

Intramuscular hemangioma presenting with multiple phleboliths: a case report

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A rare case of intramuscular hemangioma of the masseter muscle with multiple phleboliths is described, highlighting features evident in plain radiography, ultrasound, and magnetic resonance imaging (MRI). A 21-year-old woman presented with a complaint of swelling of the right masseter muscle. A plain radiograph from the soft tissue of the right cheek showed a large number of round, target-like radiopacities that varied in size. Ultrasound revealed a lobulated cystic lesion in the right masseter muscle with increased echogenicity. MRI showed a space-occupying lesion in the right masseter muscle, which was isointense on T1-weighted image close to the muscle tissue and hyperintense on T2-weighted image, containing fields with no signal septations. A plain soft tissue radiograph can demonstrate phleboliths and aid in the diagnosis of an intramuscular hemangioma. In addition, nonionized techniques such as ultrasound and MRI can provide useful information to clinicians regarding the location of calcifications and the structure of masses. (Oral Surg Oral Med Oral Pathol Oral Radiol 2013;115:e32-e36)

Developmentally, intramuscular hemangiomas (IMHs) represent congenital vascular malformations.¹ The predominant complaint is the presence of a slowly enlarging mass. IMHs are classified according to their histologic appearance as either capillary, cavernous, or mixed small/large vessel types. They generally develop in patients during the first 3 decades of life² with no gender predispositions.

Fifteen percent of IMHs occur in the head and neck region,^{2,3} with the masseter muscle is the most common site.⁴ The differential diagnosis of a mass in the masseter muscle includes benign muscular hypertrophy, lymphadenopathies, a sialoceles of the parotid gland duct, and various parotid and muscle neoplasms.^{1,5}

Phleboliths are calcified thrombi that are a characteristic feature of hemangiomas, occurring in 15%-25% of IMHs.^{2,3,6} They are generally variable in size,⁷ found in multiple number, and cause no subjective symptoms.

The diagnosis of IMHs can be difficult, because of the deep intramuscular location, rare incidence, and lack of specific symptoms that suggest a vascular-origin lesion. Definitive preoperative diagnosis has been reported in <8% of cases.⁸ Although standard radiographs are simple and constitute an important diagnostic tool in diagnosing the phleboliths within the mass,⁵ other diagnostic imaging modalities, such as comput-

erized tomography (CT), magnetic resonance imaging (MRI),^{9,10} and ultrasound,¹¹ also play a significant role in the preoperative diagnosis of IMHs.

To the best of our knowledge, no previous study has included plain radiography, ultrasound, and MRI images of an IMH with phleboliths in the maxillofacial region. The present report describes an IMH of the masseter muscle with multiple phleboliths, highlighting the features evident in plain radiography, ultrasound, and MRI.

CASE REPORT

A 21-year old woman presented with a complaint of swelling of the right masseter muscle causing a cosmetic deformity. The swelling had been present since 6 months of age and had gradually grown as the patient aged. A temporary increase in the size of the mass was observed when the patient cried, laughed, or performed handstands. Besides caries on the right upper first molar, no dental disease was observed. The patient's medical and family history were unremarkable.

On physical examination, there was asymmetry to her face (Figure 1). Bimanual palpation of the right cheek revealed a mass with hard nodules of various sizes. The mass was immobile and could not be definitively separated from the masseter muscle or the parotid gland. There were no bruits or pulsation present. Intraoral examination revealed normal-colored mucosa and normal discharge from the right parotid gland.

Periapical films from the soft tissue of the right cheek (Figure 2) and the decayed upper right first molar (Figure 3) showed large numbers of round target-like radiopacities varying in size from 2 to 10 mm. Based on radiographic and clinical findings, the lesion was suspected to be a sialolith or phlebolith, and the patient was sent for ultrasound and MRI, respectively. Ultrasound examination showed a lobulated cystic lesion, ~2 × 5 cm in size, located in the right masseter

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Fig. 1. Facial asymmetry due to intramuscular hemangioma.

