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CASE REPORT

Imaging features of myoepithelial carcinoma of the mandible with lymph node metastasis

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Abstract
Objectives To demonstrate the imaging characteristics of a patient with myoepithelial carcinoma (MEC) of the mandible accompanied by submandibular lymph node metastases, and to discuss the differential image-based diagnoses.
Patients and results The patient was a 57-year-old woman who had suffered from a dull pain in her left lower molar region for 3 months and had an elastic-soft mass in her submandibular region for 2 months. Computed tomography (CT) images showed permeative destruction of the bone trabecula and intermittent absorption of the cortical plates in the left mandible. The bone marrow of this area showed low signal intensity in a T1-weighted magnetic resonance (MR) image, slightly low signal intensity in a T2-weighted image, and marked contrast enhancement. A tumor was confirmed outside the buccal and lingual cortical plates. The left submandibular mass was shown as a well-defined, water-density mass by CT, low signal intensity in the T1-weighted MR image, and markedly high signal intensity in the T2-weighted image. The histopathological diagnosis was MEC of the mandible with submandibular lymph node metastasis.
Conclusions We demonstrated the imaging characteristics of MEC, showing permeative destruction of the bone trabecula, intermittent absorption of the mandible, and cystic degeneration of the metastatic cervical lymph node.

Keywords Myoepithelial carcinoma · Jaw · Lymph node metastasis · MRI

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Introduction

Myoepithelial carcinoma (MEC) is an uncommon tumor, accounting for fewer than 1% of all salivary gland neoplasms and fewer than 2% of all salivary gland carcinomas [1–5]. MEC occurs predominantly in the parotid gland (70–80%) but can arise in the submandibular and minor salivary glands [1–3]; few cases have been reported in the jaw [6, 7]. The mean age of patients is 55 years with a broad age distribution [1–3]. MEC occurs equally in males and females [1], sometimes with slight predominance in females [2, 3]. The prognosis of MEC is generally good, but the risk of local recurrence is high (about one-third) [1, 2, 8–11]. Cervical lymph node and distant metastasis have been reported in 20 and 10% of cases, respectively [10].

Few studies have examined the image features of MEC [3, 5–9, 12–23], with most reports presenting well-defined, sometimes poorly-defined, and heterogeneous enhanced tumors in computed tomography (CT) and magnetic resonance (MR) images. In tumors involving the bone, bone destruction is often observed [5, 6, 15]. These images of MEC were nonspecific, as in most low-grade malignant tumors. The imaging features of metastasis of the cervical lymph nodes have been described in only two reports [6, 10].

In this report we present the image characteristics for a patient with MEC of the mandible accompanied by

submandibular lymph node metastases and discuss the differential image diagnoses.

Case report

A 57-year-old woman suffered from a dull pain in her left lower molar region and underwent extraction of the left lower first and second molars at a dental clinic 3 months ago. Then, she experienced swelling and continuous dull pain in the same region. She became conscious of a mass in the left submandibular region 2 months ago. She received medication with antibiotics, and curettage of the extracted sockets was performed twice. The pain in her lower molar region continued, so she was referred to our hospital. The bone of the extracted sockets was exposed, and the surrounding gingiva showed necrosis. She had a 12 × 12 mm, elastic-soft, tender, and fixed mass in the submandibular region. She did not have paralysis of the lower lip. A panoramic radiogram showed the remains of the extracted socket and no marked absorption of the bone trabecula in the left lower molar region (Fig. 1). A biochemistry test showed high levels of alkaline phosphatase, γ-GPT, GOP, and GPT and normal levels of C-reactive protein and white blood cells.

