

CT and MRI Findings of Head and Neck Masson's Tumor: A Rare Case Report and Systematic Review of the Literature

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Background: Masson's tumor (MT) is a rare benign vascular disease. In literature, detailed description of its radiological findings is not available and functional imaging such as diffusion weighted (DW)-MRI has never been described. We aim to summarize the CT and MRI findings in our representative case and to conduct a systematic review of the literature.

Case Presentation: We reported a 54-year-old ex-smoker male patient who presented with a nodular mass to the left cheek. He denied any previous trauma. CT examination performed on initial presentation revealed a well circumscribed solid oval mass with soft tissue density, a calcified focus and no significative contrast enhancement after contrast administration. MRI showed a well circumscribed solid oval mass, with intermediate T2 signal intensity with areas of high T2 signal intensity and the presence of peripheral high-flow serpentine vessels, low T1 signal intensity. The mass showed a non-enhancing area with enhancing vessels after intravenous contrast administration. We surveyed CT and MRI findings of head and neck MT of English and French language papers, published from 1981 to 2019, together with our representative case. We included articles with a description of CT and/or MRI findings of head and neck MT.

Conclusion: We have experienced one case and have evaluated imaging findings through systematic review. Only 36 articles were eligible. CT and MRI findings were reported in 27 and 23 articles, respectively. To date, no diffusion weighted imaging (DWI)-MRI findings have been described. The most frequent findings in CT were a well-defined mass with high or soft tissue density. The most frequent MRI findings were a non-homogeneous signal intensity in T1 and T2 weighted sequences, with foci of hyperintensity, multiple septations or flow voids. After contrast administration, the enhancement could be homogeneous, non-homogeneous, nodular or peripheral. In our case, we found a non-homogeneous hyperintensity in DWI-MRI with an area of restricted diffusion and low apparent diffusion coefficient (ADC) was observed ($0.963 \times 10^{-3} \text{ mm}^2/\text{s} \pm 0.12 \text{ SD}$). The imaging characteristics cannot provide a pre-operative identikit of MT and surgical removal is necessary to accurately differentiate it from malignant angiosarcoma but radiological evaluation is useful in surgical planning.

Keywords: Masson's tumor, case report, computed tomography, magnetic resonance imaging, diffusion weighted imaging

Background

Masson's tumor (MT), also known as intravascular papillary endothelial hyperplasia (IPEH) or Masson's hemangioma, is a rare benign vascular disease with an exuberant endothelial proliferation in blood vessels that usually affects the

extremities including head, neck and upper limbs.^{1,2} It is most frequent in the digits and in rare circumstances, lesions have been observed in the abdominal organs or in intracranial aneurysms.³

The lesion can arise from normal blood vessels, and in this case it is referred to as a “pure form”, or in a pre-existing vascular lesion, the so-called “mixed form”.

It is slightly more frequent in women (1.3:1). Its aetiology is generally considered as multifactorial even though in 30% of cases it arises after a local trauma or it is associated with vascular conditions (hemangiomas, vascular malformations).⁴

Radiological diagnosis of MT is usually challenging, due to the infrequent occurrence in head and neck and the similarity with vascular tumors like low grade angiosarcoma or Kaposi's sarcoma.

In particular, it is not a typical pathologic condition of the buccal space and the knowledge of the expected CT and MR imaging findings can be helpful for the radiologist on differential diagnosis.

However, histology has to be confirmed by surgical removal and pathology.

We show here a representative case of MT of left buccal space, of which diagnostic and therapeutic experience remain limited. Further, we surveyed Computed tomography (CT) and Magnetic Resonance Imaging (MRI) findings of head and neck MT in English and French language papers, published from 1981 to 2019, to evaluate if the imaging characteristics of MT can suggest the diagnosis pre-operatively.

Case Presentation

The patient provided written informed consent for publication of this case report and accompanying images. This case presentation was accepted by the Ethics Committee of Humanitas Clinical and Research Center and it was conducted in accordance with the principles of the Declaration of Helsinki.

The subject is a 54-year-old ex-smoker male patient who presented at our ENT out-patient clinic in October 2019 with an apparently painless swelling on the left cheek, without facial palsy, complaining of a mass that appeared four months previously. He suffered from hypertension and dyslipidemia and denied any previous trauma to the cheek. Twenty five years ago he has undergone surgical removal of back lipoma and fifteen years ago a reconstruction of the anterior cruciate ligament.

