Case Report

Central Mucoepidermoid Carcinoma with Non-Hodgkin Lymphoma: A Case Report

Sayed Mohammad Razavi[1], Bijan Movahedian Attar[2], Saeedeh Khalesi[3]

[1] Professor, Dental Implant Research Center, Department of Oral and Maxillofacial Pathology, School of Dentistry, Isfahan University of Medical Sciences, Isfahan, Iran
[2] Professor, Dental Implant Research Center, Department of Oral and Maxillofacial Surgery, School of Dentistry, Isfahan University of Medical Sciences, Isfahan, Iran
[3] Assistant Professor, Dental Materials Research Center, Department of Oral and Maxillofacial pathology, School of Dentistry, Isfahan University of Medical Sciences, Isfahan, Iran

Abstract

Introduction: Central mucoepidermoid carcinoma (CMEC) is a rare primary intraosseous bony lesion. The mandible is more commonly affected than the maxilla. CMEC is frequently misdiagnosed radiographically and clinically as a benign odontogenic tumor or cyst.

Case Presentation: We present a rare case with a secondary malignant neoplasm. A male patient was initially diagnosed with B-cell non-Hodgkin lymphoma. Three months after his initial diagnosis, he presented with a large painless mass in the anterior region of his mandible. Histopathological examination revealed low-grade CMEC.

Conclusion: We reported a very rare case of CMEC in a patient treated for non-Hodgkin lymphoma without radiotherapy. Previous cases of secondary CMEC have demonstrated an increased risk in patients with leukemia/lymphoma after radiotherapy.

Key Words: Jaw neoplasms, Lymphoma, Non-Hodgkin, Carcinoma, Mucoepidermoid

Corresponding author: Saeedeh Khalesi, Assistant Professor, Dental Materials Research Center, Department of Oral and Maxillofacial pathology, School of Dentistry, Isfahan University of Medical Sciences, Isfahan, Iran

S_khalesi@dnt.mui.ac.ir

Received: 16 June 2020
Accepted: 26 Nov 2020

➢ **Cite this article as:** Razavi M, Movahedian Attar B, Khalesi S. Central Mucoepidermoid Carcinoma with Non-Hodgkin Lymphoma: A Case Report. J Iran Dent Assoc. 2021; 33(1-2):39-43.

Introduction

Mucoepidermoid carcinoma (MEC) is a common malignant salivary gland tumor (1). Intraosseous MEC of the jawbone is an extremely rare tumor and accounts for 2-4% of all central mucoepidermoid carcinoma (CMEC) cases (2). Several possibilities such as metaplasia of odontogenic cysts, entrapment of salivary gland tissues during embryonic development, and neoplastic changes of the maxillary sinus epithelium and dental lamina have been considered as the origin of CMEC (2-4). Involvement of the mandible is more common than the maxilla with a ratio of 2:1 (2). Posterior region of the mandible is the most common site of involvement (5). CMECs have a female predilection, and more commonly occur in the fourth and fifth decades of life (6,7).

Unilocular and/or multilocular radiolucency is the most common radiographic feature of CMEC which is similar to odontogenic tumors (8). In terms of histopathological features, three main cell types including epidermoid, mucus-secreting and intermediate cells have

been observed (7). CMEC is frequently misdiagnosed with odontogenic cysts or tumors and other lesions of the jawbone (2).

Limited cases of MEC with other malignant lesions such as lymphoma, leukemia and sarcoma have been reported (9-14). Herein, we present a case of CMEC in the anterior region of the mandible in a 79-year old male with malignant non-Hodgkin lymphoma. This is a very rare case of CMEC in a patient treated for non-Hodgkin lymphoma without radiotherapy.

Materials and Methods

A 79-year-old male patient presented to the Urology Clinic of Al-Zahra Hospital of Isfahan University of Medical Sciences with a chief complaint of pelvic pain and difficult urination started 3 months ago. There was no familial history or tobacco use but he had controlled hypertension. In abdominal and pelvic computed tomography, liver, pancreas, biliary system and kidneys were normal. A lesion on the right side of the pelvic space was evident extending to the lower abdomen with a compressive effect on the bladder. Furthermore, a pathologic lymph node measuring 76×127 mm was also found in the patient's right inguinal area.

For definitive diagnosis, a biopsy sample for histopathological examination was taken from the inguinal lymph nodes. The hematoxylin-eosin stained slides showed that the architecture of the nodule consisted predominantly of large cells with round to oval vesicular nuclei with pale to basophilic cytoplasm. Numerous atypical mitoses, apoptotic bodies, and areas of necrosis were observed. Immunohistochemical staining showed positive results for CD20 and Bcl2. But CD3, CD10, CyclinD1, TdT, and MUM1 were negative. Also, 90% of the nuclei were positive for Ki67. According to the results, the patient was diagnosed with diffuse large B-cell lymphoma (germinal center type) and he was referred to an oncologist for treatment.

