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Intramuscular Hemangioma of the Masseter Muscle Mimicking Parotid Sialolithiasis

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Case Report

History

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ABSTRACT

Intramuscular hemangioma occurs in less than 1% of soft tissue hemangiomas. It is equally prevalent in men and women. We examined a case of a masseter muscle hemangioma with evidence of a suspected parotid sialolithiasis. A 24-year-old woman with a 3-year history of swelling and mild pain in the cheek and ear area was referred to our Department. Due to the calcification in the anterior region of the masseter muscle observed in earlier CT images, sialography was scheduled for the differential diagnosis of sialolithiasis. Ultrasound imaging (Sonography) was performed as an initial examination that showed a complex tubular mass of 20 × 35 mm in the anterior region of the masseter muscle. The mass appeared hypervascular on color Doppler ultrasound, confirming its vascular nature. Due to its proximity to the facial nerve, a diode laser was adopted for treatment.

Key words: Hemangioma, Sialolithiasis, Masseter Muscle, Ultrasonography, Computed Tomography (CT).

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Introduction

A hemangioma is a neoplasm of endothelial origin that consists of vascular spaces composed of endothelial cells. It usually occurs in infants in their first year of life and disappears with age. [1]Hemangioma is more apparent on the skin and in the subcutaneous tissue. A rare type of hemangioma is intramuscular hemangioma (IMH), with an equal prevalence in men and women. It accounts for less than 1% of soft tissue hemangiomas[1], of which about 13.8% occur in the head and neck area, usually in the masseter, sternocleidomastoid, and trapezius muscles.[2]

IMH etiology is unknown. Among the factors that have been mentioned are Excessive muscle contraction, trauma, menstrual cycle, and pregnancy.[3] Magnetic resonance imaging (MRI), computer tomography (CT), and ultrasound imaging help diagnose IMH.[4] Unlike other hemangiomas, IMH does not regress spontaneously and is usually diagnosed in the second or third decades of life. In addition, IMH can be mistakenly diagnosed as parotid or other salivary neoplasms, emphasizing the importance of correctly diagnosing this.[5]

In this study, we examined a case of a masseter muscle hemangioma with evidence of a suspected parotid sialolithiasis in a 24-year-old woman.

Case report

A 24-year-old woman was referred to our radiology department with a 3-year history of swelling and pain in her right cheek that sometimes spread to the muscles of the ear and neck. Due to the swelling in the parotid region and the suspicion of parotid sialolithiasis, the patient was initially prescribed panoramic radiography, during which no specific findings were observed. A CT with contrast was then performed for further examination. On CT, an opacity in the anterior region of the masseter muscle indicated the presence of sialolithiasis (Figure 1). The patient was referred for a sialography examination for confirmation of sialolithiasis to the oral and maxillofacial radiology department.

On clinical examination, a smooth, round swelling was observed in the anterior region of the masseter muscle (Figure 2). It was well-circumscribed soft mobile swelling with pain and tenderness which was noticeable when chewing, manipulating, or palpation. However, no evidence of sialolithiasis-like sialagogue stimulation swelling was found. This swelling was well circumscribed soft mobile Due to swelling of the masseter muscle region, an ultrasound imaging before the sialography opted for the initial examination which was done with her consent. An ultrasound of the cheek and anatomical proximity was performed by grayscale sonography (E-CUBE7, Opinion, South Korea) with a multi-frequency linear probe (3-12 MHz) at frequencies of 8-10 MHz on the sonograms, a mixed echo tubular mass of 35 × 20 mm was observed in the anterior region of the soft tissue of the right cheek, which was

hypervascular on the color Doppler, suggesting a hemangioma and vascular malformations (Figure 3). Phlebolith echogenic foci of 3 mm diameter were also observed, which confirmed the diagnosis. A low-power diode laser was suggested for treatment because of its proximity to the facial nerve and the impossible surgery. The patient's consent was obtained to publish the case report.

