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ABOUT COVER

Editorial Board Member of *World Journal of Clinical Cases*, Dr. Iva Brčić finished medical studies at the Medical University of Graz and received her MD degree in 2003. She received her doctoral degree in 2006 at the same institution. In 2007, she enrolled in the pathology residency program at the University Hospital Center Zagreb. In 2012, she passed her board exam and, until 2015, worked as a staff pathologist at the University Hospital Center Zagreb. From 2015, she is working as the University Assistant at the Medical University of Graz. At the end of 2017, she joined the bone and soft tissue team and spent 4-mo observership at the University of Miami, FL, USA. Her ongoing research interests include bone and soft tissue neoplasms.

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CASE REPORT

Mucoepidermoid carcinoma in the infratemporal fossa: A case report

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Abstract

BACKGROUND

Mucoepidermoid carcinoma is the most common primary epithelial salivary gland malignancy. It mostly occurs in the major or intraoral minor salivary glands but rarely in the infratemporal fossa. Here, we present a case of aggressive mucoepidermoid carcinoma in the infratemporal fossa with neck lymph node metastasis and also discuss diagnostic and treatment strategies.

CASE SUMMARY

A 39-year-old woman with a mass located in the right submandibular area presented to our department. Physical examination revealed lymphadenopathy on the right submandibular side measuring 2.5 cm × 3 cm that was hard and had poor mobility. Results of nasal endoscopy were unremarkable. Ultrasound examination revealed an enlarged lymph node at level II of the right side. Fine needle aspiration cytology of the metastatic lymph node revealed malignant cells with infection. Contrast-enhanced computed tomography revealed an enhancing ill-defined soft tissue mass in the right infratemporal region. Positron emission tomography/computed tomography revealed hyperintensity in the right infratemporal fossa along with lymphadenopathy at level II of the right-side lymph node. The patient underwent extended resection of the primary tumor, and ipsilateral radical neck dissection was also completed. Hematoxylin-eosin staining and immunohistochemistry revealed a high-grade mucoepidermoid carcinoma. No signs and symptoms of recurrence of the neoplasm were present after 20 mo of follow-up.

CONCLUSION

Positron emission tomography/computed tomography play a key role in primary tumor localization. Furthermore, histopathology and immunohistochemistry play pivotal roles in disease diagnosis.

Key words: Infratemporal fossa; Mucoepidermoid carcinoma; Metastasis; Positron emission tomography/computed tomography; High-grade; Case report

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Core tip: Here, we report a patient with mucoepidermoid carcinoma in the infratemporal fossa. Because of the particular location and atypical oral presentation, we also review corresponding literature to discuss the diagnostic and treatment strategies for mucoepidermoid carcinoma.

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INTRODUCTION

Mucoepidermoid carcinoma (MEC) is one of the most common malignant salivary gland tumors and usually affects the parotid and minor salivary glands[1]. MEC accounts for < 5% of all head and neck malignancies[2]. Histologically, MEC comprises a variable percentage of epidermoid, mucous, and intermediate cells. According to the proportion of the three types of cells and cell differentiation degree, MEC has been classified as low-, intermediate-, and high-grade. High-grade tumors are highly aggressive, and regional lymph node spread is common. Furthermore, the low-grade variant is defined by a lack of aggressive invasion pattern and usually has a more benign nature.

MEC was first reported in 1945 by Stewart *et al*[3]. MECs also occur in other organs such as the larynx, mandible, breast, and thymus[4-7], and they tend to occur in the fourth and fifth decades of life and have a female preponderance[8,9]. MEC typically presents as a major salivary gland or intraoral mass. However, the effect of tumors in the infratemporal region on the oral cavity was not obvious. Comprehensive head and neck examinations and advanced imaging techniques such as positron emission tomography/computed tomography (CT) can facilitate the identification of the primary site. Thus, we present an unusual case of high-grade MEC in the infratemporal fossa of a woman and also discuss diagnostic and treatment strategies for MEC.