The clinical examination revealed a mass about 3 cm on the left cheek, of skin colour and hard consistency, which was not attached to the subcutaneous tissue or to the skin. The lesion had been increasing slightly in size over the last months.

Ultrasound examination performed in another Hospital in August 2019 revealed a hypo-anechoic oval mass about 30 x 15 mm with thin septa and a vascular pole inside.

In October 2019, he underwent ultrasound-guided tru-cut (18 G) needle biopsy and pathology examination revealed neoplasm of vascular origin.

CT examination was performed in October 2019 using a REVOLUTION CT scanner from GE Medical Systems (Illinois, United States), imaging was initiated 70 seconds after the start of intravenous administration of 100 mL of nonionic iodinated contrast agent (iopamidol, Imeron 400; Bracco-Altana Pharma, Konstanz, Germany) through an antecubital vein at a flow rate of 2 mL/s. Examination parameters were as follows: tube voltage 120 kVp, reference current-time product of 79 mAs per rotation; rotation time, 0.5 seconds; pitch, 0.9; collimation, 2 x 64 x 0.6 mm. We performed a late scan at 80 seconds with tube voltage of 80 kVp to increase iodine attenuation and image contrast of soft-tissue structures.

CT revealed a well circumscribed solid oval mass about 38 mm with soft tissue density, a calcified focus and no significative contrast enhancement after contrast administration (Figure 1) in the left buccal space.

A month later, he underwent MRI examination performed using a 1.5 Tesla MAGNETOM Avanto (Siemens Healthcare, Erlangen), with TSE T1-weighted sequences (TE 9.9 ms, TR 646 ms, Flip angle 220°, Field of view 220 mm, slice thickness 3 mm), BLADE T2-weighted sequences (TE 141 ms, TR 8.0 ms, Flip angle 90°, Field of view 220 mm, slice thickness 3 mm), 3D Volumetric interpolated breath-hold examination (VIBE) sequence (TE 1.6 ms, TR 4.4 ms, Flip angle 13°, Field of view 240 mm, slice thickness 0.8 mm).

MRI showed a well circumscribed solid oval mass about 39 mm, with intermediate T_2 signal intensity with areas of high T_2 signal intensity and the presence of peripheral high-flow serpentine vessels (low signal intensity on T_2 weighted images) (Figure 2A) and low T_1 signal intensity (Figure 2B). The mass showed a non-enhancing area with enhancing vessels after intravenous contrast administration (Figure 2C). In depth, it showed extension in the masticatory space, where it imprinted the medial pterygoid muscle.



Figure 1 CT findings of our representative case of a 54-year-old man with MT in left buccal space. Axial contrast-enhanced CT image demonstrates a well circumscribed solid oval mass (yellow arrow) with soft tissue density, a calcified focus and no significant contrast enhancement after contrast medium administration.

Diffusion Weighted Imaging (DWI)-MRI is a functional tool that can help to differentiate benign from malignant lesions. In this case, DWI was performed with 3-scan trace approach at b values of 50, 500 and 1000 s/mm². The mass

showed a non-homogeneous hyperintensity on trace images with an area of restricted diffusion and low Apparent Diffusion Coefficient (ADC) in the corresponding map (mean ADC value was 0.963×10^{-3} mm²/s) (Figure 3).

Imaging suggested a mass of the left buccal space with morphological pattern compatible with the histological results of vascular neoplasm. The signal restriction in DWI-MRI, that is possible expression of thrombus or malignant degeneration, suggested further histological characterization on the entire volume of the lesion after surgical removal.

The lesion had been totally excised by trans-oral approach in December 2019. It appeared covered by the Bichat's fat pad and it was richly vascularized and in continuity with the pterygoid muscle.

The final pathologically diagnosis was MT.

On the pathologic specimen from surgery, a mass containing dilated small caliber vessels was seen in addition to a thrombus and anastomosing network of little vessels with some fibrous septa. The mass was surrounded by muscular fibers (Figure 4).

At first follow-up visit, one month after surgery, there have not been post-operative complications. Five months after surgery no clinical signs of tumor recurrence were found and the patient has not undergone any treatment; his prognosis was good.