Discussion

Studies show that IMH is more common in people under 30, although it might also occur in older ones, according to some case reports. IMH is equally common in both sexes. However, the involvement of masseter muscles is more prevalent in men.[6] The swelling is either painless [7, 8 9]or painful [6], which is the patient's chief complaint. The patient in our study was a 24-year-old woman with painful swelling in the cheek area. Jain[10], Chandrasekar[11], ElHariti[8], and Murugan[9] case reports were women too. while in contrast to our patient, Righini[3], Suraj[2], Lee[12], Kim[13], and Murugan[9] case reports, age patients were over 30 years old. The swelling was chief complaint in all of the patients (Table 1).

Although the etiology of IMH is not yet fully understood. Studies suggest trauma plays a role in the development of hemangioma and may be related to the etiology and growth of the lesion.[1] Our patient had a history of trauma in the past three years and swelling in the same area afterward.

The turkey wattle sign is an unusual pathognomonic manifestation of hemangioma within the masseter or the parotid gland. The lesion reshapes and enlarges when the jaw is clenched or the position of the head is changed. This symptom refers to swelling of the arteries in the lesion, preventing venous return from the head to the superior vena cava. Usually, no changes are observed on the skin's surface, while clenching the jaw causes the lesion to become firm and prominent. While manipulating the target area in our patient or clenching the jaw, a swelling was visible, a symptom of the turkey wattle sign. In rare cases, the area's skin may be bluish, with increased regional warming associated with hypervascularization.[8] However, there were no specific findings on the skin surface of our patient.

Only 8% of IMH cases are diagnosed before surgery.[8] The lack of symptoms is one of the factors that cause its misdiagnosis, which is confirmed by the long history of similar cases (Table 1). Ultrasound imaging, CT, and MRI are used to diagnose IMH along with clinical examination. Conventional radiological techniques help identify phleboliths and calcifications.[14] However, these are not specific, as our patient's panoramic radiography did not contain any findings. The hemangioma appears as an ill-defined mass on non-enhanced CT scans with a similar attenuation to the muscles. Phlebolith is too small to be detected on radiography. A significant enhancement is obtained after injecting the contrast agent.[13] In our patient, because of the presence of swelling and pain in the clinical examination, CT scan was requested. On contrasted CT scan, phlebolith be enhanced. it was similar to sialolithiasis which led to a misdiagnosis. ElHariti and Lee used of CT scan like our case and report high density[8] and calcification[12] on it (Table 1).

Because of the patient's history of swelling and pain, and since no signs of sialolithiasis were observed during the clinical examination, we used ultrasound imaging for further examinations; it is inexpensive, without radiation dose and initial imaging step in patients with soft tissue swellings .[14] On the sonograms, the presence of a well-defined hypoechoic mass along with heterogeneous echotextures in the head and neck might indicate the presence of a hemangioma. In IMH, color Doppler ultrasound is helpful to diagnose vascular structures within and around the muscle, assess pathological changes such as fibrosis, and identify calcifications. Vascular lesions are characterized by abundant vascular and high blood flow.[14]

The shape of phlebolith and calcification has also been described as a diagnostic feature of hemangiomas.[8] In our patient, the hemangioma was identified on the patient's sonograms as a mixed echo tubular mass in the anterior region of the soft tissue of the right cheek, which was hypervascular on the color Doppler and confirmed the diagnosis. Chandrasekar *et al.* and Lee *et al.* reported similar observations as a mixed echo mass with calcification in the right muscle of the masseter.[12, 14] Murugan *et al.*[9] found an isoechoic lesion in the masseter muscle, while Jain *et al.* and Makkad et al. observed a lobular and hypoactive mass in the muscle.[5, 10] In these case reports, the presence or absence of phlebolith resulted in a difference in the sonograms as mixed echo or hypoechoic. Most of the articles used the MRI T2 images for IMH diagnosis, which were seen as heterogeneous and mixed hyper and hypointense. But since ultrasound has better access and less cost than MRI, it was used in our case. In the differential diagnosis of IMH of the masseter muscle, conditions including hypertrophy, myofascial pain, sialolithiasis, parotid gland tumor, lymphangioma, lymphoma, rhabdomyosarcoma, and schwannoma are considered. For our case, due to evidence such as myofascial pain, cheek swelling on the differential diagnosis of the masseter muscle hypertrophy, and the CT with contrast, parotid sialolithiasis was present and suggested.

