Central Mucoepidermoid Carcinoma of the Mandible:
A Case Report

Kuninori Sasaoka,1 Kenji Mogi,1 Akio Kanou,1 and Akihide Negishi1

A case of central mucoepidermoid carcinoma involving the mandible in a 52-year-old man is described. There was a relatively clearly bordered swelling with slight redness accompanied by soft hardness on palpation was found in the left submandibular region. Segmental excision of the left mandible and neck dissection was carried out under general anesthesia and then examined histopathologically. We performed radiation therapy with Co60 (total 60Gy). There was no sign of recurrence six years after the operation. (Kitakanto Med J 2004; 54: 219~222)

Key words: central carcinoma, central mucoepidermoid carcinoma, mandible

Introduction

Carcinoma arising in the jaw is rare, and its pathogenesis and cause are still unknown. It is difficult to diagnose clinically, because it may be radiographically similar to other cystic lesions of the mandible or odontogenic tumors.

Mucoepidermoid carcinoma was first recognized in 1945 as a distinct entity of neoplasm by Stewart, et al.1 It is regarded as being of ductal origin and is chiefly composed of both the epidermoid cells and mucus cells in varying proportions. We report a case of central mucoepidermoid carcinoma of the mandible.

Case report

A 52-year-old man complaining of swelling in the left submandibular region was referred to our department from an affiliated hospital, the Department of Oral and Maxillofacial Surgery, Kurosawa Hospital, for diagnosis and treatment on June 8, 1998. His past medical history included acute renal insufficiency 28 years previously and mandibular fracture 16 years previously.

In present illness, he had noted of a slight swelling and an abnormal sensation in the left submandibular region for about a month. However, the swelling had rapidly become larger during the preceding two weeks, so he was advised to visit the Department of Stomatology and Maxillofacial Surgery, Gunma University Hospital for diagnosis and treatment on June 8, 1998.

On physical examination, he appeared to be healthy, was 176cm tall and weighed 78kg, and was well-nourished. All routine laboratory tests were within normal limits. A relatively clearly bordered swelling with slight redness and oppressive pain on the skin accompanied by soft elasticity on palpation was found in the left submandibular region, which involved the premolars. Facial paralysis and paresthesia were also found around the left side of the lower lip and chin (Fig. 1). A canine, the first and the

Fig. 1 Preoperative photograms of the left mandible showing a relatively clearly well-demarcated swelling with a slight red surface which was elastic to palpation.
second premolar and the second molar on the left side of the mandible were recognized, and were normal and without mobility. The region exhibited a clearly bordered expansion of the left buccal region without redness and oppressive pain in the overlying mucosa which involved the premolars.

In orthopantomograms, a 40 × 30mm nearly round and slightly non-uniform monolocular radiolucency with the mandibular body of the median part of the first premolar as anterior border and that of second molar as posterior border was observed in the left side of the mandibular body. The outline of the radiolucent area was not sharply demarcated, and the cortex of the lower part of the mandibular body had disappeared (Fig. 2-A). In a computed tomogram examination, a polylocular cystic lesion was observed in the left lower border of the mandible, and the buccal and lingual cortices have been irregularly destroyed and disappeared (Fig. 2-B). Based on all findings, we made the clinical diagnosis of central carcinoma of the mandible.

Aspiration biopsy of the lesion through the skin revealed a large amount of black serous fluid with blood. The possibility of a cystic lesion was confirmed. A small specimen was taken for biopsy to a pathologist. However, the report was fibrous connective tissue, when we performed a second biopsy, we observed a cystic lesion under the platysma, and the diagnosis by the histopathologist was squamous cell carcinoma, non-keratinizing, in an odontogenic cyst. Gallium scintigraphy revealed strong uptake only in the portion of the tumor on the left side of the mandibular body.

He was admitted to our hospital on July, 30, in 1998. On August 3, segmental excision of the left mandible and neck dissection were performed under general anesthesia. We performed left-side total and right-side suprahyoid neck dissection, and en-bloc segmental excision with overlying skin. In the area of segmental excision of the mandible, the medial border was the midline and the distal border was a portion of the second molar. In Surgical specimen, the expanded portion was almost completely replaced by the tumor, and the tumor infiltrated into the surrounding buccal soft tissues. The buccal and lingual cortical plate was not intact. There were several submandibular and jugular chains of lymph nodes, but no metastasis was found in any of the specimens from these lymph nodes.

Microscopically, the tumor exhibits nests of undifferentiated epidermoid cells. In addition to these cells, mucous cells which contain mucous material are seen (Fig. 3). These features confirmed central mucoepidermoid carcinoma. The post-operative course was uneventful. But, after the removed speci-
patient made a uneventful recovery and was discharged in October 20. There was no sign of recurrence or metastasis after six years after operation.

**Discussion**

Salivary gland tumor occurring intraosseously in the jaws are rare lesions. There is general agreement that mucoepidermoid carcinoma is the most common salivary gland tumor found in an intraosseous location, and occurs mainly in females in the fourth to the fifth decades. Waldron and Koh\(^2\) reported that intraosseous mucoepidermoid carcinomas occur about three times more commonly in the mandible than in the maxilla. Browand et al.\(^3\) reported a significant number of the mandibular lesions were located in third molar angle-ramus area, and the maxillary lesions generally involved the molar-sinus-palatal area. But in our case, the patient was a 52-year old man, and the tumor was recognized in the mandibular body of the median part of the first premolar as anterior border and that of second molar as posterior border.