Discussion

MT, described for the first time in 1923 by Masson,^{5,6} may mimic other benign lesions including mucocoele, pyogenic

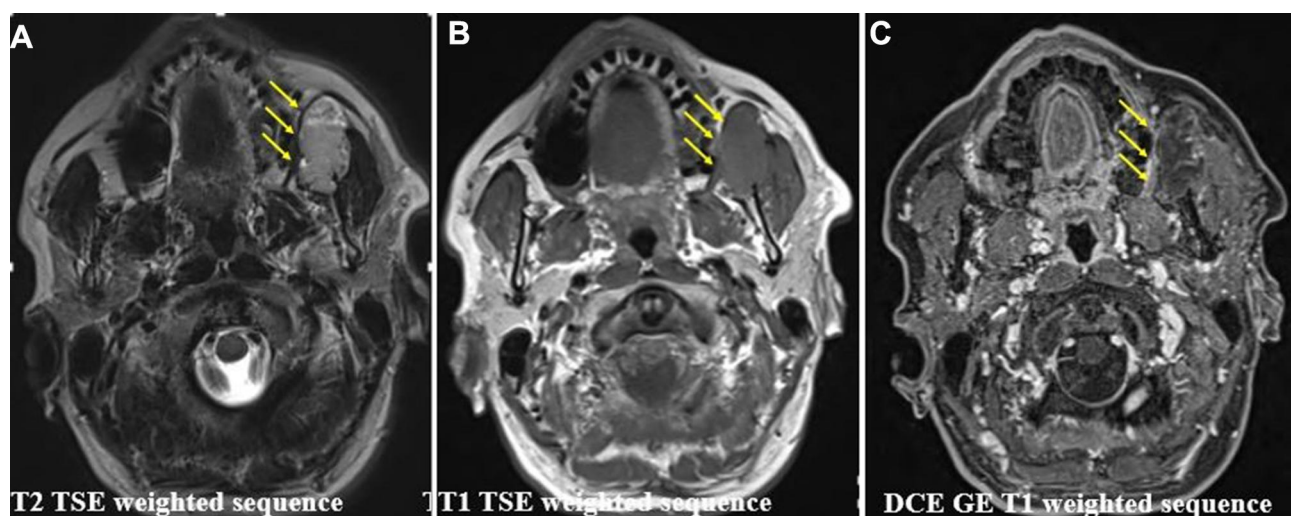


Figure 2 MRI findings of our representative case of a 54-year-old man with MT in left buccal space. (A) Axial T2 sequence showed a well circumscribed solid oval mass (yellow arrows) with intermediate signal intensity with areas of high signal intensity and the presence of peripheral high-flow serpentine vessels. (B) Axial T1 sequence showed low signal intensity of the oval lesion (yellow arrows). (C) The mass (yellow arrows) showed a non-enhancing area with enhancing vessels after intravenous contrast administration.

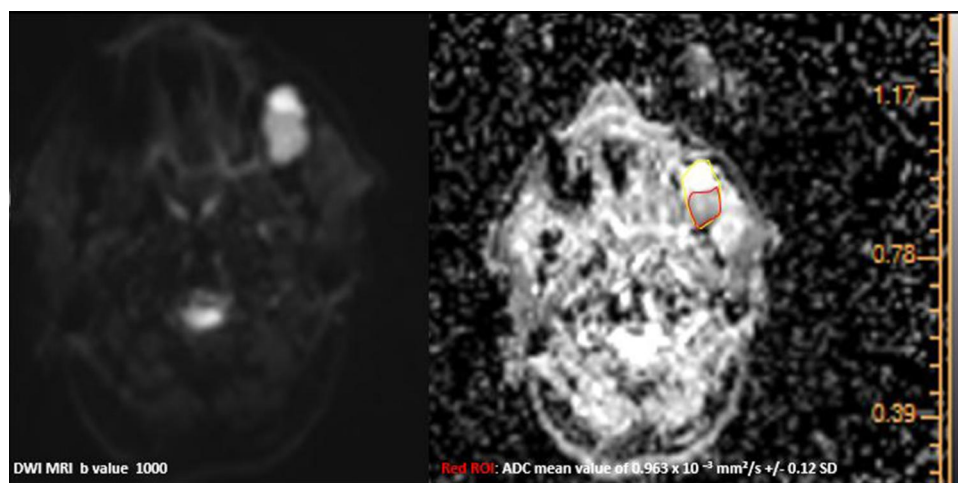


Figure 3 DW-MRI findings of our representative case of a 54-year-old man with MT (yellow circle) in left buccal space. DWI sequences at 1000 s/mm² showed a non-homogeneous hyperintensity in diffusion weighted sequences with an area of restricted diffusion and low apparent diffusion coefficient (ADC) mean value of 0.963×10^{-3} mm²/s in the corresponding map, Red region of interest (ROI).