CASE PRESENTATION

Chief complaints

A 39-year-old woman presented with a 3-mo history of a mass located in the right submandibular area.

History of present illness

The mass in the right submandibular lymph node had rapidly grown over the past 2 wk. Furthermore, it was accompanied by radiating pain in the right maxillary sinus region. The symptoms of the patient were not relieved despite taking anti-inflammatory drugs for 10 d. The results of previous examinations were as follows: Ultrasound examination revealed an enlarged lymph node at level II of the right-side lymph node; nasal endoscopy results were unremarkable; and fine needle aspiration cytology of the metastatic lymph node demonstrated malignant cells with infection but with no obvious structure to determine the tissue source.

History of past illness

She had a history of chronic hepatitis for 12 years.

Personal and family history

The patient's medical history and family history were unremarkable.

Physical examination

Physical examination revealed a 2.5 cm × 3 cm-sized lymphadenopathy of the right

submandibular lymph nodes, and the mass was tender, hard, and had poor mobility.

Laboratory examinations

Laboratory tests showed the following results: Urine analysis of blood: BLD (+-); Percentage of monocytes: 10.8%; liver and kidney function normal; hepatitis B surface antigen and hepatitis B core antibody were positive.

Imaging examinations

Contrast-enhanced CT revealed an enhancing ill-defined soft tissue mass in the right infratemporal region (Figure 1). Positron emission tomography/CT revealed an increased uptake of fluorodeoxyglucose in the right infratemporal fossa and at level II of the right-side lymph node (Figure 2).

Postoperative pathology tests

Hematoxylin-eosin staining of the specimen revealed that the tumor majorly comprised two types of lesional cells: Epidermoid and intermediate cells (Figure 3). In the tumor stroma, nests of squamous cell infiltration, cell heteromorphism, and mitotic figures were obvious, and focal necrosis was also observed, which was indicative of a high-grade MEC. Immunohistochemistry was performed for further pathological diagnosis, and the result revealed cytokeratin 7 (+), cytokeratin 5/6 (partial, +), cytokeratin 18 (partial, +), P40 (partial, +), and negative for estrogen and androgen receptors and human epidermal growth factor receptor 2/neu protein (Figure 4).

FINAL DIAGNOSIS

Based on radiographic results and histopathology, we finally confirmed the diagnosis of a high-grade MEC with lymph node metastasis. The final clinicopathologic findings were those of a high stage (T2N2M0).

TREATMENT

Under general anesthesia, the primary tumor was extensively resected *via* approach from the infratemporal fossa, and then ipsilateral radical neck dissection, facial nerve dissection, and arbitrary flap formation were performed. The tumor was completely resected. Intraoperative frozen pathology suggested that the tumor originated from the epithelium. A drainage tube was placed in the mouth and neck area. No apparent surgical complications occurred after surgery, and the patient was discharged 15 d after surgery.

OUTCOME AND FOLLOW-UP

The patient underwent radiotherapy and regular follow up. There were no signs and symptoms of recurrence of neoplasm from the past 20 mo since the surgery.

DISCUSSION

The infratemporal fossa is an irregular space in the skull base, with the anterior boundary on the posterior surface of the maxilla, posteriorly by deep lobe of the parotid gland, laterally by ascending ramus of mandible and descending lamina sphenoid bone, and superiorly by external rhytidectomy infratemporal surface of greater wing of sphenoid and squamous part of temporal bone. Common primary tumors in the infratemporal fossa are fibrosarcoma, hemangioma, pleomorphic adenoma from ectopic salivary tissue, or neurogenic tumors[10]. The incidence of MEC in this location is extremely rare.