After 3 months of chemotherapy, he was presented to the Oral and Maxillofacial Surgery Department of Isfahan Dental School with a chief complaint of progressive swelling in the anterior region of his mandible. In extraoral physical examination, there was inflammation in the chin area. The submental and submandibular lymph nodes were palpable. In intraoral examination, the swelling was smooth, firm and tender on palpation involving the anterior region of the mandible from the right second premolar to the left second premolar causing the expansion of buccal and lingual cortical plates with intact overlying mucosa. Panoramic radiograph showed a well-defined multilocular radiolucency in the anterior mandible (Figure 1). Furthermore, root resorption of premolars was observed. Based on the clinical and radiographic features, malignant lymphoma, odontogenic keratocyst, ameloblastoma, and central giant cell granuloma were considered in the differential diagnosis. Incisional biopsy was performed under local anesthesia for histopathological examination. A hematoxylin and eosin stained microscopic tissue specimen showed the presence of infiltrating sheets, nest and groups of neoplastic cells that included foci of epidermoid, mucous, intermediate and clear cells in a fibrous connective tissue (Figures 2A and 2B). The mucous and clear cells were stained by periodic-acid Schiff (Figure 2C). Based on the finding, the histopathological diagnosis of intraosseous MEC grade I was made.

The surgical treatment plan was segmental resection with safety margins. Histopathological examination of the excisional biopsy specimen confirmed the definitive diagnosis of intraosseous MEC. To prevent recurrence of the lesion, the patient was scheduled for radiotherapy. After 9 months of follow-up, there was no recurrence of the lesion.

Discussion

CMEC is the most common intrabony salivary gland tumor (15). The pathogenesis of CMEC is not known but radiation exposure has an important role in its development (14,15). Thus, radiotherapy is a risk factor for development of a secondary malignant neoplasm. MEC as a secondary malignant tumor is very rare with only few reports (14). Most of the previous

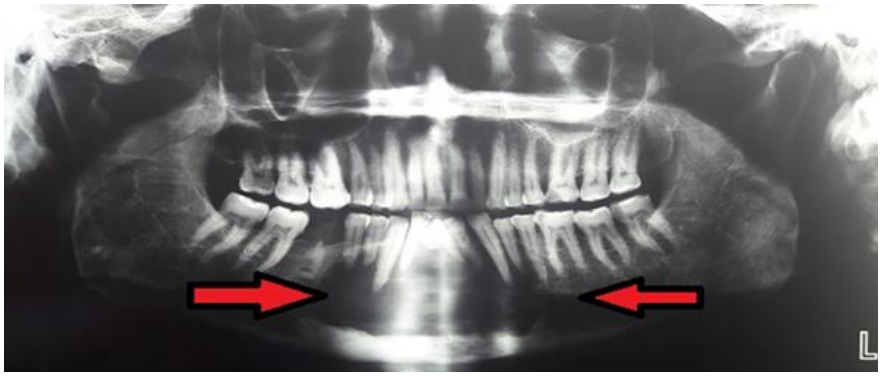

Figure 1. Panoramic radiograph showing multilocular radiolucency in the anterior mandible