She received CT, MR, and sonographic examinations of the maxillofacial and neck region because the submandibular mass had increased in size rapidly. The CT showed that the bone trabecula of the left mandible had been permeatively destroyed in a downward manner to an area beyond the mandibular canal (Fig. 2). The buccal and lingual cortical plates presented intermittent absorption. A T1-weighted MR image revealed a decrease in the signal intensity of the bone marrow in the left lower premolar and molar regions, except for the region in which gauze had

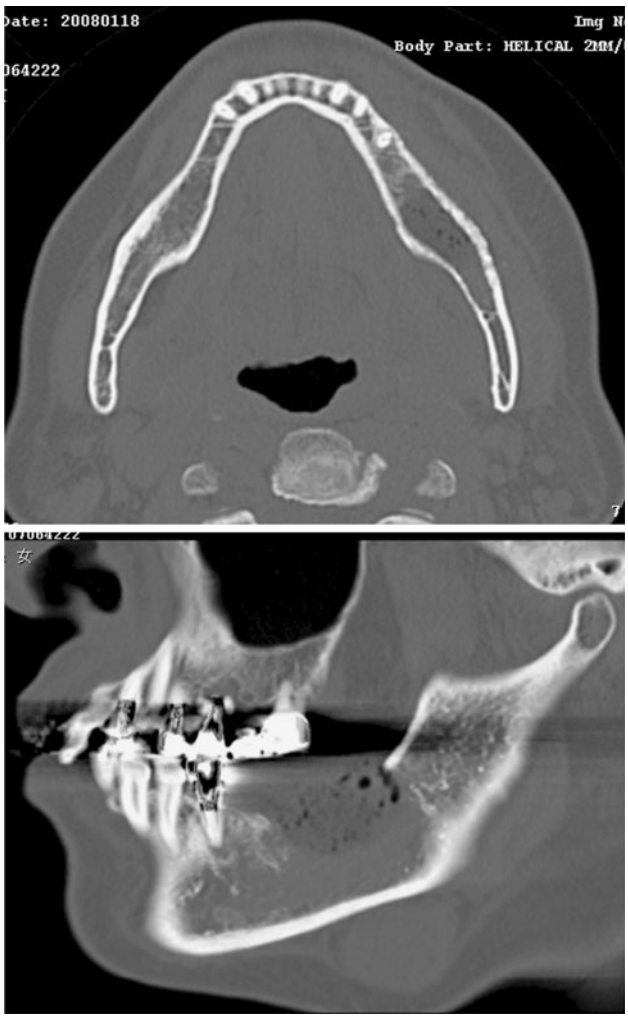

Fig. 2 CT image showing permeative destruction of the bone trabecula of the left mandible beyond the mandibular canal. The buccal and lingual cortical plates presented with intermittent absorption

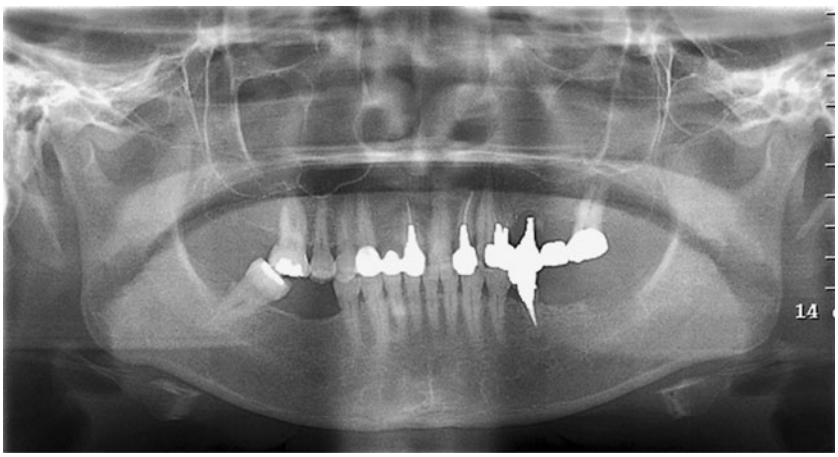

Fig. 1 Panoramic radiogram showing the remains of the extracted socket and no marked absorption of the bone trabecula in the left lower molar region

been packed into the previously curetted area (Fig. 3a). A T2-weighted image showed a slightly decreased intensity, and a T1-weighted image after contrast medium administration showed marked enhancement in the same region (Fig. 3b, c). The periosteum was elevated, and a tumor was confirmed outside the buccal and lingual cortical plates.