MEC accounts for approximately 30% of all salivary gland malignancies, and it is the most common malignant tumor of the parotid gland[11,12]. The histologic grade of MEC has prognostic value and directs adjuvant therapy[13]. The grade of MEC is determined based on the relative proportion of three types of cells and grades of differentiation. The low-grade type is characterized by > 50% mucinous cells and

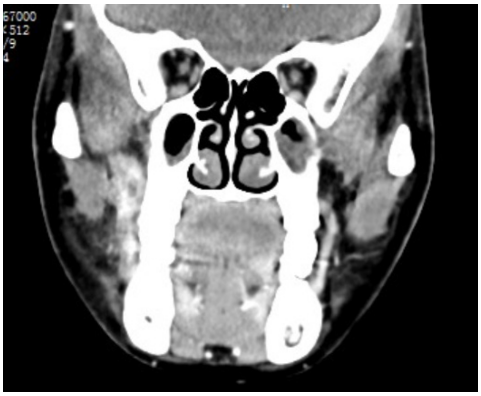

Figure 1 Computed tomography scan (axial section) showing an enhanced soft tissue mass in the right infratemporal fossa.

epidermoid cells, whereas the high-grade type is characterized by a predominance of epidermoid and intermediate cells with < 10% mucinous cells[14]. Intermediate-grade type has characteristics that are between the above two types. Because of the existence of epidermoid cells, MEC is often confused with squamous cell carcinoma, and mucicarmine staining is used to differentiate between these two types of tumors.

Intermediate- and high-grade tumors are associated with high potential risks of metastasis. Neck node metastases usually indicate a worse prognosis[15]. In this case, fine needle aspiration cytology from the neck node determined the nature of the malignancy. Localization of the primary site and accurate pathological diagnosis are particularly important for treating patients with MEC. However, because of the multiple structures that are present within the infratemporal fossa and concealed location, early diagnosis is difficult owing to the lack of atypical symptoms. Furthermore, the diagnosis of a tumor in the infratemporal fossa can be complicated by similar clinical features such as trigeminal neuralgia and temporomandibular arthropathy. In our case, because of atypical oral manifestations, it was necessary to perform a complete oncologic workup to exclude the possibility of secondary metastasis before treating the lesion as MEC in the infratemporal fossa. Positron emission tomography/CT helped determine the location of the primary tumor, and hematoxylin-eosin staining and immunohistochemical analysis confirmed the final diagnosis.

MEC is a malignancy in which histological grading and clinical behavior correlate well[16]. Ozawa *et al*[17] analyzed 43 patients with head and neck MECs and concluded that T and N stages are significant prognostic factors for MECs. Treatment is largely based on histological tumor grading, and surgical resection is the main treatment for all grades of MEC. Neck dissection is indicated when clinical evidence of regional metastasis, high TNM stage, or high histological grade is noted[18,19]. Moreover, surgical tumor resection is considered sufficient treatment for low-grade MEC. High-grade tumors are generally treated with surgical excision with wide margins or neck dissection followed by postoperative radiotherapy[13]. Furthermore, Wu *et al*[20] reported that postoperative adjuvant ^{125}I seed brachytherapy appears to be an effective and safe treatment option for MEC of the parotid gland with a clinically node-negative neck, especially when the tumor is of low or intermediate grade.

Low-grade tumors have a more benign course with a 5-year overall survival rate of approximately 90%. In contrast, high-grade tumors are much more likely to recur and have a 5-year overall survival rate of approximately 50%[21]. In our case, the patient developed neck lymph node metastasis before surgery. At 6 wk after surgery, the patient received 30 courses of radiotherapy with a total radiation dose of 60 Gy.

CONCLUSION

The incidence of MEC in the infratemporal fossa is extremely low, and the clinical symptoms are atypical. Our case findings emphasize the importance of oncologic workup to determine the primary tumor location and ensure accurate histopathology and postoperative radiotherapy. For such rare tumor sites, it is important for oral and maxillofacial surgeons to review the clinical presentation, histology, and management of MECs.

