Central mucoepidermoid carcinoma of the mandible – A case report

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SUMMARY

Introduction Mucoepidermoid carcinoma, compared to other tumors of salivary glands, occurs in 5–10% of cases. Histopathologically, it is divided into a well differentiated tumor that is of low-grade of malignancy, and a medium and poorly differentiated tumor of high grade of malignancy. Central mucoepidermoid carcinoma (CMEC) of the mandible was firstly described by Lepp in 1936, on a 66-year-old female patient. CMEC is characterized by atypical clinical image and radiological manifestation.

Case Outline A 55-year-old female patient was examined at the Clinic of Dentistry in Niš, Serbia, with anamnestic data regarding the presence of painless swelling in the right side of the mandible. Considering the histopathological results and presence of enlarged lymph nodes, right hemimandibulectomy and tumour excision from pterygomandibular space followed by supraomohyoid neck dissection was done. In due course, postoperative radiotherapy was applied (60 Gy).

Conclusion CMEC represents a rare tumor, characterized by local tissue destruction and ability to metastasize. Initial biopsy represented the key in preoperative planing. Radical excision with neck lymph node dissection followed by postoperative radiotherapy in our case represent a successful method of treating CMEC of the mandible.

Keywords: central carcinoma; mandibular swelling; biopsy; surgical therapy

INTRODUCTION

Mucoepidermoid carcinoma (MEC), compared to other types of tumor of salivary glands, occurs in 5–10% of the cases. It may be present in large salivary glands (86% in parotid gland, 8% in submandibular, and 6% in sublingual gland) [1]. Most frequent intraoral localization of MEC are small salivary glands of the hard palate with 41.1% [2]. Histopathologically, there are three modes of MEC – well differentiated tumor with low-grade of malignancy, medium, and poorly differentiated tumors with high grade of malignancy. By observing intraoral localization, a well differentiated tumor is encountered in 58.4% of the cases, medium differentiated in 38.3%, while poorly differentiated tumor is present in 3.2% of the cases [3].

Central mucoepidermoid carcinoma (CMEC) of the mandible was described in literature for the first time by Lepp [4] in 1936, in a 66-year-old female patient. However, this is a very rare localization of the tumor, which belongs to primary intraosseous carcinomas. Assumptions are that CMEC can originate from ectopic glandular tissue, transformed mucosa cells, existing untreated odontogenic cist or intra-bony propagation of epithelial cells from maxillary sinus and submucous salivary glands [5]. CMEC is characterized by atypical clinical image and radiological manifestation [6].

CASE REPORT

A 55-year-old female patient was referred to the Clinic of Dentistry in Niš, Serbia, with a painless swelling in the right region of the mandible, present for the previous 30 days. The patient said that four years prior to this she underwent an extraction of lower lateral teeth for prosthetic rehabilitation of the jaw, which included the extraction of the lower right impacted wisdom tooth. Extraoral palpation revealed an extremely hard swelling in the region of the body, angle and ramus of the mandible (Figure 1). Enlarged lymph nodes were also palpated in levels Ib and II of the neck, smaller than 1 cm in diameter, movable and painless. Intraoral examination revealed a complete lack of teeth in the upper and lower jaw and expanding