The left submandibular tumor was a 30 × 30 mm, round, well-defined, water-density (14 H.U. of CT number) mass with a peripheral soft-tissue-density thin rim in CT images (Fig. 4a). A color Doppler sonogram showed an ill-defined heterogeneous mass (Fig. 4b). The mass consisted of a central anechoic area with a septum structure and a peripheral thick echogenic area. Posterior echo enhancement was observed. No internal color signal was seen, and the peripheral color signal was partially observed. The mass displayed low signal intensity in the T1-weighted

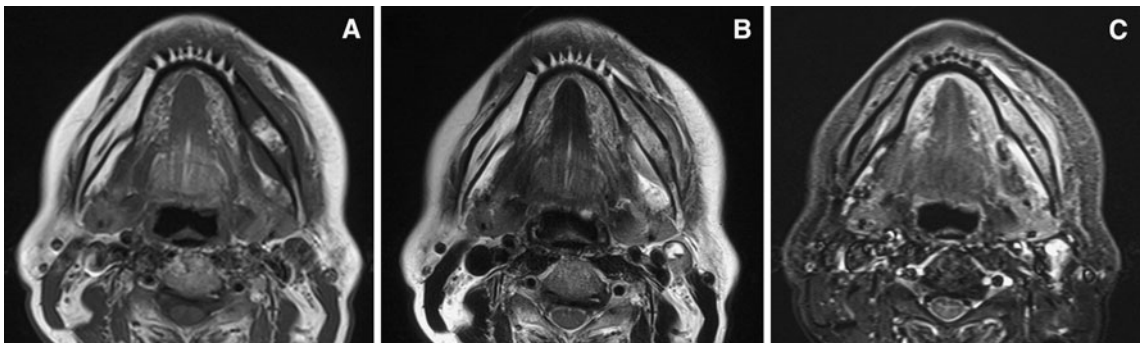

Fig. 3 MR images of the mandible. **a** T1-weighted MR image showing a decrease in the signal intensity of the bone marrow in the left lower premolar and molar regions, except for the region in which gauze had been packed into the previously curetted area. **b** T2-weighted image showing a slight decrease in the same region. **c** T1-weighted image after contrast medium administration showing marked enhancement in the same region. The periosteum was elevated, and a tumor was confirmed outside the buccal and lingual cortical plates