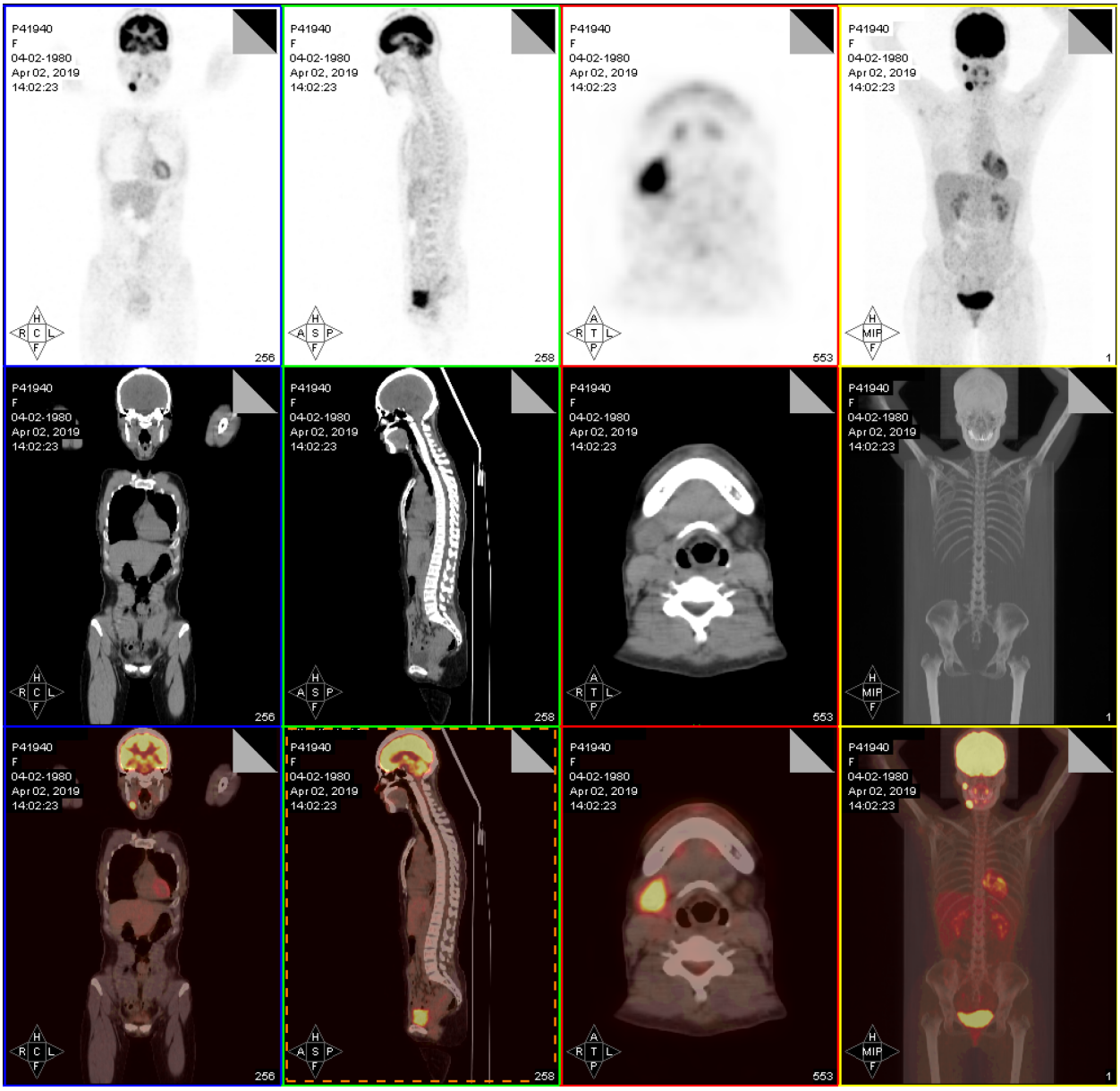

Figure 2 Representative fusion positron emission tomography/computed tomography images demonstrate intense uptake in the right infratemporal fossa and the lymph node of right II.

