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Central mucoepidermoid carcinoma: Report of a case with 11 years' evolution and peculiar macroscopical and clinical characteristics

Renata Tucci [1], Luciana-Fasanella Matizonkas-Antonio [1], Artur-Aburad de Carvalhosa [2], Paulo-Henrique-de Sousa Castro [3], Fabio-Daumas Nunes [4], Décio-dos Santos Pinto Jr [4]

[1] DDS, MS, PHD, Institute of Health Research, Sao Paulo, Brazil
[2] DDS, PHD, Cuiaba Cancer Hospital, Mato Grosso, Brazil
[3] DDS, MS, Cuiaba Cancer Hospital, Mato Grosso, Brazil
[4] DDS, MS, PHD, Oral Pathology, School of Dentistry, University of Sao Paulo, Brazil

Correspondence:
Institute of Health Research
Av. Indianópolis, 153
São Paulo - SP - Brazil
CEP:04063-000
renata-tucci@uol.com.br

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Abstract

Central mucoepidermoid carcinomas (CMC) are uncommon tumours, comprising 2-3% of all mucoepidermoid carcinomas reported. They have been reported in patients of all ages, ranging from 1 to 78-years, with the overwhelming majority occurring in the 4th and 5th decades of life. They are histologically low-grade cancers, usually affecting the mandible as uniocular or multiocular radiographic lesions. The authors report a case of CMC of the mandible with a long evolution, and peculiar clinical and macroscopical features related with the long term evolution of the disease. A 53-year-old male patient had expansion of buccal and lingual cortices of the anterior region of the mandible, covered by ulcerated mucosa, with 11 years evolution. An incisional biopsy was performed, and the histopathological findings confirm low-grade mucoepidermoid carcinoma. The patient was treated with a mandibulectomy, followed by supraomohyoid neck dissection. There was no evidence of local recurrence, regional or distant metastasis revealed; and the patient was alive and without disease after a follow-up interval of 36 months.

Keywords: *Mucoepidermoid carcinoma, central, intraosseous, salivary gland neoplasms.*

Introduction

Some 90% of oral cancers consist of squamous cell carcinomas that arise from the oral mucosa. The remaining 10% of malignancies consist of malignant melanomas, carcinomas of the intraoral salivary glands, sarcomas of the soft tissues and the bones, malignant odontogenic tumors, non-Hodgkin's lymphomas and metastases from primary tumors located elsewhere in the body (1). Salivary gland tumours are an important part of the Oral and Maxillofacial Pathology (2) and represent 3-5% of all head and neck neoplasms (3).

Central mucoepidermoid carcinomas (CMC) are extremely rare, comprising 2-3% of all mucoepidermoid carcinomas reported (4). We found reports of about 120 cases of central mucoepidermoid carcinomas (5,6).

The origin of the CMC is controversial and several pos-

sibilities have been considered, including: metaplasia of odontogenic cysts epithelium, entrapment of salivary tissues from the submandibular, sublingual or minor salivary glands, during embryonic development, entrapment of minor salivary glands from the retromolar area, maxillary sinus epithelium, iatrogenic entrapment of minor salivary glands (e.g. chronic osteomielitis and sinusitis) and odontogenic remnants of the dental lamina (7,8). More recently, intraosseous salivary tissue was demonstrated in 0.3% of bone specimens of all jawbones studied by Bouquot et al. (8), providing new evidences for the origin of intraosseous salivary carcinomas (9). Although its etiology is questionable, CMC is a well-accepted entity (9).

We report a case the CMC in a mandible of a 53-year-old man with 11 years evolution, showing clinical, tomographic, macro and microscopical characteristics.

Case Report

A 53-year-old man was referred to the Oral Medicine Service at the Cancer Hospital, Cuiabá, Mato Grosso, Brazil, for the diagnosis of mandibular lesion with 11 years evolution (Fig.1A). The patient refers growth of the lesion in the last month. His medical history was not significant. Clinical examination of the oral cavity revealed expansion of buccal and lingual cortices of the anterior region of the mandible, covered by ulcerated mucosa (Fig.1B). Cervical lymphadenopathy was absent. A computed tomography showed lobulated osteolytic lesion inside the mandibular body with inexate limits and cortical rupture (Fig.1C and 1D).

An incisional biopsy of the intraosseous lesion was performed. Histological examination of the specimen revealed a neoplasm composed predominantly of cystic spaces, containing myxoid material produced by clear

Fig. 1A. Expansive lesion in mandible with 11 years evolution.