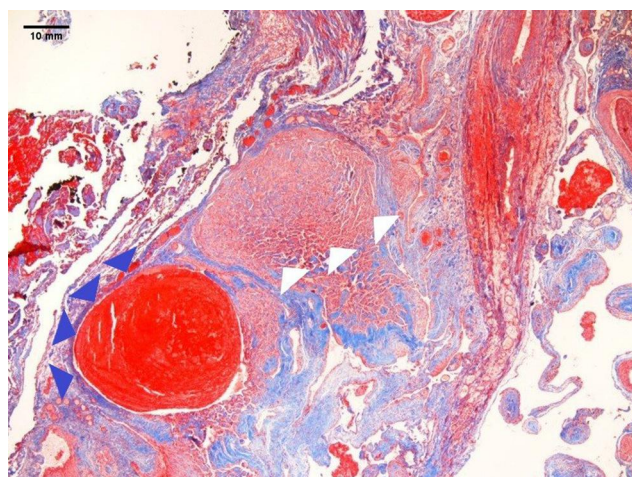


Figure 4 Pathologic specimens (Masson stain 4x) of our representative case of a 54-year-old man with MT in left buccal space. On the pathologic specimen from surgery a mass containing vessel of dilated small calibre (white triangles) was seen in addition to a thrombus (purple triangles) and anastomosing network of little vessels, some fibrous septa and muscular fibers around the lesion.

granuloma and hemangioma and malignant neoplasms such as angiosarcoma and Kaposi's sarcoma.³ The most frequent locations in head and neck were dermis, lip, tongue and buccal mucosa, followed by thyroid, orbit, parotid gland, masseter muscle, nose, sinus, mandible, pharynx and central nervous system.⁷

Histologically, MT is characterized by formation of papillary structures lined by hyperplastic endothelial cells in a thrombosed vascular lumen.³ These pathologic features do not allow to easily differentiate MT from angiosarcoma and other benign and malignant lesions,⁸ like haemangioma,⁹ thrombosed aneurysm,¹⁰ and Kaposi's sarcoma.^{11–13}

MT treatment is less invasive than malignant tumors, such as angiosarcoma that is able to metastasize and may not be fully eradicated by localized surgical removal. Pre-operative radiological diagnosis can be difficult as it has similar CT and MRI appearance to other benign or malignant lesions. Depending on its location, MT should be differentiated from other head or neck soft tissue tumors. In particular, lesions in the orbits should be differentiated from inflammatory pseudotumor, giant cell angiofibroma, rhabdomyosarcoma. Solitary fibrous tumors, sinonasal glomus tumors and sarcomas (fibrosarcoma, rhabdomyosarcoma, malignant fibrous histiosarcoma, leiomyosarcoma) should be included in the differential diagnosis of MT in the sinonasal cavities. MT of the skin should be distinguished from dermatofibroma, dermatofibrosarcoma, while inter- or intra-muscular MT from synovial sarcoma, myositis ossificans, pleomorphic sarcoma, liposarcoma, rhabdomyosarcoma.

In consideration of the rarity of MT, we reported our experience and evaluated image findings through systematic review to identify key features that could help in pre-operative radiological diagnosis.

A comprehensive literature search of English and non-English studies using PubMed was completed up to January 2020 to identify studies evaluating head and neck MT. The following keywords in different combinations were used: “intravascular papillary endothelial hyperplasia” OR “, IPEH” OR “Masson hemangioma” OR “Masson pseudoangiosarcoma” OR “Masson tumor”.

The initial search yielded 1745 hits, whose titles and abstracts were screened for eligibility. We removed 1477

of these because there was not a description of MT cases and we screened 268 studies. Out of these, full text was available in 248 articles, but only 36 were eligible.

