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Case Report

A Case of Mucoepidermoid Carcinoma Associated with Maxillary Cyst

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Abstract

We report a case of a mucoepidermoid carcinoma associated with a maxillary cyst. The patient was an 18-year-old man presenting with the chief complaint of left buccal swelling. The left maxillary third molar was semi-impacted in the direction of the crown on the buccal side. Orthopantomography revealed a cystic radiolucency extending over a wide area, ranging from the left maxilla to the maxillary sinus and nasal cavity. Computed tomography revealed a multilocular lesion surrounded by a thin shell of bone. Biopsy findings revealed a cystic lesion, but the cause could not be identified preoperatively. The cystic lesion was resected under general anesthesia. The lesion was multilocular and surrounded by a bone shell, and had expanded into the maxillary sinus. At the same time, indurated soft tissue adhering strongly to the palatine bone on the inferior palatine side of the lesion was resected. Histopathological examination showed a cystic lesion and mucoepidermoid carcinoma. An additional resection was planned and the maxilla partially resected. Mucoepidermoid carcinomas usually occur in the parotid and minor salivary glands, but in rare cases appear in the center of a jaw bone, almost always in the mandible. In the present case, the carcinoma was associated with a cystic lesion believed to have developed from maxillary bone and involved an impacted tooth adjacent to the tumor. This suggested a central mucoepidermoid carcinoma in the maxillary cyst. Postoperatively, the missing teeth have been replaced with a denture and the course has been good, with no recurrences or metastases identified.

Key words: Mucoepidermoid carcinoma — Maxillary cyst — Central carcinoma

Introduction

Mucoepidermoid carcinomas are derived from ductal epithelial cells of the salivary gland and contain mucus-producing, epidermoid and intermediate cells. Although
usually occurring in the parotid gland in the head and neck region, they are often found in the palate when minor salivary glands are affected. Here we report a case of mucoepidermoid carcinoma associated with progressive cyst in the maxillary sinus of the left maxilla.

Case Report

The patient was an 18-year-old man. On examination at a local dental clinic in October 2008, extraction of an impacted third molar in the left maxilla was recommended and he was referred to our hospital. He presented with the chief complaint of left buccal swelling at initial examination at our department later the same month. Nothing noteworthy was found in his medical history. He had noticed a painless swelling in the left buccal region from around July 2008.

No notable systemic findings were made, but a facial examination revealed a slight horizontal asymmetry due to protrusion of the left buccal region. Protrusion of the buccal alveolar region around the left molars and swelling of the palatine alveolar region were found in the oral cavity (Fig. 1). The upper and lower first premolars on both sides were extracted for orthodontic treatment. No dental caries or tooth mobility were observed, but electrical resistance of the dental pulp from the left maxillary incisors to the second molar was reduced. The left maxillary third molar was semi-impacted in the direction of the crown on the buccal side. Orthopantomography revealed a cystic radiolucency extending over a wide area, ranging from the left maxilla to the maxillary sinus and nasal cavity (Fig. 2). Computed tomography (CT) revealed a multilocular lesion surrounded by a thin shell of bone. The upper part of the lesion extended to
near the orbital floor and compression and resorption of the buccal cortical bone and left wall of the nasal cavity were observed (Fig. 3). The diagnosis was an impacted left maxillary third molar and odontogenic tumor or maxillary cyst.

1. Treatment and course

Needle aspiration was performed on the lesion in November 2008, yielding a large amount of yellowish, serous semitransparent liquid. The left maxillary third molar was extracted and a biopsy of the adjacent cyst wall performed. Fenestration was also undertaken in the same region. The biopsy findings revealed a cystic lesion, but the cause could not be identified. Repeated irrigation was subsequently performed in the fenestrated area, and the size of the cystic cavity decreased. Therefore, removal of the residual lesion was planned. Another biopsy was performed preoperatively, but again yielded a diagnosis of a cystic lesion, as in the previous biopsy. In March 2009, the cystic lesion was resected under general anesthesia. The lesion was multilocular and surrounded by a shell of bone, and had expanded into the maxillary sinus. Indurated soft tissue adhering strongly to the palatine bone on the inferior palatine side of the lesion was resected. After making a relaxation incision of the mucoperiosteal flap, the wound was closed by suturing and the operation completed. Histopathological examination of the lesion revealed a cystic lesion and mucoepidermoid carcinoma. Given these findings, an additional resection was planned and the maxilla partially resected in April 2009. The operation consisted of extraction of the left maxillary second premolar and en bloc resection of the first and second molars and palatine bone (Fig. 4). The wound was closed and the defect covered with a buccal fat flap. Histological examination of fast-frozen tissue performed intraoperatively revealed no neoplastic changes in the vicinity of the resected sample. Postoperative course was favorable and the patient is currently being followed up using periodic CT and magnetic resonance imaging. As of 28 months postoperatively, the missing teeth have been replaced with a denture and the course has been good, with no recurrences or metastases identified.

2. Histopathological findings

A tumor was observed on the wall of the maxillary cyst (Fig. 5a). In the cystic area, the wall of the cyst was lined with pseudostratified ciliated epithelium including germ cells and stratified squamous epithelium. Infiltration

![Fig. 3 CT findings](Image)
A multilocular lesion surrounded by a thin shell of bone. Lesion extended to near orbital floor, the buccal cortical bone and left wall of nasal cavity.

