INTRAMUSCULAR VASCULAR MALFORMATION OF THE MASSETER MUSCLE - A CASE REPORT

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ABSTRACT

Intramuscular vascular malformations are occasionally diagnosed as hemangioma, as similar characteristic features have been reported, particularly on image findings. Haemangioma are benign endothelial cell-derived tumors, while vascular malformations are not true tumors, but instead are congenital abnormalities without increases in endothelial cell proliferation. Haemangiomas become evident shortly after birth as expansive lesions that gradually disappear with age. Vascular malformations are present at birth and continue to grow throughout the patient's lifetime. In this report, we present an intramuscular vascular malformation of the right masseter muscle in an 8-year-old boy. Swelling with intramuscular calcification was observed on computed tomography (CT) images. Magnetic resonance imaging (MRI) showed a well-defined space-occupying lesion of the masseter muscle that was isointense to the surrounding muscle on T1-weighted images, heterogeneously hyperintense on T2-weighted images, and heterogeneously enhanced by gadolinium contrast medium. This lesion included several low signal areas on MR images. Compared to the CT images, some low signal areas were considered phleboliths, whilst others were flow-void or stroma-like fibrous septa. We finally diagnosed this lesion as an intramuscular vascular malformation based on the MR findings.

Key words: Hemangioma, Masseter Muscle, MRI, Vascular Malformation

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INTRODUCTION
Vascular malformations are not true tumors, but instead are congenital abnormalities that demonstrate no endothelial cell proliferation. Unlike a hemangioma, vascular malformations are present at birth, do not resolve spontaneously, and become more apparent as the child grows. In addition, vascular malformations may increase in response to trauma, infection, and changes in the endocrine environment during adolescence or pregnancy. In 1982, Mulliken et al. 2 compiled a clinical and histopathological classification to separate haemangioma and vascular malformations. Vascular malformations are further divided into four subgroups based on the type of blood vessel predominantly affected: capillary, venous, arterial, and lymphatic. A more recent study showed that intramuscular vascular malformations presented a mixed density on non-enhanced CT image. However venous malformations are not easily detectable on enhanced CT due to their slow vascular flow. Phleboliths are a useful diagnostic finding for this type of lesion on CT imaging. Venous malformations and haemangioma are difficult to distinguish through magnetic resonance imaging (MRI). An arterial malformation displays a high-flow lesion corresponding to an arterio-venous fistula or malformation, resulting in the appearance of a flow-void on MRI. Karaman et al. 4 reported spiculated flow boid vascular structures on MRI of their huge arteriovenous malformation in right masseter muscle. The co-existence of both venous and arterial malformations is rare. In the present case, the characteristic signal patterns of both the stroma in the venous malformation and the high-flow artery in the arterial malformation were also observed on MRI. Vascular malformations tend to develop in the head and neck regions, with the lips, tongue, and buccal mucosa the most predominant sites. Vascular malformations appear as a type of birthmark and are almost completely visible superficially. An intramuscular lesion is comparatively rare. In this study, we present an intramuscular vascular malformation of the masseter muscle in an 8-year-old boy.

CASE REPORT
An 8-year-old boy visited our dental hospital with a painless swelling on his right cheek. His mother had noticed this swelling several years earlier, but since no pain was present, the swelling was not assessed. Following trauma to the right cheek a few days earlier, pain appeared following both biting and the application of pressure. In the intra- and extra-oral examination, a well-defined elastic hard mass with a diameter of approximately 30 mm was palpated in the right cheek. Trismus, odynophagia, and a tenderness of the submandibular lymph node were not observed. Abnormal findings, such as phleboliths were not observed on panoramic radiographs (Figure 1); however, a swelling of the right masseter muscle with some intramuscular calcification was observed on CT images (Figure 2). A slight decrease in CT number in the intramuscular region was also evident. However, a well-defined margin of the lesion was not confirmed. Destruction or compression of the mandible was not evident near the right masseter muscle. The right parotid gland and other right muscles, such as the lateral and medial pterygoid muscles and the temporal muscles were intact on CT images.

