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Case Report

A case of ameloblastoma with extensive pulmonary metastasis survived for 14 years without treatment of the lung

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ABSTRACT

Although ameloblastoma is generally considered to be a benign tumor, local recurrences have often been noted. On the other hand, however, lung metastasis is quite rare. Here, we present a case of a patient who had ameloblastoma in the mandible with metastases in a lung and several recurrences. The patient was a 24-year-old man who was treated, but developed lung metastasis 1 year later. Once it had metastasized, recurrences occurred twice. Histologically, both the primary tumor and lung metastasizing tumor were benign. The patient died 27 years after the initial treatment and 14 years after detection of pulmonary metastasis. To our knowledge, this patient was the longest-survival case in Asia for such conditions.

1. Introduction

Ameloblastoma represents 11–13% of all odontogenic tumors [1], with 80–85% occurring in the mandible [2–5]. Ameloblastoma is generally considered to be a benign tumor of the odontogenic epithelium. However, recurrences have been noted for more than 10 years after the initial treatment [6]. Moreover, there is a type of ameloblastoma known as metastasizing ameloblastoma or ameloblastic carcinoma which metastasizes to the lung. The metastasizing ameloblastoma, however, is different from ameloblastic carcinoma. Metastasizing ameloblastoma causes distant metastasis despite it having the benign histological appearance. Ameloblastic carcinoma behaves more aggressively and demonstrates distinct cytologically malignant tumors [7]. However, lung metastasis is not common.

We present a case of metastasizing ameloblastoma of a patient with bilateral pulmonary metastases, who survived for 14 years after detection of metastasis without treatment for lung lesions.

2. Case report

In 1983, a 24-year-old man was referred to our hospital for a swelling of the right premolar region in his mandible. A panoramic radiograph showed a cystic radiolucent image. Biopsy and histological examination revealed an ameloblastoma. The patient was admitted to our hospital and underwent a marginal resection of the right premolar region in the mandible under general anesthesia. There was no sign of invasion into the bone at the margin. The tumor was not attached to the surrounding bone and therefore easily removed. Then we finally performed curettage of the surrounding bone with a burr. The pathological findings included nests which had formed follicular variant and increased in the connective tissue; the nests consisted of stellate reticulum and spindle cells in the central region, and palisaded cuboidal or columnar cells in the peripheral region. In the nests, clear cells without atypia were occasionally seen (Fig. 1).

After the surgery, there was no event for more than 10 years. In January 1996, the patient had a medical check-up and an abnormality was detected in his chest radiograph (Fig. 2). We referred him to the department of internal medicine in our hospital to assess the lesion in his lung. A CT showed some small nodules in both lungs; other organs had no abnormality. As we suspected metastasis of the mandibular ameloblastoma, the patient underwent an open biopsy of the pulmonary tumor. The pathological findings of the biopsy included tumor cells forming small nests in the pulmonary alveoli tumor cells forming small nests in the pulmonary alveoli. The nests consisted of spindle cells in the central region and cuboidal cells in the peripheral region (Fig. 3).

☆ Asian AOMS: Asian Association of Oral and Maxillofacial Surgeons; ASOMP: Asian Society of Oral and Maxillofacial Pathology; JSOP: Japanese Society of Oral Pathology; JSOMS: Japanese Society of Oral and Maxillofacial Surgeons; JSOM: Japanese Society of Oral Medicine; JAMI: Japanese Academy of Maxillofacial Implants.

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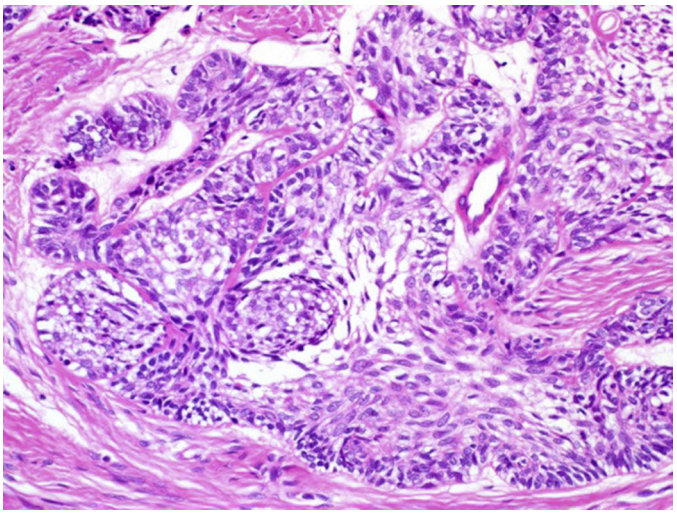

Fig. 1. The nests consisted of stellate reticulum and spindle cells in the central region, and palisaded cuboidal or columnar cells in the peripheral region. In the nests, clear cells without atypia were occasionally seen.