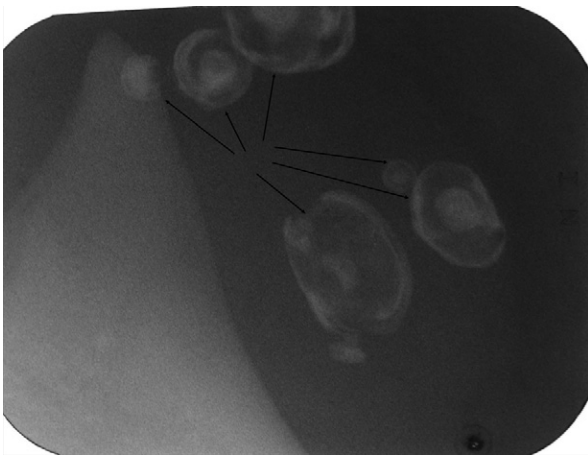


Fig. 2. A periapical film from the soft tissue of the right cheek clearly showing multiple round target-like radiopacities varying in size from 2 to 10 mm (arrows).

muscle. Nodular structures with heterogeneous internal echogenicities were found within the lesion (Figure 4). MRI showed a space-occupying lesion in the right masseter muscle, 45 × 22 mm in axial dimensions, isointense on T1-weighted images, close to the muscle tissue and hyperintense on T2-weighted images, containing fields in which no signal septations or calcific foci were detected. The benign lesion showed heterogeneous intense contrast in contrasted sections and nonuniform intense septations in the center; however, it did not show diffusion limitation in diffusion-weighted sections, but did contain large no-signal fields along with microcalcifications (Figure 5). The diagnosis of an IMH with phleboliths was made based on these findings.



Fig. 3. A periapical film from upper right molar, showing phleboliths (arrows).

The patient was informed about her condition, and treatment options were discussed; however, the patient elected to forego treatment at that time.

DISCUSSION

IMHs are congenital lesions representing 1% of all hemangiomas^{2,3} with 15% of lesions occurring in the head and neck region.^{2,3} The most common site is the masseter muscle.^{12,13}

Hemangiomas usually occur in the first 3 decades of life.¹³ It has been suggested that they arise from malformed tissue that has been subjected to repeated trauma¹³ or are the result of hormonal factors.¹²

IMHs rarely display any clinical symptoms or signs that reveal their vascular nature.⁵ There are usually no overlying skin changes,¹⁴ although there may be occasional reddish-blue discoloration.^{14,15} Thrills, bruits, compressibility, and pulsation are usually absent¹⁴; however, pain can be present.^{5,15} Situations that increase the venous pressure in the head increase IMH size.¹ In the present case, the mass significantly enlarged during crying, laughing, or performing handstands, which all increase the blood pressure in the head and neck area. In this case, the oral mucosa of the patient was normal and pain was not present.

The formation of phleboliths typically causes no symptoms. Phleboliths consist of a mixture of calcium carbonate and calcium phosphate salts¹⁶ and are thought to form when a fibrous component attaches to a developing phlebolith and becomes calcified. Radiologically, they have either a radiolucent or a radiopaque core, and repetition of this calcification causes an onion-like appearance or concentric rings. In the maxillofacial region, they are usually multiple and vary in size. The differential diagnosis of phleboliths includes other causes of calcifications in the head and neck area, such as sialolithiasis, tonsilloliths, healed acne lesions,