Various methods have been proposed for treating hemangiomas, including sclerosing agents, radiation, and surgery. However, the optimal accepted treatment is the excisional surgery of the lesion and the muscle around it. However, excisional surgery is associated with an increased risk of damage to the facial nerve. On the other hand, the diode laser is also effective in treating hemangiomas and vascular malformations. It reduces the lesion size and the disease symptoms and is better tolerated by the patient. Because of the reduced risk of facial nerve damage along with the benefits of using a diode laser [15], low-power laser treatment was suggested for our patient. For follow-up, the ultrasound was not performed on the patient, but after a few months, the patient expressed an improvement in pain and swelling in the area clinically.

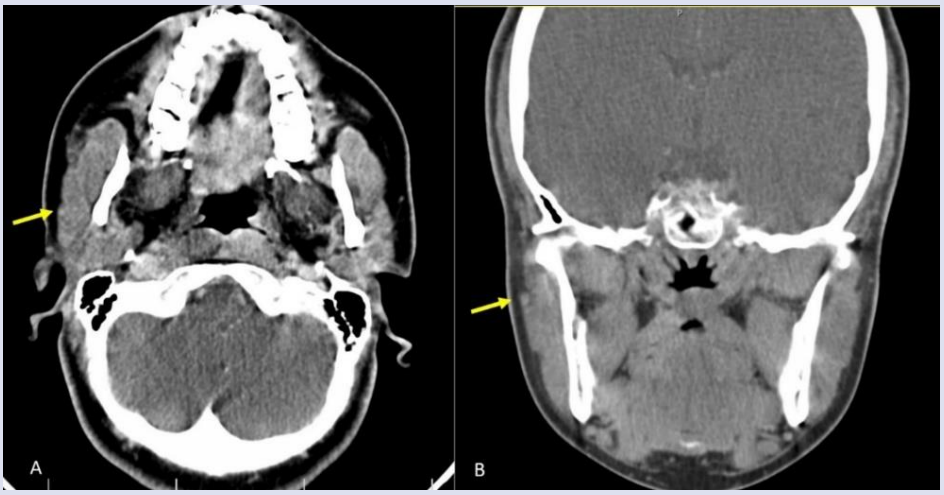

Figure 1. CT scans axial and frontal view: hypo to Iso attenuation mass in the right masseter muscle with phlebolith(arrow).

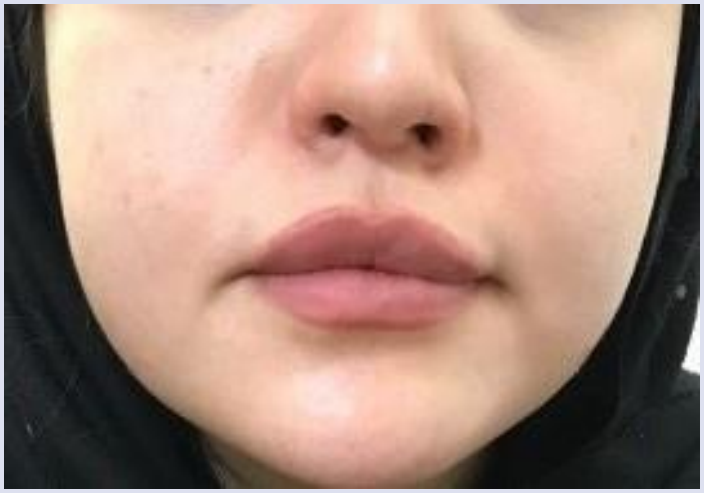

Figure 2. Slight swelling was observed in the anterior region of the right masseter muscle