Those were compatible with the specimen that we obtained from his mandible in 1983. The lung specimen showed no malignant feature. As we considered that chemotherapy was not effective in the lung, the patient was followed up periodically. The doctor in the internal medicine department prescribed him a traditional Chinese medicine but nothing else. In July 2003, the patient visited our hospital with a chief complaint of persistent gingival swelling of the right molar region in the mandible for several months. Examination revealed an elastic hard mass, and radiography showed a radiolucent image at the root apex and surrounding bone of the right molar (Fig. 4). Biopsy was performed and histological examination revealed recurrent ameloblastoma. Under general anesthesia, the operation which included extraction of the molar and marginal resection of the mandible was performed. No malignancy was found pathologically in the resected specimen. However in the specimen, clear cells consisting of ovoid, clear cytoplasm and pyknotic nuclei were observed in the nests at various degrees (Figs. 5 and 6). From 2004 to 2006, the healing course was uneventful. Periodic CT and radiography of the lung at 3-month intervals demonstrated stability of the nodules. There was no sign of growing pulmonary foci. The condition of the oral wound was satisfactory. The patient received a new partial denture. And we checked his oral hygiene and took periodic radiographs of his mandible.

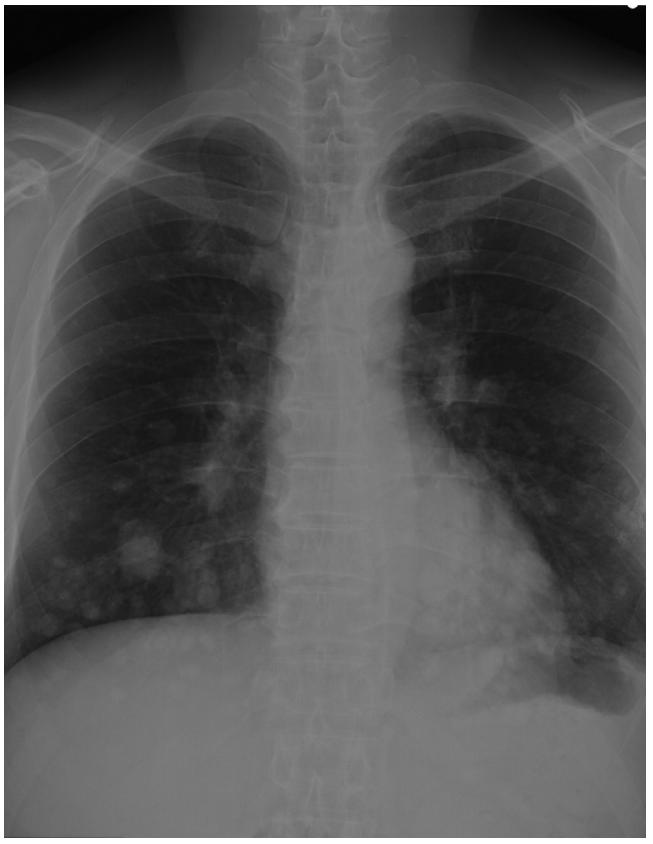

Fig. 2. Chest radiograph showing some nodules in both lungs in 1996.

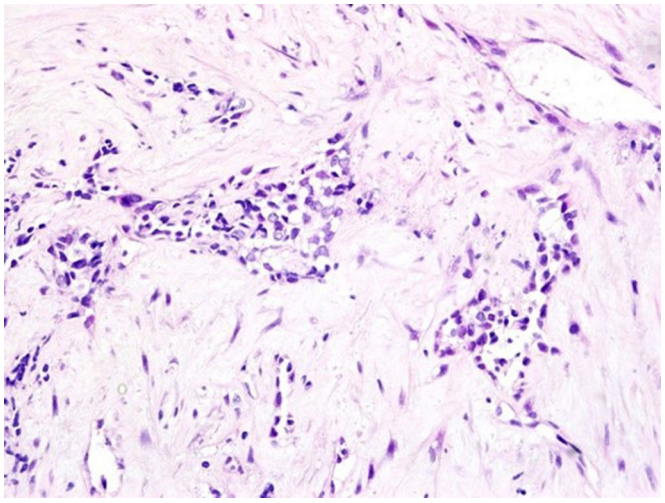

Fig. 3. Tumor cells forming small nests in the pulmonary alveoli. The nests consisted of spindle cells in the central region and cuboidal cells in the peripheral region.

In 2007, although the oral wound showed no signs of recurrence, the lung nodules had increased in size and spread out. By December 2008, the size of each pulmonary tumor increased even more. By August 2009, gingival swelling of the right mandible was detected. It was an elastic hard mass and radiograph showed radiolucent image of the anterior tooth apex (Fig. 7). Pathologic examination of a biopsy sample of the molar gingiva and anterior tooth apex revealed a recurrence of ameloblastoma. The patient underwent a partial resection of the right molar region of the mandible, including extraction of the anterior incisors. In May 2010, his oral condition was stable, with no recurrence. However, the patient felt dyspnea and returned to our hospital. Examination of a chest radiograph by the physician in charge revealed bilateral pleural effusion, which was drained. In June 2010, the patient's oral intake gradually worsened and his body weight decreased noticeably (Fig. 8). Thereafter, fluid drainage from his lungs was less efficacious. The pleural effusion gradually increased and compressed his heart, and the lung and heart function worsened. In July 2010, the patient died, 27 years after the initial treatment and 14 years after the diagnosis of pulmonary metastasis.

3. Discussion

According to the 2005 World Health Organization classification of odontogenic carcinomas among malignant tumors derived from odontogenic tissues, ameloblastomas are divided into metastasizing ameloblastoma, ameloblastic carcinoma-primary type, and ameloblastic carcinoma-secondary type (intraosseous, peripheral) [8]. Metastasizing ameloblastoma is defined as ameloblastoma that metastasizes despite benign histological appearance. Ameloblastic