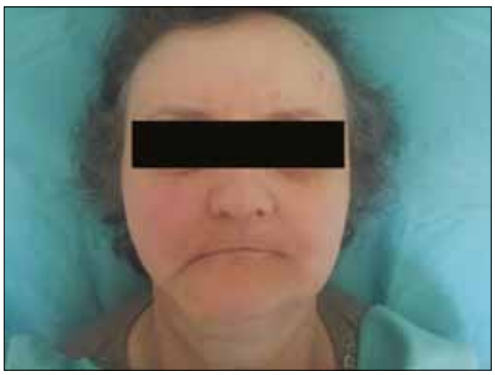

Figure 1. Swelling in the right region of the mandible

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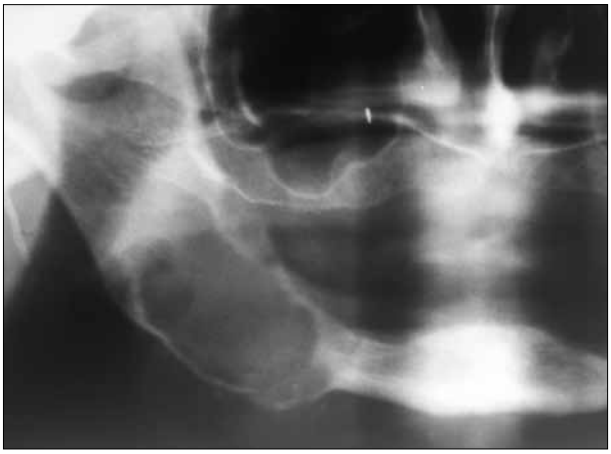

Figure 2. Presence of multi-locular irregular osteolytic lesion in the region of the body, corner, and branch of the mandible

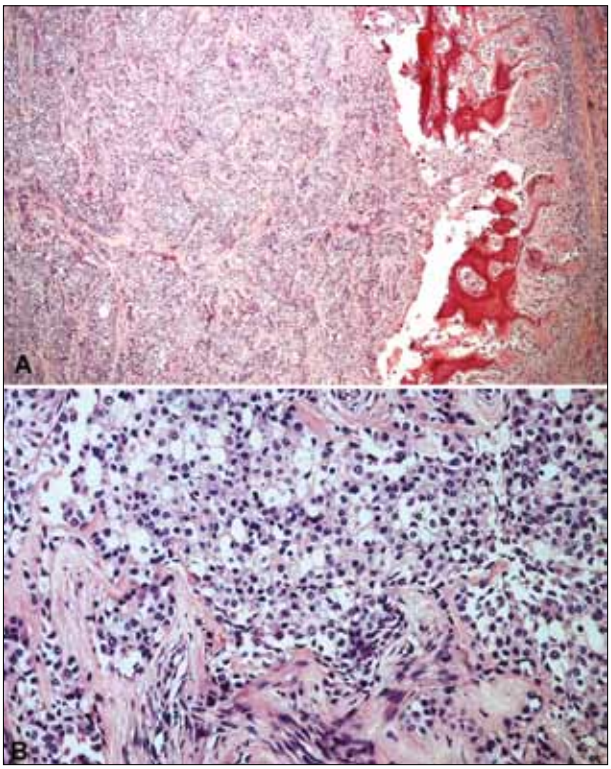

Figure 3. Mucoepidermoid carcinoma composed of basal, intermediate, and mucous cells (HE; A: ×4; B: ×20)

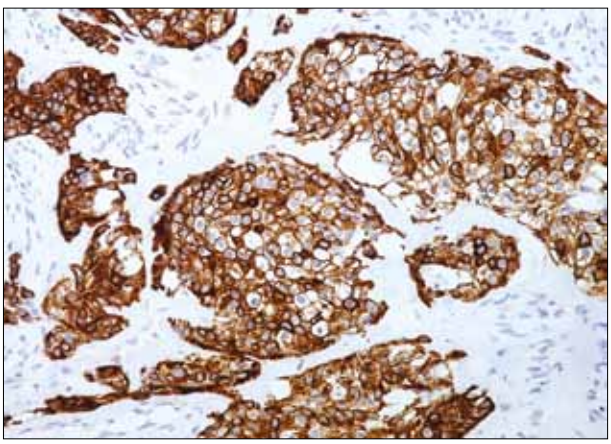

Figure 4. Tumor cells randomly positive on CK7 (cytoplasmic staining; ×20)

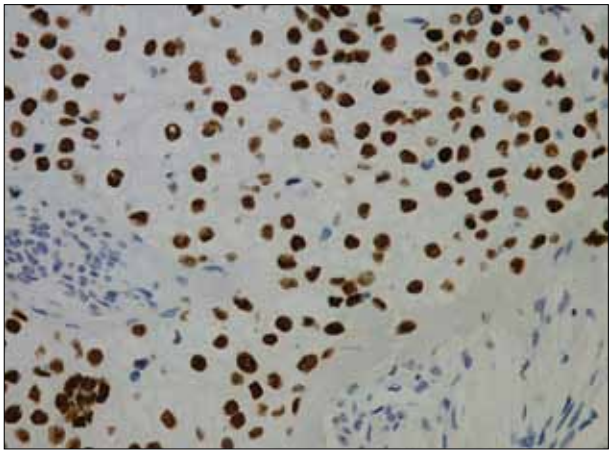

Figure 5. Tumor cells randomly positive on P63 (nuclear positive staining; ×40)

