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Case Report

Erectile intramuscular hemangioma of the masseter muscle: A report of a case

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ABSTRACT

We report a rare case of erectile intramuscular hemangioma of the masseter muscle. The patient, a 41-year-old female, complained of a mass on the left cheek. Physical examination revealed a soft mass that was apparent only when the patient masticated. Computed tomography with contrast enhancement revealed a hypovascular mass with peripheral rim enhancement. On contrast-enhanced magnetic resonance imaging, a well-defined mass was identified within the posterior portion of the masseter muscle. The mass exhibited high signal intensity at its peripheral rim on T2-weighted images and showed isointense signal intensity to muscle on T1-weighted images. Fine-needle aspiration biopsy revealed only blood. These findings were supportive of an erectile intramuscular hemangioma. The mass was completely removed by the submandibular approach without postoperative complications. Histopathological examination of the mass revealed a vascular tumor composed of variably sized, thin-walled veins between skeletal muscle fibers, confirming the diagnosis of a racemose hemangioma. Computed tomography with vascular enhancement and contrast-enhanced magnetic resonance imaging were useful in the presurgical diagnosis of this intramuscular hemangioma.

1. Introduction

Intramuscular hemangiomas (IMH) are rare tumors, accounting for 0.8% of all hemangiomas and approximately 13.8% of cases occurring in the head and neck region [1,2]. The most frequently affected muscle is the masseter (36% of cases), followed by the trapezius (24%). Erectile intramuscular hemangioma (EIMH) of the masseter muscle is rare, and it becomes apparent only when the patient masticates. Usually, hemangiomas are easily distinguished from other soft-tissue tumors by computerized tomography (CT), magnetic resonance imaging (MRI), and arteriography. However, it is well known that most cases of IMH are misdiagnosed before surgery [3]. In the case of an EIMH, the diagnosis is simplified by the characteristic clinical findings. Here, we describe the clinicopathological findings in a case of EIMH.

2. Case report

A 41-year-old Japanese female presented at our hospital with a mass on the left cheek. She had first noticed the swelling 2 months previously. Physical examination revealed a soft, painless, and mobile subcutaneous mass that became apparent only when the patient masticated (Fig. 1). The mass was 20 mm × 15 mm in diameter and the overlying skin color was normal. Computed tomography with contrast enhancement revealed a well-circumscribed, hypovascular mass with peripheral rim enhancement located within the left masseter muscle (Fig. 2). On contrast-enhanced MRI, a well-defined mass was identified within the posterior portion of the masseter muscle. The mass was isointense to muscle on T1-weighted images and had high signal intensity at its peripheral rim on T2-weighted images (Fig. 3). Fine-needle aspiration biopsy revealed only blood. These findings strongly suggested an EIMH.

The operation was performed through a submandibular approach under general anesthesia (Fig. 4). The branches of the facial nerve were identified and preserved. A dark blue mass was identified within the masseter muscle and excised with a margin of normal muscle around it. The mass was well-circumscribed and measured 20 mm × 15 mm in size. Microscopically, the vascular tumor was composed of variably sized, thin-walled veins between

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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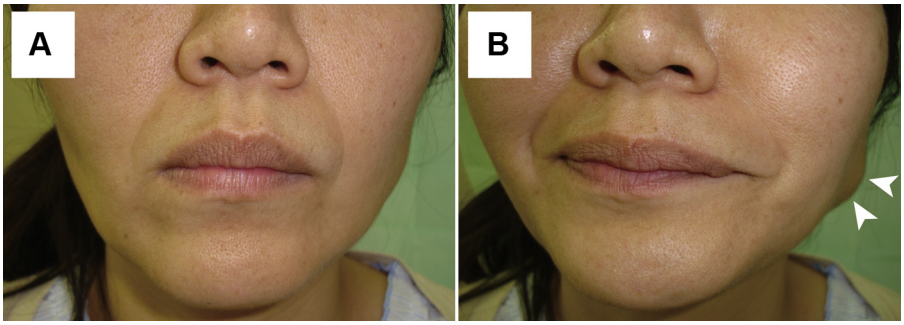

Fig. 1. Clinical presentation. (A) With the masseter muscles relaxed, no swellings were noted at the mandibular angle. (B) A soft, painless, and mobile subcutaneous mass (arrowheads) was visible when the patient masticated.

skeletal muscle fibers. An organized thrombus with fibrosis and fibrin deposition was observed within one of the dilated vascular channels, suggesting the presence of hemodynamic changes (Fig. 5). This histopathologic diagnosis was consistent with a racemose hemangioma.