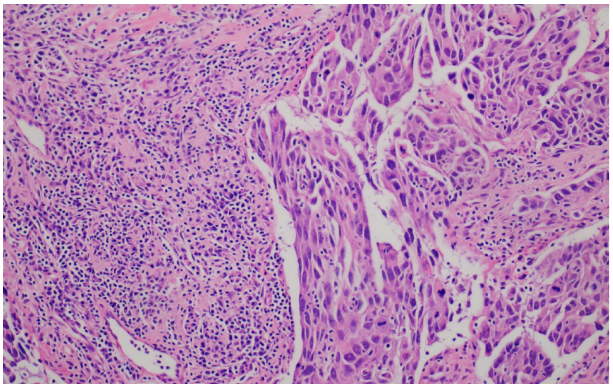

Figure 3 Hematoxylin-eosin staining of mucoepidermoid carcinoma. High grade mucoepidermoid carcinoma: Infiltration and solid growth pattern (magnification: 200 ×).

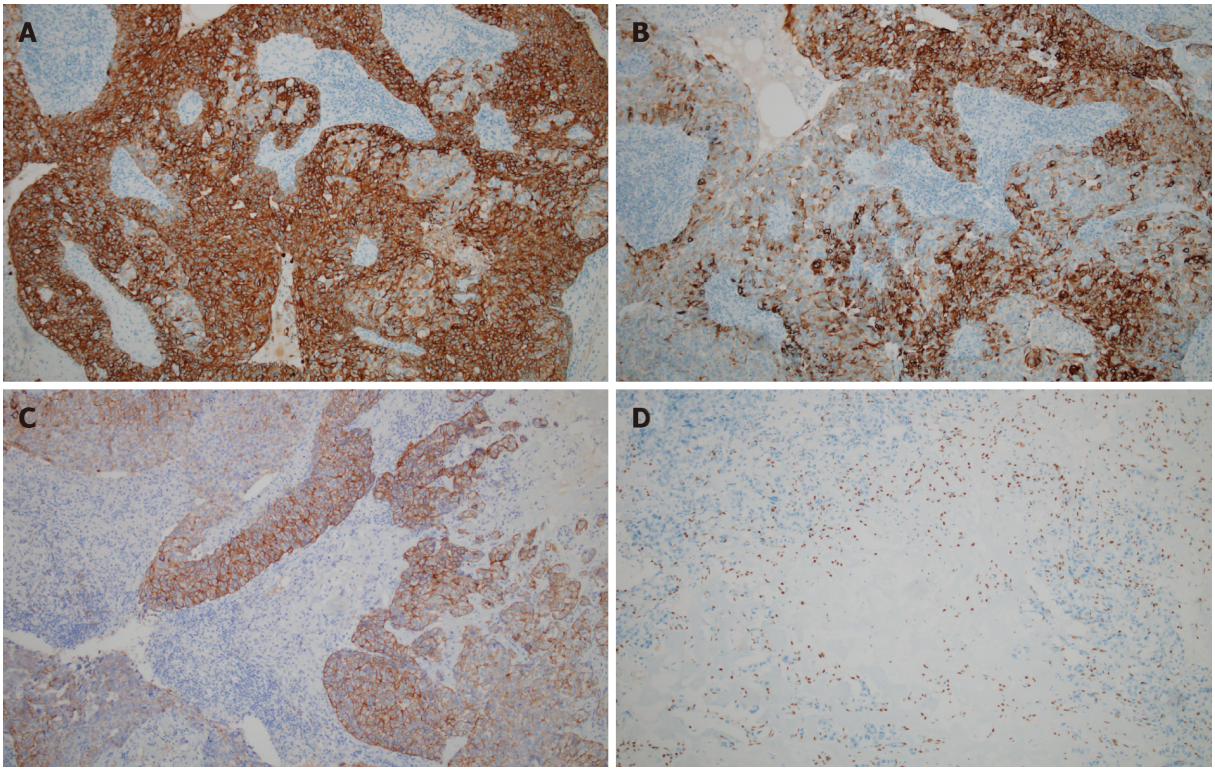

Figure 4 Immunophenotypic features of mucoepidermoid carcinoma. A: Cytokeratin 7 showing diffusely positive staining; B: Showing positive staining for cytokeratin 5/6; C: Showing a strong positive reaction to cytokeratin 18; D: Focal positive expression of P40 (magnification: 200 ×).

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