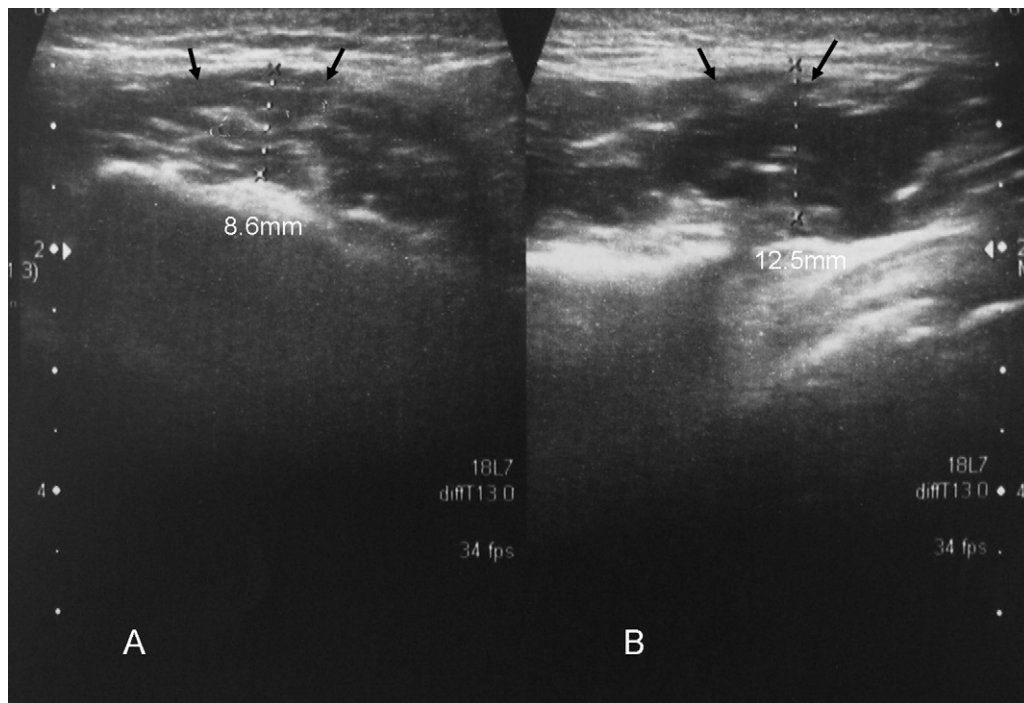


Fig. 4. Ultrasound examination shows 2 nodular structures with heterogeneous internal echogenicity ~9 (A) and ~13 (B) mm thick, with smooth margins in the subcutaneous area (arrows).

cysticercosis, miliary skin osteomas,⁷ calcified lymph nodes, and carotid artery calcifications.

Plain radiographs can show soft tissue calcifications. The presence of round, smooth, and laminated phleboliths is pathognomonic for a cavernous hemangioma.¹⁷ The plain radiograph (Figure 2, periapical film taken from the cheek) of the present patient demonstrated multiple small (2- to 10-mm diameter) target-like opacities within the mass. In addition, other diagnostic imaging modalities such as CT,¹⁵ MRI,^{9,10} and ultrasound¹¹ increase the accuracy of a preoperative diagnosis for this lesion. Ultrasound has been shown to be a reliable method for diagnosing alterations in the masseter muscle.¹⁸ MRI is considered to be the most useful imaging modality for tissue characterization and recognition of the extent of a lesion.⁹ In general, vascular malformations and hemangiomas demonstrate hyperintensity on T2-weighted images and isointensity on T1-weighted images⁹ owing to the increased free water present within stagnant blood in the vessels.

The therapeutic approach is based on clinical factors such as age, cosmetic appearance, size, location, and depth of involvement. Many forms of therapy have been suggested, including cryotherapy, radiotherapy, sclerosing agents, and steroids,¹² but the ideal treatment is complete excision of the tumor, thereby eliminating the phlebolith.⁷ Even with this approach, local recurrence rates ranging from 9% to 28% have been re-

ported.¹² In addition, total excision of the masseter muscle has been recommended.¹⁹ In our case, the patient was informed about the condition and treatment options discussed; however, she elected to forgo treatment at the time.

In conclusion, palpation of small hard nodules deep within the muscle that are diffuse and compressible should alert the clinician to the possibility of an IMH with phleboliths. Plain soft tissue X-ray image can show phleboliths, and nonionizing techniques, such as ultrasound and MRI, can provide useful information to clinicians about the location of calcifications and the extent of the lesion.

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Fig. 5. **A**, T1-weighted sagittal MR scan shows isointense mass $\sim 5 \times 6$ cm in size with distinct borders in the right masseter muscle (*black arrows*). Millimetric slightly hyperintense nodular areas are seen within the homogeneous mass (*white arrows*). **B**, T2-weighted coronal MR scan shows millimetric hypointense structures (*black arrows*) in the hyperintense mass. **C**, Contrast-enhanced T1-weighted MR scan demonstrates many nonenhancement nodular structures (phleboliths) (*black arrows*) in the strongly enhancing mass with smooth borders (*white arrow*).

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