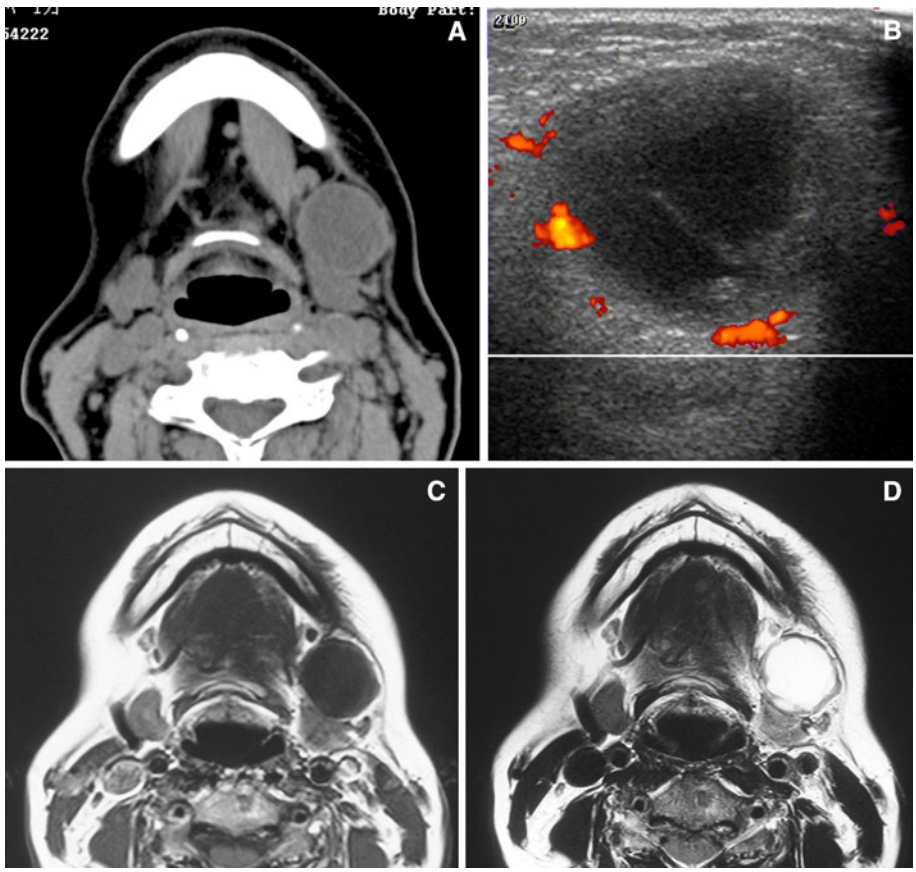

Fig. 4 CT, sonographic, and MR images of the left submandibular mass. **a** CT image showing a 30 × 30 mm, round, well-defined, water-density (14 H.U. of CT number) mass with a peripheral soft-tissue-density thin rim. The submandibular gland was deviated posteriorly by this mass. **b** Color Doppler sonogram showing an ill-defined heterogeneous mass. **c** T1-weighted MR image showing low signal intensity. **d** T2-weighted MR image showing markedly high signal intensity. A low-intensity capsule was observed around the mass

image, and a markedly high signal intensity in the T2-weighted image (Fig. 4c, d). A low-intensity capsule was observed around the tumor.

The patient received curettage of the molar region of the mandible and resection of the submandibular tumor. The specimens from the buccal cortical plate of the mandible and from the buccal mass outside the mandible revealed proliferation of the spindle-shaped and round-shaped tumor cells in the solid form. The nucleus was variable in size, and demonstrated high mitotic activity and nuclear polymorphism. The specimen from the trabeculae around the mandibular canal showed invasion of the tumor cells into the bone marrow. The submandibular mass was separated easily from the submandibular gland. The mass showed cystic degeneration. In the peripheral area of the mass, the spindle-shaped and round-shaped tumor cells formed solid or restiform patterns. The nucleus was variable in size, and demonstrated high mitotic activity and nuclear polymorphism (Fig. 5a). Focally, the tumor infiltrated the surrounding connective tissues. The immunohistochemical findings were positive for cytokeratin (AE1/AE3, Ck13, Ck17), smooth muscle actin (Fig. 5b), Vimentin, D2-40, p63, Ki-67, and proliferation cell nuclear antigen. The final diagnosis of our patient was a myoepithelial carcinoma of the mandible with metastasis of the submandibular lymph node.

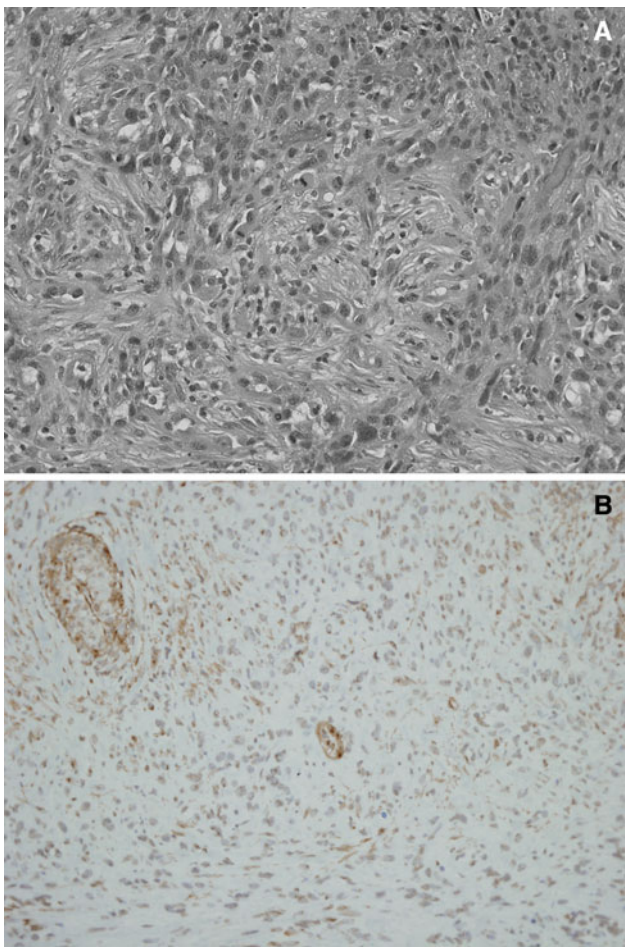

Fig. 5 Histopathological features. **a** HE (×272). Hyaline and spindle-shaped myoepithelial cells demonstrated prominent mitotic activity and abundant eosinophilic cytoplasm. **b** Smooth muscle actin showing positivity