Fig. 1B. Intra-oral examination revealed a large and firm lesion partially covered by an ulcerated mucosa causing expansion of buccal and lingual cortices of the anterior region of the mandible

Fig. 1C and 1D. A computed tomography showed lobulated osteolytic lesion inside the mandibular body with inexate limits and cortical rupture.

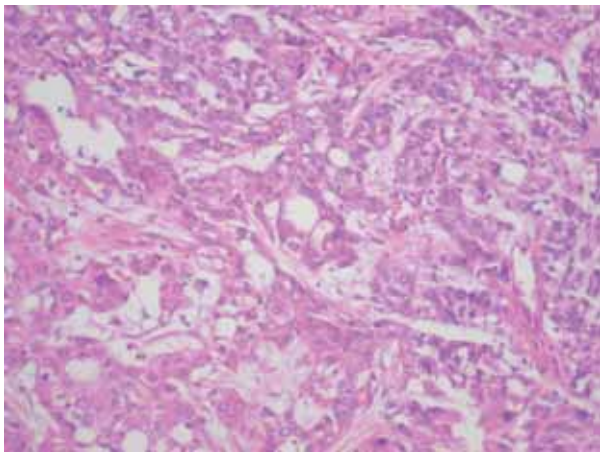

Fig. 2. Histological examination of the specimen revealed a neoplasm composed predominantly of nests of epidermoid cells with intermediate differentiation in a fibrous stroma and clear cells. (Haematoxylin an eosin, magnification X200).

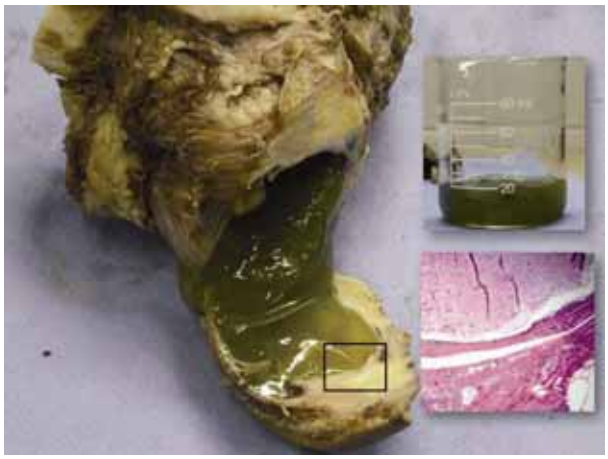

Fig. 3. A. After sectioning the surface, pseudo-cystic cavities showing green mucous secretion.
3. B. 20 ml of mucous secretion were removed from the cavities.
3. C. The histopathological characteristics of pseudo-cystic cavity showing mucous secretion are observed (Haematoxylin an eosin, magnification X100).

cells, and nests of epidermoid cells with intermediate differentiation in a fibrous stroma (Fig.2).

With a diagnosis of low-grade mucoepidermoid carcinoma a mandibulectomy with preservation of the condiles was performed, because the size of tumor. Gross examination of the ressected specimen revealed brownish tumoral mass involving all the jaw body, buccal and lingual cortical, from left to right molars. After cutting the surface, pseudo-cystic cavities showing green mucous secretion were observed (Fig.3A). 20 ml of mucous secretion were removed into the cavities (Fig.3B). The histopathological characteristics of pseudo-cystic cavity showing mucous secretion are observed in (Fig.3C). A histopathological analysis the surgical specimen reconfirmed the diagnosis of the low-grade mucoepidermoid carcinoma. The patient is on regular follow-up and is disease free after 3 years.

Discussion

Primary central salivary gland carcinomas of the mandible are uncommon neoplasms (10). Mucoepidermoid carcinoma generally affects the salivary glands and only rarely is located in the jaws (11,12). CMC affects females twice more frequently than males and involves the mandible twice more often than maxilla. The most common site of occurrence is the premolar-molar-angle region of mandible (13).

The criteria for diagnosing CMCs include: (a) presence of a radiographic distinct osteolytic lesion; (b) positive mucicarmine staining; (c) absence of rupture of one or more cortical plates; (d) clinical and histological exclusion of a metastasis or an odontogenic lesion; (e) exclusion of the origin from a soft tissue salivary gland; (9) histologic confirmation (14).