Most reported cases of central mucoepidermoid carcinoma have been described as radiographically varied and nondiagnostic. Waldron and Koh\(^2\) reported radiographic findings in about half of the cases indicated a unilocular radiolucent lesion, and about half have been stated to be multilocular lesions. Browand et al.\(^3\) reported most reported cases have been described as radiographically radiolucent and often multilocular, a few lesions were reported as diffusely destructive with vague or poorly defined borders, the most frequent radiographic diagnosis may be ameloblastoma or dentigerous cyst. In our case, a polilocular cystic lesion was observed and the outline of the radiolucent area was not sharply demarcated and the buccal and lingual cortices have been irregularly destroyed and disappeared, so we made the radiographical diagnosis in CT of central carcinoma of the mandible.

The microscopic appearance of the central mucoepidermoid carcinoma is similar to that noted in mucoepidermoid carcinomas arising in salivary glands. The three basic cell types are present in varying proportions, mucous, epidermoid and intermediate cells. Tsang et al.\(^4\) suggested the proportion of these cell types and their architectural configurations vary between tumor and these histological differences reflect the various clinical features.

Ito et al.\(^5\) suggested the low malignant type is characterized by multiple cell types, abundant production of microcysts and mucus pools and relative predominancy of stroma. On the other hand, the high malignant type tends to be more uniform in cell types and shows predominancy of epidermoid cells with relatively few mucus cells, microcysts and mucus pools. Waldron and Koh\(^2\) suggested that vast majority of central mucoepidermoid carcinoma of the jaws have been low grade, predominantly cystic lesions. In our case, on H.E-stained feature of the specimen, the tumor exhibits nests of undifferentiated epidermoid cells. In addition to these cells, mucus cells which contain mucous material are seen. The majority of these cells were dispersed singly or in tiny clusters within the epidermoid cells. These features confirmed the malignant type of central mucoepidermoid carcinoma.

Commonly accepted criteria for the diagnosis of a central mucoepidermoid carcinoma suggested by several authors\(^2,5,6\) include the following: (1) presence of intact cortical plates, (2) radiographic evidence of bone destruction, (3) histologic confirmation, (4) positive mucin staining, (5) absence of a primary lesion in the salivary glands or elsewhere which can mimic the histologic features of a mucoepidermoid carcinoma, and (6) exclusion of an odontogenic tumor. In our case, the buccal and lingual cortical plate was not intact at the time of first visit, it was impossible to get the proof of the presence of intact cortical plates.

A variety of treatments have been employed, including conservative enucleation and curettage, wide surgical excision, en bloc resection, partial and radical jaw resection with or without lymph node dissection, electrocoagulation, and radiation therapy. The studies of Gray and et al.\(^7\) have shown that the majority of mucoepidermoid tumors pursue a relatively benign course and are curable when adequately treated. But, Alexander et al.\(^8\) suggested that aggressive therapeutic efforts are essential in view of the high incidence of recurrence, and it should be emphasized that conservative enucleation or small-marginal excision of these lesions must be avoided to prevent recurrence or the more serious sequelae associated with distant metastases. Browand et al.\(^3\) reported that conservative therapy has been employed in about 40 per cent of reported cases, while various forms of more extensive therapy have been used in the remaining cases. And the Review of the 41 previously reported cases and the nine case in the present series showed thirteen instances of recurrence, four instances of metastasis, and four deaths attributable to the tumor. Jenkins et al.\(^8\) suggested the radio-therapy could improve the cure rate, particularly if there was doubt about the surgical margin. Because our case was diagnosed as squamous cell carcinoma in an odontogenic cyst on the second biopsy, we performed segmental excision of the left mandible and neck dissection. And we received a report of surgical specimen suggesting the possibility of tumor in the lingual border of the mandible, so we performed radiation
Discussions of histogenesis have embraced three main theories for the source of origin for these tumors suggested by several authors: (1) mucous metaplasia of the lining epithelium of odontogenic cysts; (2) ectopic submandibular or possibly sublingual salivary gland tissue - the so-called static bone cavity or "Stafne defect"; (3) embryologically entrapped retromolar mucous salivary gland tissue. No single theory can adequately explain the origin of all these lesions, and it may be that all three are valid. In our case, the fractures were recognized on both sides of the condylar process, the right side of the coronoid process, and the left side of lower border of the mandible in the orthopantomograph taken three months after mandibular injury 16 years previously (Fig. 4). Concerning the relations between the origin and the fracture line of the lower border of the mandible, there are possibilities of ectopic salivary gland tissue and some influences of the fracture. Concerning this case of mucoepidermoid carcinoma, we supported that the occurrence of odontogenic cyst is possible, but we cannot rule out the possibility of ectopic salivary gland tissue due to fracture. Reporting and analysis of additional cases of central mucoepidermoid carcinoma of the mandible may help to clarify this point.

References

2. Waldron CA and Koh ML. Central mucoepidermoid carcinoma of the jaws. J Oral Maxil-

Fig. 4 Panoramic radiograms taken 16 years previously showing fracture lines of the mandible (arrows).