Discussion

MEC is an uncommon tumor, with very few cases reported in the jaw [6, 7]. The patients may have no symptoms or complain of a long-standing mass without pain. The occurrence of pain or nerve paralysis is rare [1, 20, 24]. The patient in our study complained only of a dull pain in the mandibular molar regions for 3 months or more and she had no nerve palsy. Her clinical features seemed to suggest an inflammatory lesion, such as poor healing of the extracted sockets. Only the high-level of alkaline phosphatase suggested a pathology involving bone remodeling.

Few studies have reported the imaging features of MEC [3, 5–9, 12–23]. A summary of the imaging features of MEC in the maxillofacial region is provided in Table 1. MEC is unencapsulated, but may be well-defined with a nodular surface [1]. Among the imaging features of the border or margin of the tumor, some reports have revealed smooth and well-defined tumors without invasion of the adjacent fat planes, suggesting benign tumors [3, 8, 9, 13, 19], whereas others have reported poorly circumscribed lobular tumors [12, 17, 21]. The description of the tumor interface has been reported to differ depending on imaging modality: MRI displays the interface more clearly than CT [8]. Therefore, the method used for evaluation is very important.

MEC displays uniform low or intermediate signal intensity in T1-weighted MR images and homogeneous moderately high or slightly nonhomogeneous high signal intensity in T2-weighted images [8, 9, 13, 14, 21, 22]. The high signal intensity in T2-weighted images is consistent with low-grade malignant salivary gland tumors, although it is not specific to MEC; high-grade salivary gland malignant tumors are reported to show a low signal intensity in T2-weighted images [9, 13]. The tumor is enhanced by contrast media with various patterns, either slightly, well, or heterogeneously [3, 8, 9, 13, 17, 18, 21]. Contrast enhancement suggested a physiologically rich blood supply for the tumor, or highly vascularized characteristics of MEC [9]. These image findings might be very important for malignant tumors, although relatively high vascularized histopathological characteristics of MEC have not been reported [9].

Originally, MEC has the character of local destruction [1, 12, 23], and destruction of the surrounding bone has

Table 1 Summary of image characteristics of myoepithelial carcinomas in the maxillofacial regions

Authors	Published year	Gender	Age (years)	Primary site	Size (mm)	Modalities	Image features						Metastasis	
							Border	MR intensity		CT density	Enhancement	Bone destruction	LN	Distant
								T1 WI	T2 WI					
Kusafuka et al.	2008	Man	70	Parotid gland	68 × 47 × 70	MRI/CT	Ill-circumscribed, lobulated							
Piscioli et al.	2007	Woman	81	Parotid gland	22	MRI/CT	Well-defined	Low	Homo-geneous high		Slight			
Silvers et al.	1996	Man	64	Parotid gland	40	MRI/CT	Smooth	Inter-mediate	High		Hetro-geneous			
Yano et al.	2005	Woman	62	Parotid gland	10 × 15	MRI			High					
Asai et al.	1995	Woman	1.5	Parotid gland	30 × 20	PA/ oblique						Sun-ray spiculae/bone destruction		
Moriniere et al.	2003	Woman	8	Parotid gland (×2 mass)	15, 17	MRI/CT	Irregular							
Amin et al.	2002	Woman	76	Parotid gland (×2 mass)	40, 20	MRI	Poorly circumscribed				Hetro-geneous	Intracranial invasion/ bone erosion		
Yamada et al.	2007	Man	72	Submandibular gland	20 × 15	MRI/CT	Irregular shaped	Low	Abnormal	Low	Hetro-geneous			
Zim et al.	2006	Woman	12	Floor of mouth	30 × 40	MRI	Well-circumscribed				Enhanced			
Kumai et al.	2006	Man	76	Base of tongue	40 × 40 × 50	MRI	Irregular				Enhanced			
Li et al.	2000	Woman	72	Hard palate	20 × 15	CT	Lobulated					No bone resorption		
Hagiwara et al.	1995	Woman	69	Hard palate	10	Occusal						No bone destruction		
Jain et al.	2006	Woman	25	Buccal mucosa	12 × 10	CT						Bone erosion	Multiple	
Imate et al.	2000	Woman	68	Parapharyngeal space	20	MRI	Sharply defined							
Yamanegi et al.	2008	Woman	70	Nasal cavity		CT						Bone destruction		
Jung et al.	2007	Man	32	Lacrimal gland		CT					Well-enhanced			
Takayama et al.	2006	Man	29	Floor of mouth[a]	30	MRI/CT	Smooth	Low	High					
Karatzanis et al.	2005	Woman	70	Soft palate[a]	46 × 60	MRI			Necrosis					

