

## Bilateral Hemangioma of Temporo-Mandibular Joint

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### Abstract

Hemangiomas are proliferative lesions characterized by increased endothelial cell turnover, may be present anywhere in the body, and rarely involve the skeleton. It is extremely uncommon to come across a case of Hemangioma of the temporomandibular joint (TMJ) and even more rare to find a case of bilateral Hemangioma of TMJ. As a result of the low incidence of such anomalies in the region of the TMJ, most literature descriptions of vascular lesions in this area are case reports. Clinical Diagnosis of hemangioma consists of a clinical, radiological and possibly an angiographic examination. Magnetic Resonance Imaging (MRI) allows clear visualization of joint involvement and is particularly useful to define the extension of the hemangioma and its relationship with surrounding soft tissues. We believe that MRI should be the modality of choice to diagnose hemangioma in the oro-facial region and recommend that Dental surgeons and oral medicine experts should be aware of clinical implications of such an entity during their clinical interactions.

**Keywords:** Hemangioma; Proliferative lesions; Condylar head; Temporomandibular

### Introduction

Hemangioma was first described in the literature by Liston as early as 1843 [1a]. Starting in 1982, Mulliken and Glowacki's classification [1] has been used to separate vascular anomalies into hemangiomas and vascular malformations based on clinical manifestations, natural history, and histologic findings. While majority of the vascular malformations in the orofacial area are found in the mandible, especially the ramus and body [2-6], it is a rare entity in the temporomandibular joint. The peak incidence of hemangioma is in the second and fifth decades of life [7]. According to some authors, hemangiomas are more common in females than males (2:1), and more in whites than other racial groups [8]. There is a significant preponderance of hemorrhage and exsanguination accompanying biopsy or dental extraction associated with a diagnosis of Hemangioma which has been well documented by several authors [9,10]. The clinical presentation is usually a slow growing non-tender mass causing facial deformity or difficulty in mastication [11].

### Case Report

A 36 years old man presented to the Department of Dental Surgery, Dr. RML Hospital with difficulties in chewing and obvious facial deformity. The swelling had gradually increased to the present size. The swelling did not show any change in size during meals, bending or straining. There was no history of sudden increase in the size of swelling. The patient had no numbness or any abnormal sensation. Clinical examination revealed as a firm, nontender swelling of approximately 2 x 2 cm in size with ill-defined margins bilaterally. It was immobile from side to side and the overlying skin was normal in color, texture and temperature similar to surrounding skin. FNAB (Fine Needle Aspiration Biopsy) was done which yielded only blood. Even though a history, clinical examination and CT scan, potentiated the

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impression of benign condition, an MRI was ordered and excision of the mass was planned. T2 hyperintense areas can be seen extending in both TMJ cavities then medially in adjacent soft tissues. Contrast study was not done. With careful dissection, the mass revealed to be attached to the capsule and the TMJ disc and encroaching on the condylar head making obvious discolored condylar head. Histopathological examination of the specimen revealed lobules of variably sized large congested blood filled cavernous spaces with interstitial hemorrhages. The final histopathological diagnosis was cavernous hemangioma.

### Discussion

There are certain distinguishing features [1,12-15] which restrict the term hemangioma to the common vascular tumor of infancy, which grows rapidly during infancy and involutes by adolescence. Hemangiomas are proliferative lesions characterized by increased endothelial cell turnover, may be present anywhere in the body, and rarely involve the skeleton [16]. Most of the reported central hemangioma cases were located within the bone sometimes presenting hard and non-tender swellings but generally without symptoms [17]. Unni, *et al.* [18] reported that while hemangiomas were common, they are extremely rare in bone. It is imperative for a dental surgeon to recognize the importance of recognizing hemangiomas of the tooth-bearing jaws as they not only mimic other tumors of the region but may present with a life-threatening hemorrhage if a tooth is extracted or a trans-oral biopsy is attempted and this has already been stressed by authors of previous studies [19]. However, there is a serious lack of available literature which discusses in detail about hemangioma involving temporomandibular joint. Diagnosis consists of a clinical, radiological and possibly an angiographic examination. Based on microscopic appearance, hemangiomas are classified into two general patterns. The cavernous form is composed of large, thin-walled vessels and sinuses which are lined by a single layer of flat endothelial cells [20]. The capillary form is more cellular and composed of fine densely grouped capillary loops [18]. In lesions with high vascular pressure patients have had a sensation of pulsation [21,22] and consequently some lesions extending into the soft tissue have audible bruits [23]. However, intrabony lesions make auscultation of a bruit unlikely. Occasionally patients have had paresthesia in the region of the lesions [21,24,25]. Most of the lesions in the present study exhibited diffuse borders. The literature is inconsistent regarding radiographic borders of the lesion [26,27]. The cause of bony hemangiomas is thought to be congenital [28]. However, in a significant number of cases a history of antecedent trauma can be elicited. In what is considered to be the pioneering report of this tumor, Toynebee suggested that the lesion was related to the centers of ossification of the involved bones [29].