There were no original articles focusing on CT and MRI accuracy and technique with valid results. All the articles were case reports or clinical series published between 1981 and 2019 (Table 1). Among the 36 articles, we found 37 cases (14 M, 23 F) of patients with head and neck MT, who underwent CT and/or MRI examination.

The median age was 34.79 (range 0–70). We found 9 MT cases in the head and skull base,^{16,18,31–33,35,40,43} 6 in the orbit,^{3,14,20,26,29,37} 7 cases in paranasal sinuses,^{8,9,17,25,28,44,46} 1 case within the parotid gland,⁴¹ 2 in parapharyngeal space,^{19,22} 2 in larynx and hypopharynx;^{27,39} 4 in buccal space or cheek,^{23,24,34,36} 1 in submandibular space,⁴² 1 in the lip,³⁰ 1 in the mandible,³⁸ 3 in the neck.^{15,21,45}

CT examination was reported in 27 out of 36 articles and in 28 patients, but CT findings were described in 23 articles. MR examination was reported in 23 out of 36 articles and in 23 patients, but MR findings were described in 19 articles. To date, no Diffusion Weighted Imaging (DWI)-MRI findings were described.

Our representative case would be the first one reported with description of CT, MRI and DWI-MRI findings.

Summarizing the descriptions reported, the most frequent CT findings were a well-defined mass with high or soft tissue density, with calcifications in some cases. After contrast medium administration, the enhancement could be homogeneous, non-homogeneous, nodular or peripheral. Bone erosion and lytic lesion were reported in MT of paranasal sinuses and scalp.

The most frequent MRI findings were a well-defined mass with a non-homogeneous signal intensity on T1 and T2 weighted sequences. In particular, MRI showed hypointensity in T1 sequences with foci of hyperintensity (hemorrhage) and hyperintensity on T2 sequences with multiple septations or flow voids (vascular channels). The MRI contrast enhancement is similar to that reported in CT. Bone erosion and fatty infiltration were reported in some cases.

The most frequent CT and MRI findings can mimic angiosarcoma and Kaposi's sarcoma.⁴⁷ The different MRI and CT patterns found in literature could represent the different stages of thrombus organization. In fact, older MT presents growth of endothelial cells, smooth muscle cells, and fibroblasts into the fibrin-rich thrombus. Over time, MT is characterized by formation of capillary channels that may anastomose to create conduits from one end

of the thrombus to the other, reestablishing to some extent the continuity of the original lumen.³ This could explain the presence of high and low flow serpentine vessels seen in the MRI findings reported in the literature and in our case. If the thrombus is recanalized, it becomes a vascularized mass of connective tissue, incorporated in the sub-endothelial layer of the vessel wall. This could explain the presence of homogeneous or peripheral contrast enhancement. In late stage, the thrombus is replaced by a fibrous nodule originating from mesenchymal cell contraction.³

DWI may also play a role in the differentiation of head and neck malignant and benign tumors, because tumoral Apparent Diffusion Coefficient (ADC) values tend to vary depending on the stage of tumor cell differentiation, the degree of tumor cellularity, the presence of necrotic tissue, and the degree of degenerative change in interstitial tissues. Malignant soft tissue tumors usually have low ADC values, which are represented as low signal intensity on ADC maps, whereas benign tumors tend to have higher ADC values.^{48–51}

In particular, it has been shown that the ADC correlates with tumor cellularity in soft tissue sarcomas and it is lower in malignant non myxoid soft tissue tumors than in benign tumors.^{52–54}

In the case presented, MT showed mixed areas of restricted diffusivity, with a mean ADC value of $0.963 \times 10^{-3} \text{ mm}^2/\text{s}$ that overlaps the mean ADC value range $0.41\text{--}1.45 \times 10^{-3} \text{ mm}^2/\text{s}$ of angiosarcoma with solid portions, reported by Sero et al.⁵⁵ The area of non-restricted diffusion could be interpreted as a low cellularity area. This could limit the accuracy of the technique in distinguishing between the two conditions. It is reported that angiosarcomas with predominantly dilated vascular spaces may correspond to low cellularity with no diffusion restriction, and angiosarcomas with predominantly solid portions may correspond to high cellularity with diffusion restriction. Moreover, fibrosis, necrosis and hemorrhage are frequently encountered in angiosarcomas and low ADC values may be induced by hematoma or fibrosis. In our case, histology showed a growth of papillary structures lined by hyperplastic endothelial cells into the fibrin-rich thrombus and capillary channels (Figure 4). Low ADC values may have been induced by hyperplastic endothelial cell proliferation or by the latest stage of fibrin-rich thrombus.⁵⁶ The region with no DWI restriction may correspond to capillary channels.