The patient's postoperative course was uneventful; facial palsy and skin deformity were not observed. At one year after surgery, no evidence of recurrence has been seen.

3. Discussion

IMH of the skeletal muscle is a rare, benign tumor characterized by the proliferation of blood vessels. The lesion was first described by Liston in 1843 [4]. IMH typically presents in the 2nd and 3rd decades of life, and patients are usually asymptomatic until a growth spurt occurs. The cause of IMH remains unknown, although trauma and abnormal sequestration of embryonic tissue have been postulated [5]. A few cases of IMH occurring in the masseter muscle have been labeled as erectile hemangiomas [3,6–8], which become apparent only when the patient masticates. In these cases of EIMH, the diagnosis is made easier by this clinical finding. Histopathologically, most of the IMH reported previously were of the cavernous type. In our case, the pathological diagnosis was racemose hemangioma and it is thought to be rare. However, the correlation between these clinical findings and pathological findings remains unknown. Further study is required to clarify the pathogenesis.

Typically, hemangiomas are distinguished from other soft tissue tumors by CT, MRI, and arteriography. Magnetic resonance imaging has been shown to provide better detection of IMH and more accurate delineation of their extent than CT [9]. On MRI, hemangiomas are characteristically much brighter on T2-weighted versus T1-weighted images, and they are enhanced by gadolinium. On multiphase contrast-enhanced CT or MRI, hemangiomas show characteristic peripheral nodular enhancement in the early phase and gradual centripetal filling in the delayed phase. However, it is well recognized that giant hemangiomas show incomplete central filling because of central fibrosis or thrombosis [10,11]. However, a small number of hemangiomas show atypical enhancement patterns, such as the presence of intratumoral non-enhanced or less-enhanced components [12–15]. In the present case, high T2-weighted signal intensity was only seen at the mass's peripheral rim for this reason.

The differential diagnosis of IMH includes both parotid and other soft tissue tumors [2,16], but US, CT, MRI and arteriography are useful in distinguishing these. FNA typically shows only hemorrhage and is commonly non-diagnostic. FNA helps to exclude other soft tissue tumors, including malignant tumors, and it can be a supportive diagnostic tool [8].

The treatment of IMH should be individualized according to the tumor's size, location, extent, anatomical accessibility, and rate of growth as well as cosmetic considerations. With the accuracy of MRI, some patients can merely be observed, particularly young children [17]. However, wide surgical excision that includes normal

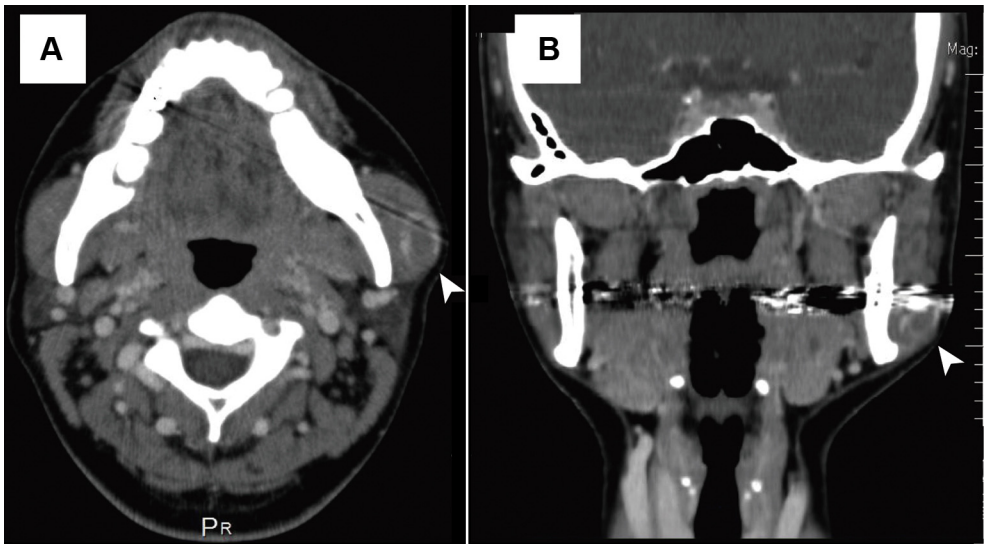

Fig. 2. Computed tomography with vascular enhancement. (A) CT with vascular enhancement (axial view) shows a well-circumscribed, hypovascular mass with peripheral rim enhancement located within the left masseter muscle (arrowhead). (B) Coronal reformatted image. The mass is indicated by the arrowhead.