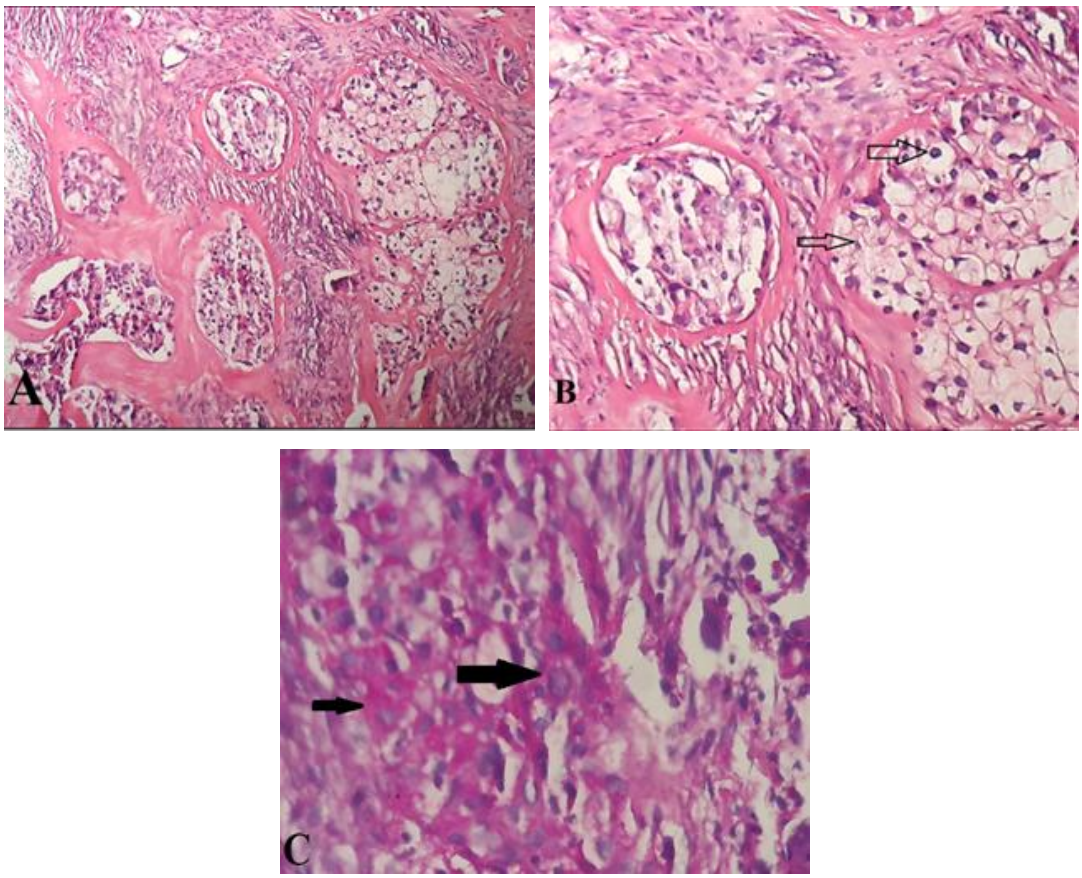

Figure 2. (A) Presence of infiltrating sheets, nest and groups of neoplastic cells in fibrous connective tissue of incisional biopsy specimen (H&E staining, original magnification ×100). (B) Foci of mucous and clear cells in the connective tissue. (C) Positive result for periodic-Acid-Schiff staining (original magnification ×400)

cases of secondary MEC diagnosed as leukemia and lymphoma were treated by multi-drug chemotherapy and radiotherapy (10-12,14). But, a few cases did not receive radiotherapy (9,16,17). Our patient received multi-drug chemotherapy without radiotherapy for treatment of non-Hodgkin lymphoma.

In general, the parotid gland is the most common site of MEC (15). In some studies, MEC of the parotid gland is a secondary malignancy in patients with acute lymphoblastic leukemia (12,14). Furthermore, the parotid gland is the site involved with MEC in patients with pediatric sarcoma such as Ewing sarcoma and osteosarcoma (10). Also, pulmonary MEC with anaplastic lymphoma has been reported (11). Savelli et al. reported MEC of the parotid gland as a secondary malignant neoplasm in a patient with B-cell lymphoblastic lymphoma (13). In this paper, we presented a CMEC in the anterior mandible in a patient with B-cell non-Hodgkin lymphoma which was a very rare occurrence. Similar to the most previous reports, our patient had low grade MEC (10,12-14).

In many patients, the interval between the treatment of primary malignant lesion and the secondary tumor occurrence was several months to years (10,12,14). However, in our patient, a secondary tumor developed shortly after treating the primary tumor. Similar to other reports of CMEC, our patient was an elderly (2). But unlike most other studies, the lesion involved the anterior region of the mandible (1-3,7). Our patient had a multilocular lesion on radiographic examination which was similar to the case reported by da Silva, et al (7). Unlike other malignant tumors of the jaw, CMEC appears mainly as a cyst or a benign tumor. A slow-growing swelling with pain, paresthesia of the inferior alveolar nerve and dissemination to lymph nodes is usually the most common clinical manifestation (1,7). Our patient only complained of swelling in the affected area.

Although most cases have been reported in females, Hernández-Arenas et al, (1) Nallamilli et al (5), da Silva et al (7), and Costa et al. (8) reported male patients with CMEC, similar to our patient.

Surgical removal of the tumor and postoperative radiotherapy are the best treatments for CMEC (3,4). According to the literature, curettage, enucleation and marginal resection of the jaw with or without postoperative radiation leads to a high recurrence rate of nearly 40%, while the recurrence rate with segmental resection of the mandible is only 4% (18).

Early and correct diagnosis of CMEC is very important especially in patients with other malignant tumors.

Conclusion

CMEC is a rare malignant tumor which can be misdiagnosed radiographically. Careful clinical examination and obtaining a precise medical history are essential for early and correct diagnosis.