Our case showed rupture of cortical plate, but it has been shown in the literature that intact cortical plates should not be an essential feature for diagnosis of CMCs (15). Moreover we excluded other possibilities as metastases and odontogenic lesions.

Brookstone et al. in 1992 (14) proposed a staging system based on the condition of the overlying bone. Lesions with intact cortical plates with no evidence of bone expansion are Staged I; tumors with intact plates but intraosseous expansion are Staged II; and lesions associated with cortical perforation or nodal disease are Staged III. Our case is in staged III, because showed rupture of cortical plate.

Most of the reported, CMC are histologically low-grade tumours and usually carry a favorable prognosis (10). As a rule, even being low-grade tumours, CMC should be managed by wide local resection. Our case shows that, although the CMC is considered a low malignant potential carcinoma, in long-term evolution, it can be locally aggressive and requires wide en bloc resection.

In conclusion, CMC is a rare entity. Although CMC don't show clinic characteristics and un favorable prognosis, it can, how our case, showed destruction, local infiltration and ulcerated mucosa commonly in long time evolution cases. In these cases, bloc resection and effective follow-up are necessary for the success of the treatment.

References

1. Van der Waal R, Van der Waal I. Oral non-squamous malignant tumors; diagnosis and treatment. Med Oral Patol Oral Cir Bucal. 2007;12:E486-91.
2. Ledesma-Montes C, Garces-Ortiz M. Salivary gland tumours in a Mexican sample. A retrospective study. Med Oral. 2002;7:324-30.
3. Pons Vicente O, Almendros Marqués N, Berini Aytés L, Gay Escoda C. Minor salivary gland tumors: A clinicopathological study of 18 cases. Med Oral Patol Oral Cir Bucal. 2008;13:E582-8.

4. Goldfarb D, Mikaelian D, Keane WM. Mucoepidermoid carcinoma of the mandible. Am J Otolaryngol. 1994;15:54-7.
5. De Mello-Filho FV, Brigato RR, Mamede RC, Ricz HM, Saggioro FP, Xavier SP. Central mucoepidermoid carcinoma: report of 2 cases. Br J Oral Maxillofac Surg. 2008;46:239-41.
6. Simon D, Somanathan T, Ramdas K, Pandey M. Central Mucoepidermoid carcinoma of mandible - A case report and review of the literature. World J Surg Oncol. 2003;1:1.
7. Browand BC, Waldron CA. Central mucoepidermoid tumors of the jaws. Report of nine cases and review of the literature. Oral Surg Oral Med Oral Pathol. 1975;40:631-43.
8. Bouquot JE, Gnepp DR, Dardick I, Hietanen JH. Intraosseous salivary tissue: jawbone examples of choristomas, hamartomas, embryonic rests, and inflammatory entrapment: another histogenetic source for intraosseous adenocarcinoma. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2000;90:205-17.
9. Pires FR, Paes de Almeida O, Lopes MA, Elias da Cruz Perez D, Kowalski LP. Central mucoepidermoid carcinoma of the mandible: report of four cases with long-term follow-up. Int J Oral Maxillofac Surg. 2003;32:378-82.
10. Martínez-Madrigal F, Pineda-Daboin K, Casiraghi O, Luna MA. Salivary gland tumors of the mandible. Ann Diagn Pathol. 2000;4:347-53.
11. Namin AK, Moshref M, Shahoon H, Mashhadi A, Khojasteh A. Intraosseous mucoepidermoid carcinoma of the maxilla in a teenager: a case report and review of literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2005;100:e93-6.
12. Maremonti P, Califano L, Mangone GM, Zupi A, Guida C. Intraosseous mucoepidermoid carcinoma. Report of a long-term evolution case. Oral Oncol. 2001;37:110-3.
13. Gingell JC, Beckerman T, Levy BA, Snider LA. Central mucoepidermoid carcinoma. Review of the literature and report of a case associated with an apical periodontal cyst. Oral Surg Oral Med Oral Pathol. 1984;57:436-40.
14. Brookstone MS, Huvos AG. Central salivary gland tumors of the maxilla and mandible: a clinicopathologic study of 11 cases with an analysis of the literature. J Oral Maxillofac Surg. 1992;50:229-36.
15. Inagaki M, Yuasa K, Nakayama E, Kawazu T, Chikui T, Kanda S, et al. Mucoepidermoid carcinoma in the mandible: findings of panoramic radiography and computed tomography. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 1998;85:613-8.