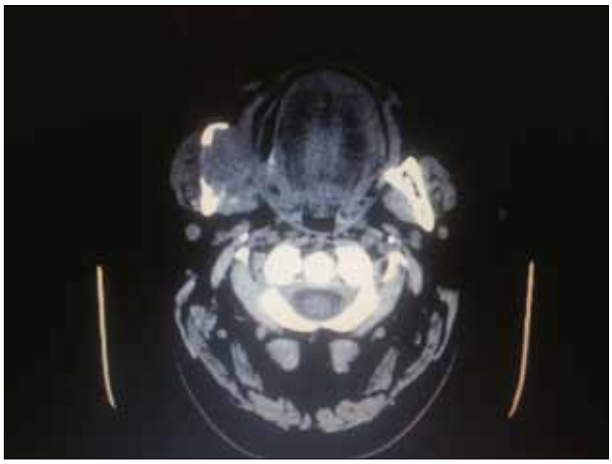

Figure 6. Multi-slice computer tomography (MSCT) revealed that lingual and buccal cortices are destroyed by tumour

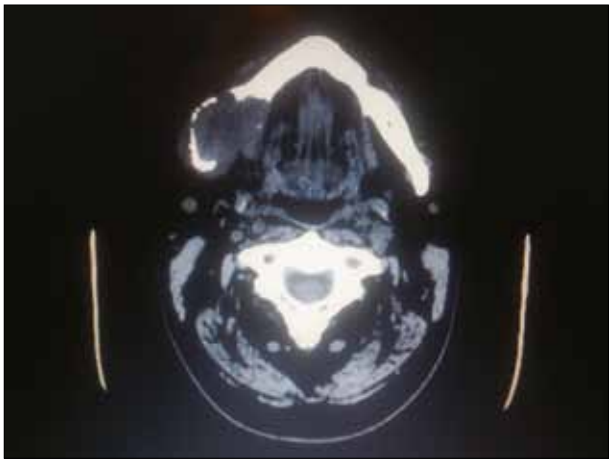

Figure 7. MSCT revealed an extension of tumor mass in the pterygomandibular space

tumor mass in the region of the body and angle of the mandible. The abovementioned region was covered with intact mucosa. Panoramic radiograph revealed a multilocular irregular osteolytic lesion in the region of the body, angle, and ramus of the mandible (Figure 2).

An incisional biopsy was performed due to large spectrum of pathological entities that could be diagnosed, and the result was a low-grade MEC (Figures 3–5). The tumor

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was composed of basal intermediary and mucosal cells. Presence of mucin was proven by alcian-blue PAS (AB-PAS) staining. Immunohistochemically, tumor cells were positive to CK7 and P63, and negative to S100. Magnetic resonance imaging revealed an expansive tumor formation with signs of bone destruction of the body, angle, and ramus of mandible, an extension of tumor mass to pterygomandibular space, as well as enlargement of lymph nodes at neck levels IIb, Ib, and IIa (Figures 6 and 7).

Considering the histopathological results and presence of enlarged lymph nodes, right hemimandibulectomy and tumour excision from pterygomandibular space, followed by supraomohyoid neck dissection, were performed. Operative histopathological result confirmed the presence of low-grade mucoepidermoid carcinoma, while the neck result was negative. Postoperative radiation therapy was applied after a successful postoperative period. The patient was followed up for the next two years through regular medical checkups every three months and then every six months. Clinical examination and insight into ultrasonography of the neck did not reveal signs of recurrence of the tumor or appearance of secondary deposits in the neck. A reconstruction of the postoperative defect of the mandible was proposed, which the patient refused.