Angiography to show a tumor blush offers an additional mode of diagnosis when the lesion is large [30]. As a result of the low incidence of such anomalies in the region of the TMJ, most literature descriptions of vascular lesions in this area are case reports [4,6] CT scan shows tumor with enhancing quality of blood vessels and will confirm the location of the tumor, as seen here in the present case, the tumor presented in the superficial lobe. However the soft tissue density of the tumor, its homogeneity, and any areas of extension are best demonstrated by MRI. Magnetic resonance imaging demonstrates hyper intensity on T1 weighted images and isointensity with muscle on T2 weighted images. MRI scan which allows clear visualization of joint involvement is useful to define the extension of the hemangioma and its relationship with surrounding soft tissues [31]. We believe that MRI should be the gold standard when it comes to investigation of choice to diagnose hemangioma in the oro-facial region and recommend that Dental surgeons and oral surgeons should be aware of clinical implications of such an entity during their clinical interactions.

### Conclusion

Hemangioma of the Temporomandibular Joint is a very rare pathological entity with an atypical radiographic appearance which also has scarce mention in the literature till date. From that perspective, we, authors sincerely believe it is our privilege to add this case report to the existing medical literature on these lesions. It is also our recommendation that MRI should be the modality of choice to diagnose hemangioma in the oro-facial region. Hemangiomas present as a benign growth and in most cases without any history of hemorrhage. Therefore, they pose a major diagnostic and clinical challenge especially to dentist, ENT and Oral Maxillofacial surgeons as a result of a biopsy or even a simple tooth extraction. Such health care professionals would be advised to consider this clinical entity in their differential diagnosis as this will be of significant help in devising their treatment plan and preventing any inadvertent clinical outcome.

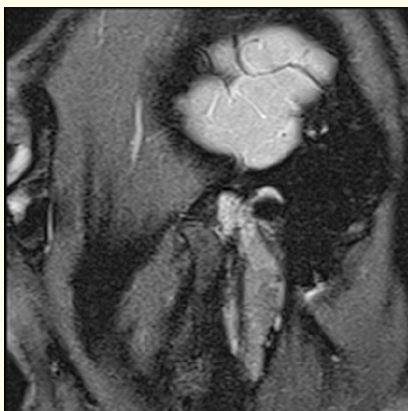


Figure 1:

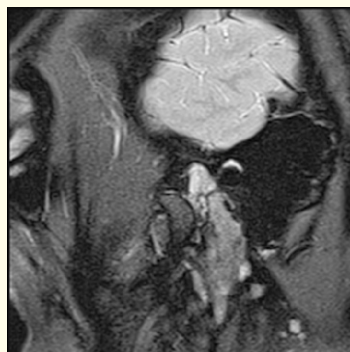


Figure 2:



Figure 3:

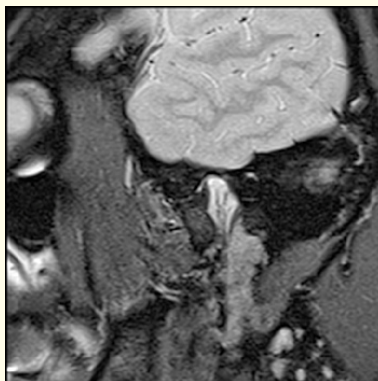


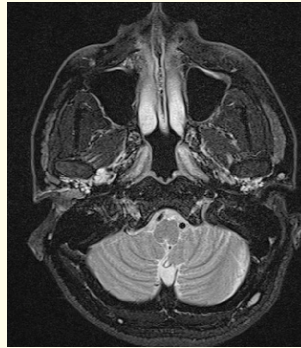
Figure 4:



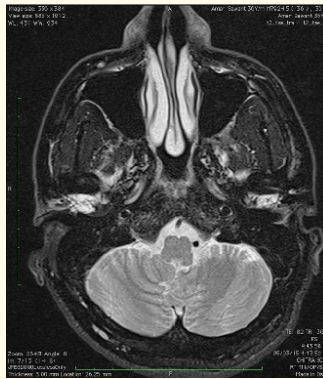
Figure 5:



Figure 6:



**Figure 7:**



**Figure 8:**

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