![Fig. 4 En bloc resection of first and second molars and palatine alveolar bone](Image)
of inflammatory cells and vasodilatation and angiogenesis were observed. Connective tissue with fibrotic tendencies was seen in the outer layer, and trabecular bony tissue was found in the outermost layer. In the tumoric area, the tumor growth showed low dysplasia and consisted of squamous epithelial cells, mucus-producing cells and clear cells that were either connected with the lining epithelium or inside the connective tissue. Although a small amount of dysplasia was observed, oncocytic metaplasia was also observed (Fig. 5b). Tumor cells proliferated in the wall of the maxillary cyst in some areas. Glands of various sizes consisting of cuboidal and columnar cells were also observed in some areas. Some mucicarmin-positive tumor cells were observed (Fig. 5c). Part of the tumor had infiltrated the bone matrix in a nested pattern (Fig. 5d).

**Discussion**

Mucoepidermoid carcinoma is a tumor derived from ductal epithelium of the salivary gland, and was first reported by Stewart *et al.*\(^{21}\) as a mucoepidermoid tumor in 1945. Normally, these tumors grow slowly in a clinical setting and histologically the tumor cells exhibit low cellular atypia. Such findings suggest a benign tumor, but since metastasis has been reported, the name was changed to mucoepidermoid carcinoma in the World Health Organization (WHO) classification of 1991\(^ {18}\).
Mucoepidermoid carcinoma can be classified as well-differentiated or poorly-differentiated based on the relationship between its cell component ratio and malignancy\(^{18}\). The well-differentiated type consists of \(>50\%\) mucus-producing cells in the tumor parenchyma and includes well-differentiated squamous epithelial cells, while the poorly-differentiated type consists of undifferentiated intermediate cells with \(<10\%\) mucus-producing cells. Prognosis is good for well-differentiated lesions, as recurrences and metastases are rare. They are generally treated with surgical excision alone and the 5-year survival rate is 100%. However, the poorly-differentiated type shows a high local recurrence rate of about 50% and metastasis to the regional lymph nodes is also common. It is treated with wide surgical excision of the primary lesion and lymphadenectomy when cervical metastases are present, together with adjuvant external beam radiotherapy\(^{14}\). The 5-year survival rate is approximately 60%. The well-differentiated type was identified in the present case and partial resection of the maxilla was performed without postoperative radiation therapy.

Unlike mucoepidermoid carcinoma of the salivary gland, the histologic grade of central mucoepidermoid carcinoma does not appear to correlate with prognosis\(^4\). Metastases are reported in 9% of central mucoepidermoid carcinomas, mainly to the regional lymph nodes. As regards therapy, procedures that fall short of complete en bloc resection result in a high recurrence rate; enucleation or curettage result in a recurrence rate which varies between 40 and 45%\(^{3,6,24}\), whereas en bloc resection or radical excision have a 13% recurrence rate\(^5\). Freje et al.\(^8\) recommend radiotherapy for high grade tumors and suggest that an evaluation of lymph nodal status is always indicated and that a neck dissection should be performed in the presence of metastases.

Mucoepidermoid carcinomas often occur in the parotid and minor salivary glands, but in rare cases appear in the center of a jaw bone, almost always in the mandible\(^1-4,6-17,20,22-24\). Clinical findings include uni- or multi-locular radiolucencies on imaging. Histological onset has been suggested to involve neoplastic changes in the lining epithelium of an odontogenic cyst\(^23\). With respect to the origin of mucoepidermoid carcinoma, Eversole et al.\(^7\) described a relationship with odontogenic cysts or impacted teeth in about half of cases of mucoepidermoid carcinomas occurring in the center of the mandible. According to Brookstone and Huvos\(^5\), the percentage is about 32%. However, Pires et al.\(^17\) reported that establishing the origin and pathogenic relationship of odontogenic and glandular lesions can be difficult. For central mucoepidermoid carcinoma in the maxilla, classification of the origin as the palatine mucosa, gingiva or maxillary sinus mucosa is more difficult than for such tumors in the mandible. This may be one reason for the small number of case reports to date\(^{25}\).

One possibility in the present case is that the jaw cyst developed in the maxilla or maxillary sinus near to the mucoepidermoid carcinoma on the palatine side. There are three types of cyst associated with the maxillary antrum: mucoceles, retention or pseudocysts, and postoperative maxillary cysts\(^19\). Although no surgical treatment was performed in this region in the present case, the histological features of the cyst wall were similar to those of retention or pseudocysts. On the other hand, the higher possibility of a central mucoepidermoid carcinoma in the maxilla associated with a jaw cyst was suggested, as the tumor showed almost no outward progression and there was deep infiltration into the maxillary bone and base of the maxillary sinus; the tumor and cystic lesion were very close together; findings showed that the cyst had a bony shell, had arisen in the maxilla and had progressed into the maxillary sinus; and the spatial relationship with the maxillary impacted third molar differed from that of ordinary dentigerous cysts. Browand and Waldron\(^4,24\) reported that maxillary central mucoepidermoid carcinomas appear as multilocular or cystic radiolucencies in the posterior area, with no apparent continuity with the maxillary sinus, and generally involve the
molar-sinus-palatal area.

As no tumor cells were identified from the multilocular cyst-like lesion in the two biopsies, a diagnosis of mucoepidermoid carcinoma was not obtained preoperatively. However, a good course was achieved with partial resection of the maxilla. Treatment for mucoepidermoid carcinoma occurring in a jaw bone usually involves en bloc resection including the jaw bone. Indications for chemo- or radio-therapy have not been established\(^3,8\). The 5-year survival rate for this type of tumor is approximately 90% overall, but differs depending on the histological type and operative method. Therefore, the postoperative course must be observed very closely over a long period to identify local recurrences or metastases.

In conclusion, although the origin of the tumor was not clear in the present case, the clinical findings suggest a central mucoepidermoid carcinoma in the maxillary cyst.

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