DISCUSSION

CMEC is a very rare tumor, illustrated by only 200 described cases of this lesion in literature [7]. In average, it represents 2–3% of all MEC described in literature [8]. It is usually seen in women in the fourth and fifth decade of life, often in the region of the mandibular body [9]. Criteria that clearly confirm the diagnosis of CMEC include the following: 1. intact cortical bone; 2. radiological proof of bone destruction; 3. absence of primary tumor that would metastasize to the mandible; 4. absence of odontogenic tumor; 5. histopathological verification; 6. presence of intracellular mucin [10].

Degree of development of the disease is defined by the state of the surrounding bone [11]. Stage I represents an intact cortical bone, stage II intact cortical bone with minimal signs of expansion, and stage III represents a cortical destruction with encompassing surrounding periosteum. Our patient could be classified as stage III due to presence of buccal and lingual cortex of the mandible. Our patient had no metastases, although literature data show 9% of metastases in regional lymph nodes. Distant metastases are described in the region of the lungs and the brain [12].

True origin of the CMEC is still not explained in detail. Bouquot et al. [13] present data that intrabony presence of small salivary glands was spotted in 0.3% of the upper and lower jaw, which could be one of the causes. Ellis et al. [14] also presents data that confirm ectopic glandular tissue to be a rare pathological entity even in cases of histologically proven CMEC. Brookstone and Huvos [15] show evidence that CMEC is related to the presence of odontogenic cists in 32% of cases, while 1–2% of jaw tumors are related to the presence of impacted wisdom teeth.

Considering that our patient said that four years previously she had an extraction of the lower impacted wisdom tooth, it is possible that malignant transformation of the remaining cyst epithelial cells around the impacted tooth led to the creation of the CMEC.

The treatment included surgical removal of the tumor and post-operative radiation therapy. More conservative surgical methods like curettage, enucleation, and marginal resection of the jaw, with or without post-operative radiation, lead to relapse of primary disease in 40% of cases, while in the case of segmented resection of the mandible the relapse is in 4% of the cases [5]. Gradation of the tumor is one of the most important predicting factors for the survival of patients suffering from CMEC. High grade is characterized by larger bone destruction, enlarged frequency of relapses, and presence of regional and distant metastases [16].

CMEC represents a rare tumor, with causes still not fully defined in detail, which is characterized by signs of local destruction of tissue and ability to metastasize. Long-lasting asymptomatic clinical period, atypical radiological manifestation and wide spectrum of pathological entities can present a problem for the differential diagnosis during planning of the treatment. Initial biopsy represents the key to creating an operative plan. Radical excision with neck lymph node dissection followed by postoperative radiotherapy in our case represents successful method of treating CMEC of the mandible.

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Централни мукоепидермоидни карцином доње вилице – приказ болесника

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КРАТАК САДРЖАЈ

Увод Мукоепидермоидни карцином (МЕК) у односу на остале туморе пљувачних жлезда јавља се у 5 до 10% случајева. Хистопатолошки је подељен на добро диферентоване туморе који су ниског степена малигнитета, средње и лоше диферентоване туморе високог степена малигнитета. Централни мукоепидермоидни карцином (ЦМЕК) доње вилице је први описао Леп 1936. године, код 66-годишње пацијенткиње. ЦМЕК се одликује атипичном клиничком сликом и радиолошком презентацијом.

Приказ болесника Пацијенткиња старости 55 година јавила се на Клинику за Стоматологију у Нишу са анамнестичким подацима о присуству безболног отока у пределу доње вилице са десне стране. У складу са хистопатолошким резултатима и присуством увећаних лимфних нодуса дренажне зоне врата, урађена је супраомохиоидна лимфаденектомија праћена хемимандибулектомијом и екстирпацијом тумора у птеригомандибуларној области. Након уредно протеклог постоперативног тока спроведена је радиотерапија.

Закључак ЦМЕК представља редак тумор који карактеришу знаци локалне деструкције ткива и способност метастазирања. Иницијална биопсија представљала је кључ стварања оперативног плана. Хируршка ресекција инфилтрисаног подручја, лимфаденектомија врата и постоперативна радиотерапија били су успешни у лечењу ЦМЕК.

Кључне речи: централни карцином; оток доње вилице; биопсија; хируршка терапија

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