Acknowledgment

This study was supported by Isfahan University of Medical Sciences.

References

1. Hernández-Arenas Y, Álvarez-Martinez EDC, Ardila CM. Diagnosis and treatment for central mucoepidermoid carcinoma in the mandible: report of a clinical case in a young patient. J Clin Exp Dent. 2019; 11(12):e1190-3.

2. Singh H, Yadav AK, Chand S, Singh A, Shukla B. Central mucoepidermoid carcinoma: case report with review of literature. Natl J Maxillofac Surg. 2019;10(1):109-13.

3. Aghakouchakzadeh A, Kargahi N, Shahnaseri S, Shakeri S. Central unilocular mucoepidermoid carcinoma of the mandible: A case report and literature review. Dent Res J (Isfahan). 2019;16(2):127-30.

4. Razavi SM, Yahyaabadi R, Khalesi S. A case of central mucoepidermoid carcinoma associated with dentigerous cyst. Dent Res. J 2017;14(6): 423-6.

5. Nallamilli SM, Tatapudi R, Reddy RS, Ravikanth M, Rajesh N. Primary intraosseous mucoepidermoid carcinoma of the maxilla. Ghana Med J. 2015;49(2):120-3.

6. Kechagias N, Ntomouchtsis A, Mavrodi A,

Christoforidou B, Tsekos A, Vahtsevanos K. Central mucoepidermoid carcinoma of the anterior region of the mandible: report of an unusual case and review of the literature. Oral Maxillofac Surg. 2015;19: 309-13.
7. da Silva LP, Serpa MS, da Silva LA, Sobral AP. Central mucoepidermoid carcinoma radiographically mimicking an odontogenic tumor: A case report and literature review. J Oral Maxillofac Pathol. 2016;20(3):518-22.
8. Costa AL, Ferreira TL, Soares HA, Nahas-Scocate AC, Montesinos GA, Braz-Silva PH. Cone beam computed tomography diagnostic imaging of intra-osseous mucoepidermoid carcinoma in the mandible. J Clin Exp Dent. 2017;9(9):e1158-61.
9. Sukumaran Nair RK, Rajeswari B, Thankamony P, Parukuttyamma K. Mucoepidermoid carcinoma of parotid gland as a subsequent neoplasm in children treated for acute lymphoblastic leukemia. J Cancer Res Ther. 2015;11(3):655.
10. Rutigliano DN, Meyers P, Ghossein RA, Carlson DL, Kayton ML, Kraus D, et al. Mucoepidermoid carcinoma as a secondary malignancy in pediatric sarcoma. J Pediatr Surg 2007;42(7):E9-13.
11. Onodera K, Sato N, Kobayashi K, Kurotaki H. [A Pulmonary Mucoepidermoid Carcinoma with Anaplastic Lymphoma Kinase Fusion Responding to Alectinib Hydrochloride]. Kyobu Geka. 2019;72(11):889-92.
12. Miyatima Y, Ogawa A, Kuno K, Toda K, Suzuki K, Mituya A. Mucoepidermoid carcinoma of the parotid gland as a secondary malignancy developed ten years after chemotherapy for childhood acute lymphoblastic leukemia. Rinsho Ketsueki. 2007;48(6):491-4.
13. Savelli SL, Klopfenstein KJ, Termuhlen AM. Mucoepidermoid carcinoma of the parotid gland as a second malignant neoplasm. Pediatric Blood Cancer 2005;45(7):997-1000.
14. Tikku G, Jain D, Kumari A, Grover R. Mucoepidermoid carcinoma of parotid as a second malignancy in acute lymphoblastic leukemia. Indian Pediatrics 2015;52 (15): 979-80.
15. Neville BW, Damm DD, Allen CM, Chi AC. Oral and Maxillofacial Pathology. 4th ed. St. Louis: Elsevier; 2016. 11, p:461-4.
16. Sandoval C, Jayabose S. Parotid mucoepidermoid carcinoma following chemotherapy for childhood acute lymphoblastic leukemia. Pediatr Hematol Oncol 2001;18: 217-20.
17. Védrine PO, Coffinet L, Temam S, Montagne K, Lapeyre M, Oberlin O, et al. Mucoepidermoid carcinoma of salivary glands in the pediatric age group: 18 clinical cases, including 11 second malignant neoplasms. Head Neck 2006; 28:827-33.
18. He Y, Wang J, Fu HH, Zhang ZY, Zhuang QW. Intraosseous mucoepidermoid carcinoma of jaws: report of 24 cases. Oral Surg Oral Med Oral Pathol Oral Radiol 2012;114:424-9.