[a] Recurrent tumor

often been observed [5, 6, 17]. Furthermore, Asai et al. reported the formation of sunray spiculae of the mandible [15]. Panoramic radiography in our case did not reveal definite bone destruction. However, CT and MR images revealed intermittent resorption of the buccal and lingual cortical bone, maintaining the outward form of the mandible. Such resorption may not be evidenced by panoramic radiography. The bone destruction had spread downward beyond the mandibular canal. This intermittent resorption of the cortical bone is also observed in adenoid cystic carcinoma and actinomycosis [25, 26]. It might be a characteristic finding of the tumor or inflammation that invades the medullary cavity of the bone widely, which is different from squamous cell carcinoma. Furthermore, the MR image showed permeative invasion of the bone marrow space and mass formation inside and outside the mandible. These might be typical features of malignancies, for example adenoid cystic carcinoma, malignant lymphoma, osteosarcoma, and other sarcomas [25, 26].

Submandibular masses may be inflammatory, neoplastic, or developmental lesions originating from the major structures of the submandibular space, for example lymph nodes or the submandibular salivary gland [27]. Cystic lymph nodes present as inflammatory reactions or as neoplastic conditions such as metastatic diseases [28–30]. Among salivary gland neoplasms, prominent cystic changes may occur in pleomorphic adenoma [31], Warthin tumor [32], mucoepidermoid carcinoma [33], or acinic cell carcinoma [34]. A cystic mass in the submandibular region must be differentiated from other nonneoplastic cystic swellings, such as submandibular branchial cyst [35], dermoid/epidermoid cyst [36], mucocele [27, 37], rare cases of hydatid cyst [38], cystic hygroma [39], and salivary duct cyst [40]. Areas of necrosis and cystic degeneration have been found in some MECs [1, 23]. Yamada et al. reported that examination of MEC of the submandibular gland by CT showed a low-density area within the tumor [9]. MEC involves cervical lymph node metastases in approximately 20% of cases [10, 12]. Although their imaging features have not been reported, such cystic masses might be compatible with metastatic lymph nodes of MEC.

MEC is composed almost exclusively of myoepithelial differentiated cells surrounding duct-forming cells [1, 5]. Immunohistochemical examination with specific myoepithelial markers is helpful for diagnosis; i.e., diagnosis of MEC is established by cytokeratin activity and at least one of the other myoepithelial markers, including smooth muscle actin, glial fibrillary acidic protein, CD-10, calponin, and smooth muscle myosin heavy chain [1, 2, 11, 16, 18, 19, 23]. Our patient was positive for cytokeratin, smooth muscle actin, and Vimentin, and the diagnosis was compatible with MEC.

The prognosis for MEC is generally good, but the risk of local recurrence is high, at about one-third [1, 2, 8–11]. Death due to the disease is relatively rare, with a 10-year disease-free survival of 81.8% [1, 2, 4, 19]. Low-grade malignant salivary gland tumors are sometimes misdiagnosed as benign tumors preoperatively. Such a situation leads to selection of an inappropriate surgical technique, outcomes of frequent local recurrence, and poor prognosis [9].

In conclusion, we presented the imaging characteristics of MEC of the mandible with cervical lymph node metastasis. A cyst-like appearance of lymph node is rare. When radiologists encounter such an appearance, they should consider cervical lymph node metastasis of MEC as one of the differential diagnoses.

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