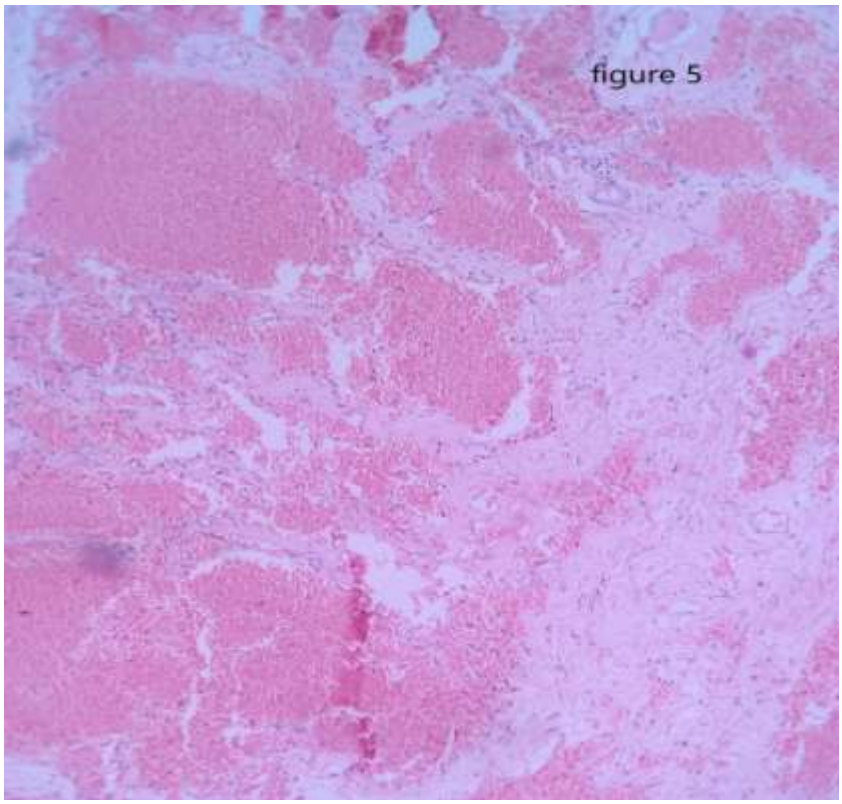

Figure 5:- Histopathology.

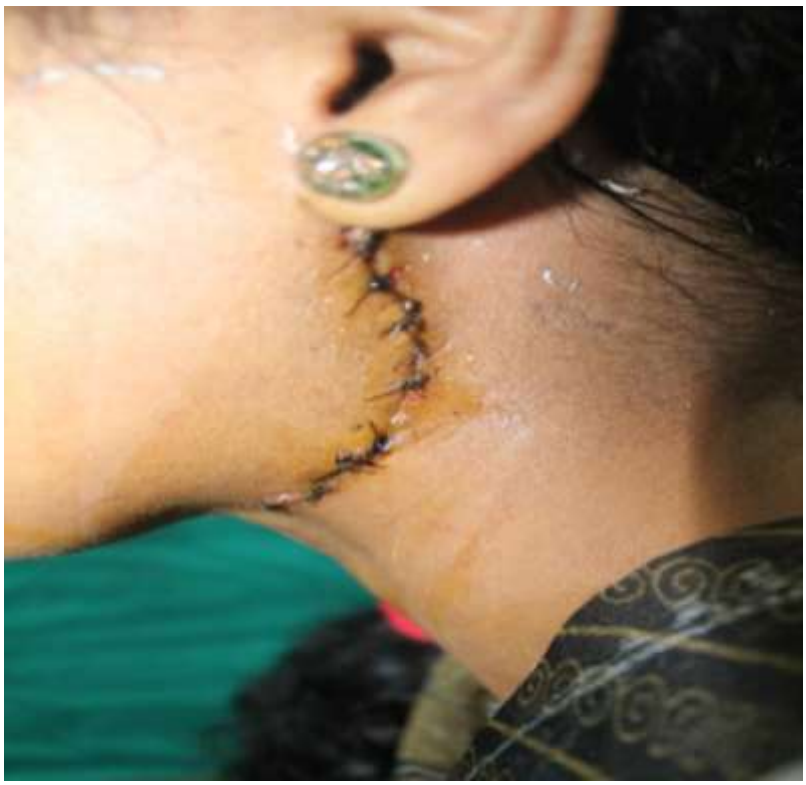

Figure 6:- Post-operative picture.

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