MRI showed a space-occupying lesion of the masseter muscle that was isointense to the surrounding muscle on T1-weighted images, heterogeneously hyperintense on T2-weighted images, and heterogeneously enhanced by gadolinium contrast medium (Figures 3-5). The intramuscular vascular lesion was surrounded by an indistinct moderate signal area on T1-weighted images. Several low signal linear or round areas were observed in the intramuscular region on T2-weighted images, suggesting a well-defined high signal mass lesion (Figure 4). Heterogeneity of the intramuscular lesion was highlighted and the margin of the lesion became unclear on gadolinium-enhanced T1-weighted images (Figure 5). Under the diagnosis of an intramuscular vascular malformation, surgical extirpation was performed extra- orally under general anaesthesia (Figure 6). There were no complications and a good prognosis following surgery. Histopathological examination revealed the composition of an irregular hyperplasia of the venous vessel and the formation of phleboliths (Figure 7).

DISCUSSION
In 1982, Mulliken et al. 2 histopathologically divided "common hemangioma" into two groups: a hemangioma that develops with the hyperplasia of the vascular endothelium and a vascular malformation with no increase in vascular endothelium proliferation. Jackson et al. 8 additionally classified haemangioma into three groups based on vascular dynamics: a hemangioma, a vascular malformation, and a lymphatic malformation. Based on this
classification scheme, it was considered that the treatment of a hemangioma should be delayed until involution is complete. Sclerotherapy represents a useful procedure for the treatment of low-flow vascular malformations. An embolization or partial resection should be considered for high-flow vascular malformations. In 2003, the International Society for the Study of Vascular Anomalies (ISSVA) established a strict classification of vascular lesions (Table 1). Recent policies regarding diagnosis and treatment were based on this classification. Vascular malformations tend to develop in superficial areas such as the lips, tongue, and the buccal mucosa of the oral cavity. However, an intramuscular vascular malformation...
Clinically, the diagnosis of intramuscular vascular malformations is difficult, as no clear colour changes are evident, and no characteristic view exists. Rai et al. described that intramuscular vascular malformations showed a mixed density on non-enhanced CT image. However, in our case, a mixed density was unclear in the right masseter muscle because of an edge artefact of the mandible on non-enhanced CT images. Since phleboliths occasionally develop in low-flow vascular lesions, an intramuscular calcification observed on CT images may be a useful indicator for the differential diagnosis of vascular lesions. Compared with CT examination, MRI may represent a useful diagnostic tool to determine the extent and internal characteristics of these lesions. Three characteristic MR image findings regarding the vascular component, the stroma component, and the neighbouring muscular tissue have been reported. Firstly, the intramuscular vascular lesion in this case was isotense and surrounded by an indistinct moderate signal area on T1-weighted images. Cohen et al. also described a similar margin of vascular lesions (short-TR/TE sequence), suggesting that fatty linear strands surround the multiple blood vessel lesions. Secondly, a serpiginous internal lesion pattern was observed in our case on T2-weighted images. This may indicate that the intervening linear or round areas of low-signal intensity defining the high-signal areas correspond with the fibro-fatty septa and thrombosed vascular channels on T2-weighted images. Thirdly, Kadota
et al.\textsuperscript{13} reported amyotrophy of the affected muscle due to the growth of the vascular lesion. However, adipose degeneration of the masseter muscle was not observed in this case.

We diagnosed the present case as an intramuscular vascular malformation based on clinical findings, CT and MR images, and histopathological examination. Haemangioma in children typically spontaneously disappears with age; however, our case showed a widely spreading mass within the masseter muscle with some phleboliths. A co-existing arterial component was suspected based on several sizes of slow flow signal areas on MRI, indicating a flow void; however, a simple venous malformation did not display a flow-void on MRI. In fact, Karaman et al.\textsuperscript{4} reported the speriginous flow boid vascular structures on MRI of their case. As indicated by Smith et al.\textsuperscript{17} MRI remains the imaging modality of choice for the management of vascular lesions of the masseter muscle.

**CONCLUSION**

In conclusion, when an intramuscular vascular malformation is doubted, MRI represents the first choice to determine the range and internal characteristics of the lesion and should be employed for determining the most appropriate treatment strategy for vascular lesions.

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**REFERENCES**


2. Mulliken JB, Glowacki J. Haemangioma and vascular malformations in infants and children: a classification based on...