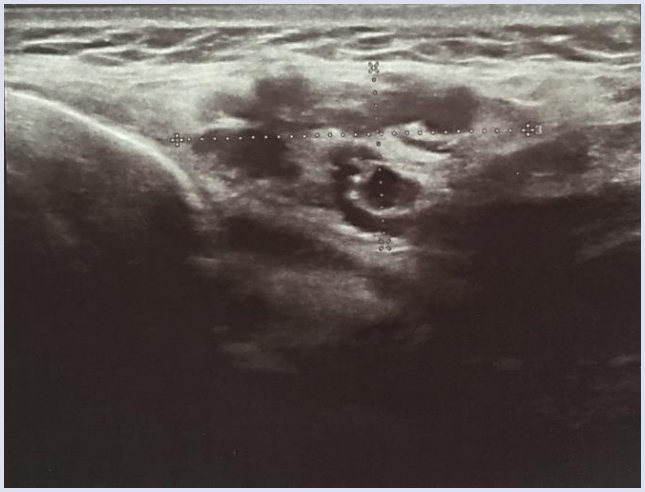

Figure 3. Ultrasound image revealing a mixed echo lesion with a phlebolith in the right masseter

Table 1. Intramuscular Hemangioma case reports and radiographic findings

Study	Year	Age	Gender	Chief complain	History	Imaging modality	Imaging findings
Jain et al. (13)	2011	8	Female	Facial swelling	3 year	USG Color Doppler USG MRI	USG: Lobulated hypoechoic mass Color Doppler ultrasound: Evidence of color flow (internal vascularity) MRI: Isointense on T1 Nonhomogeneous hyperintense in T2 and PD
Righini et al. (3)	2014	70	Male	Firm painless well-contoured swelling	2 year	MRI	MRI: Well-Contoured mass, contrast-medium uptake
Chandrasekar et al. (10)	2014	23	Female	Painful swelling	6 months	USG Color Doppler USG MRI	USG: Mixed echoic lesion with a speck of calcification Color Doppler: Dilated vascular channels with good flow MRI: T2 mixed (hypo and hyperintense) mass signal, space-occupying lesion
Surej Kumar et al. (2)	2016	35	Male	Firm swelling	3 year	MRI Angiogram	Enhanced- well-circumscribed intramuscular mass
Lee et al. (12)	2016	42	Male	Muscular hypertrophy and Swelling during clenching Several months after treatment for hypertrophy: painful swelling	At childhood	USG CT	USG: Non-homogenous echo CT: swelling with calcification
ElHariti et al.(8)	2017	16	Female	Painless, well-circumscribed, firm swelling	2 year	CT scan MRI	CT: High density MRI: Voluminous encapsulated tissue mass in iso signal T1 and T2
Kim et al. (13)	2017	48	Male	Swelling	--	MRI External carotid angiography	MRI: strongly enhanced with heterogeneous T2 With multiple vascularity as signal voids. External carotid angiography: The blushed mass by contrast agent injection and gradually disappeared with time
Murugan et al. (9)	2018	37	Female	Painless dependent swelling	4 year	USG Color Doppler USG MRI	USG: Isoechoic lesion Color doppler USG: minimal color flow MRI: Intermediate signal on T2W1 and strong enhancement embedded in the anterior side of the masseter muscle
Makkad et al. (5)	2021	25	Male	Incidental finding of well-swelling	Gradual growth	USG Color Doppler USG	USG: lobulated, hypoechoic mass Color Doppler USG: Enhanced vascularity within the lesion
Present Case report	2023	24	Female	Dull pain and swelling	3 year	CT USG Color Doppler USG	CT: Opacity anterior of the masseter USG: Mixed echo tubular mass Color Doppler USG: Hyper vascular

Conclusions

Masseter muscle hemangioma can be associated with vague symptoms such as myofascial-sialolithiasis pain, parotid gland tumor, and masseter muscle hypertrophy. A careful ultrasound examination of the parotid gland and the anatomical proximity of the area, especially the masseter muscle, can help diagnose the condition promptly. It is suggested that in such cases, similar to the present case, before prescribing CT, which has a high amount of radiation,

ultrasound should be used as the initial image, and after that, CT should be prepared if necessary.

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Conflicts of Interest Statement

The authors declare that they have no conflict of interest.

Informed consent

Informed consent of the patient has been obtained for this case report.

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