Histomorphological findings (the lack of necrosis and mitosis and presence of organizing thrombi) in conjunction

Table 1 Articles Included with Demographic Data and CT and MRI Findings

Author	Year	Head and Neck Site	N° of Patients	Age (Sex)	CT Findings	MRI Findings	Shape and Margins
Weber ¹⁴	1981	Orbit	1	20yy (F)	Not described	Not described	Well-shaped
Youngerman ¹⁵	1984	Anterior neck (subcutaneous tissue anterior to thyroid)	1	59yy (F)	Soft tissue	Not performed	Not described
Izukawa ¹⁶	1987	Parieto-occipital	1	55yy (F)	Mixed density	Not performed	Elliptical
Stern ¹⁷	1991	Maxillary sinus	1	17yy (M)	Soft tissue density	Not performed	Not described
Lam ¹⁸	1995	Scalp	1	14yy (M)	Relatively dense with slight contrast enhancement	Not performed	Not described
Moon ⁹	2000	Ethmoid and sphenoid sinuses	1	35yy (F)	Not performed	T1 iso-hyperintense with hyperintense foci (hemorrhage)	Not described
						T2 hyperintense with multiple septations and hypointense peripheral rim	
						Heterogeneous contrast enhancement	
Pantanowitz ¹⁹	2000	Parapharyngeal space	1	26yy (M)	Low density with a peripheral enhancement	Non-homogeneous signal intensity, with mandibular erosion, non-homogenous central enhancement	Well defined
Barras ²⁰	2001	Medial canthus	1	59yy (F)	Not described	Not performed	Not described
Suh ²¹	2003	Neck (soft tissue)	1	12yy (M)	Internal spot-like regions of high density; multiple foci of nodular, peripheral enhancement	Not performed	Ovoid mass
Nishimoto ²²	2004	Parapharyngeal Space	1	66yy (M)	High density, calcifications, heterogeneous enhancement	T1 hypointense	Not described
						T2 heterogeneous/high intensity	
						Heterogeneous contrast enhancement	

Wang ²³	2006	Buccal space	I	55yy (F)	Not performed	Hypointense centrally with a peripheral rim of signal intensity	Well-circumscribed mass
Ucok ²⁴	2007	Face (masseter-buccal space)	I	28yy (F)	Not performed	T1 + T2: non-homogeneous intensity with signal void areas; patchy contrast enhancement	Not described
Hooda ²⁵	2007	Ethmoid sinus	I	45yy (F)	Soft tissue mass	Marked post-contrast enhancement	Not described
Schulze ²⁶	2008	Orbit	I	58yy (F)	Not performed	T1 + T2: non-homogeneous intensity with small coagulations/calcifications	Not described
Güvenç ²⁷	2008	Hypopharynx and larynx	I	18yy (F)	Not performed	Not described	Not described
Wang ²⁸	2009	Left nasal cavity, maxillary sinus, ethmoid sinus and frontal sinus	I	42yy (M)	Not described	No contrast enhancement	Not described
Aggarwal ²⁹	2010	Orbit	I	45yy (F)	Hypodense, with two central hyperdense masses	Not performed	Ill defined
Yonezawa ³⁰	2010	Lower lip	I	62yy (F)	Not performed	T1: marginal enhanced line with no signal intensity at the center of the lesion T2: high signal intensity with no signal intensity inside two low signal intensity dots	Not described
Moriyama ³¹	2011	Forehead (aneurysm of the superficial temporal artery)	I	67yy (F)	Homogeneous contrast enhancement with a thrombus	Not performed	Not described
Park ³²	2012	Frontal scalp	I	10yy (F)	Soft tissue density	T1 isointense T2 heterogeneous/high intensity Heterogeneous contrast enhancement	Well defined
Lee ³³	2012	Left frontal bone	I	14yy (M)	Osteolytic bone	Dilated or prominent vascular structures in bone	Not described

(Continued)

Table 1 (Continued).

Author	Year	Head and Neck Site	N° of Patients	Age (Sex)	CT Findings	MRI Findings	Shape and Margins
Narwal ³⁴	2013	Buccal mucosa	1	19yy (F)	Soft tissue density	Not performed	Not described
Miller ³⁵	2013	Clivus (petrous apex)	2	56yy (F)	Bone erosion, peripheral calcifications	T1: hypointense	Lobulated
				39yy (F)		T2: hyperintense	
						Homogenous contrast enhancement	
Guledgud ³⁶	2014	Cheek (anterior to the right masseter muscle)	1	40yy (F)	Iso to hyperdense, with hyperdense calcific foci	T1 + T2: isointense with hypointense foci and fatty infiltrations	Round to oval mass, well defined
Nguyen ³⁷	2014	Orbit	1	27yy (F)	Not described	Not described	Not described
Xu ³⁸	2014	Mandible	1	14yy (M)	Low density inside the lesion (CT value 43–50HU), no central contrast enhancement with peripheral slight enhancement	Not performed	Not described
Tedla ³⁹	2014	Larynx	1	48yy (M)	Density of 30–45 HU, with vascular areas and vessels	Not described	Polypoid mass, well-defined
Shah ⁴⁰	2014	Scalp	1	3mm (M)	Not performed	T1: isointense	Not described
						T2: heterogeneous hyperintensity with multiple flow voids (vascular channels)	
						Intense contrast enhancement	
Sim ⁸	2015	Ethmoid and sphenoid sinuses	1	11yy (M)	High density	T1: isointense	Round mass, well-defined margins
						T2: mixed high intensity	
Mignogna ⁴¹	2016	Parotid gland	1	70yy (F)	Not performed	T1: isointense	Irregular mass, smooth margins
						T2: heterogeneous, high intensity	
						Homogeneous contrast enhancement	

Kim ⁴²	2017	Submandibular space	I	18yy (F)	Low density, vascular enhancement after contrast administration	Not performed	Not described
Gupta ⁴³	2017	Scalp	I	13yy (F)	Hyperdense lesion with irregular bone cortical erosion	T1: hypointense	Oval mass, well demarcated
						T2: heterogeneous hyperintensity	
D'Aguanno ⁴⁴	2018	Maxillary sinus	I	67yy (F)	High density, bone erosion	T1: hypointense with high intensity spot	Not described
						T2: heterogeneous hyperintensity	
						Irregular contrast enhancement, bone erosion	
Alotaibi ⁴⁵	2019	Right lateral neck	I	56yy (M)	Cystic mass with peripheral vessels	Not performed	Well-circumscribed
Cooke ⁴⁶	2019	Maxillary sinus	I	28yy (M)	Bone erosion	Not performed	Not described
Dryden ²	2019	Orbit	I	63yy (M)	Not performed	Enhancing mass	Not described

with immunohistochemical staining (CD31 and CD34) of the entire lesion can differentiate MT from angiosarcoma and other malignant head and neck soft tissue tumors.

Currently, no standard treatment of MT had been established, and localized lesion has been usually treated with radical resection. Radical excision was performed in our case and the prognosis was good.

According to literature data and our experience, a pre-operative identikit of MT based only on imaging is not achievable and surgical removal is necessary to accurately differentiate it from malignant angiosarcoma.

Despite this, pre-operative radiological evaluation is useful in surgical planning, suggesting a low aggressive behavior more compatible with a benign tumor like MT or low grade angiosarcoma than malignant lesion, preventing unnecessary aggressive surgical approach.

In summary, the present case and literature review suggest that MT may show following imaging features: (1) high or soft tissue density in CT examination, with calcifications in some cases; (2) hypo-isointensity in T1 sequences with foci of hyperintensity (hemorrhage) and hyperintensity on T2 sequences with multiple septations or flow voids in MRI examination; (3) mixed presence areas of restricted diffusivity in DW-MRI; (4) homogeneous, non-homogeneous, nodular or peripheral contrast enhancement; 5) bone erosion and lytic lesion in some cases.

We hope that our report provide further information for the diagnosis, classification and treatment of the disease in the future.

Abbreviations

CT, computed tomography; MRI, magnetic resonance imaging; DWI, diffusion weighted imaging.

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Salvatore Politi and Luca Balzarini share senior authorship.

Disclosure

The authors report no conflicts of interest in this work.

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