Spindle cell variant of ameloblastic carcinoma: a case report and literature review

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Spindle cell variant of ameloblastic carcinoma is an extremely rare tumor. Severe dedifferentiated spindle cell variants are diagnostically challenging, particularly in small biopsy specimens. Here, we report a case of spindle cell variant of ameloblastic carcinoma in the mandible of a 69-year-old male patient and review the available literature. The tumor was surgically resected under general anesthesia. Histopathologic diagnosis of spindle cell carcinoma was made on incisional biopsy, and the final diagnosis was confirmed as spindle cell variant of ameloblastic carcinoma. Immunohistochemistry using cytokeratin and CK19 is helpful in determining the origin of spindle cell variant of ameloblastic carcinoma, particularly CK19 indicated that sarcomatoid spindle cells are derived from odontogenic epithelium. A review demonstrated higher mean age of patients compared with that of other types of ameloblastic carcinoma. The rates of mortality and local recurrence were concurrently 30%. No recurrence or metastasis was seen in the 23-month follow-up period in the present case. (Oral Surg Oral Med Oral Pathol Oral Radiol 2016;121:e54-e61)

Odontogenic tumors have been defined and classified by the World Health Organization (WHO). Malignant odontogenic tumors have been classified into odontogenic carcinomas and odontogenic sarcomas, but odontogenic carcinosarcomas have been removed from the WHO classification of 2005.[1] Ameloblastic carcinoma has been characterized as an odontogenic tumor with the histologic features of an ameloblastoma accompanied by cytologic atypia, regardless of the presence of metastasis. It is subcategorized into (1) primary type; (2) secondary type (dedifferentiated), intraosseous; and (3) secondary type (dedifferentiated), peripheral.

We encountered an extremely rare case of spindle cell variant of ameloblastic carcinoma in the mandible of a 69-year-old Japanese male. There have been reported a few cases of spindle cell variant of ameloblastic carcinoma.[2-10] Some cases of this variant are considered a secondary type of ameloblastic carcinoma. They showed malignant transformation accompanied by the appearance of atypical spindle cells in ameloblastic islands and infiltration of these spindle cells into the surrounding stroma.[8,9] Intense dedifferentiation and proliferation lead to carcinomatous and sarcomatoid histologic features, which no longer resemble ordinary ameloblastic carcinoma. Here, we report the case and discuss the histopathologic diagnosis and clinical differences with ordinary ameloblastic carcinoma, along with a literature review.

CASE REPORT

A 69-year-old male was referred to the Nagasaki University Hospital with a complaint of abnormal sockets of extracted bilateral lower first premolars, detected at a dental clinic. He had no clinical symptoms. His facial appearance was asymmetric, and a bonelike swelling was observed on the right side of the mental region. A clinical oral examination revealed a swelling on the buccal side covered with intact mucosa (Figure 1A). A panoramic radiographic image revealed widespread bone destruction in the mandible (Figure 1B). Computed tomography (CT) showed a poorly marginated, 60-mm, multilocular lesion that extended on both sides of the mandible (Figure 1C). The mandibular bone was buccolingually destroyed. There appeared to be two centers of mass in the mandible separated by a bony wall. The lesion infiltrated just under the skin. Magnetic resonance imaging (MRI) yielded observations that were similar to

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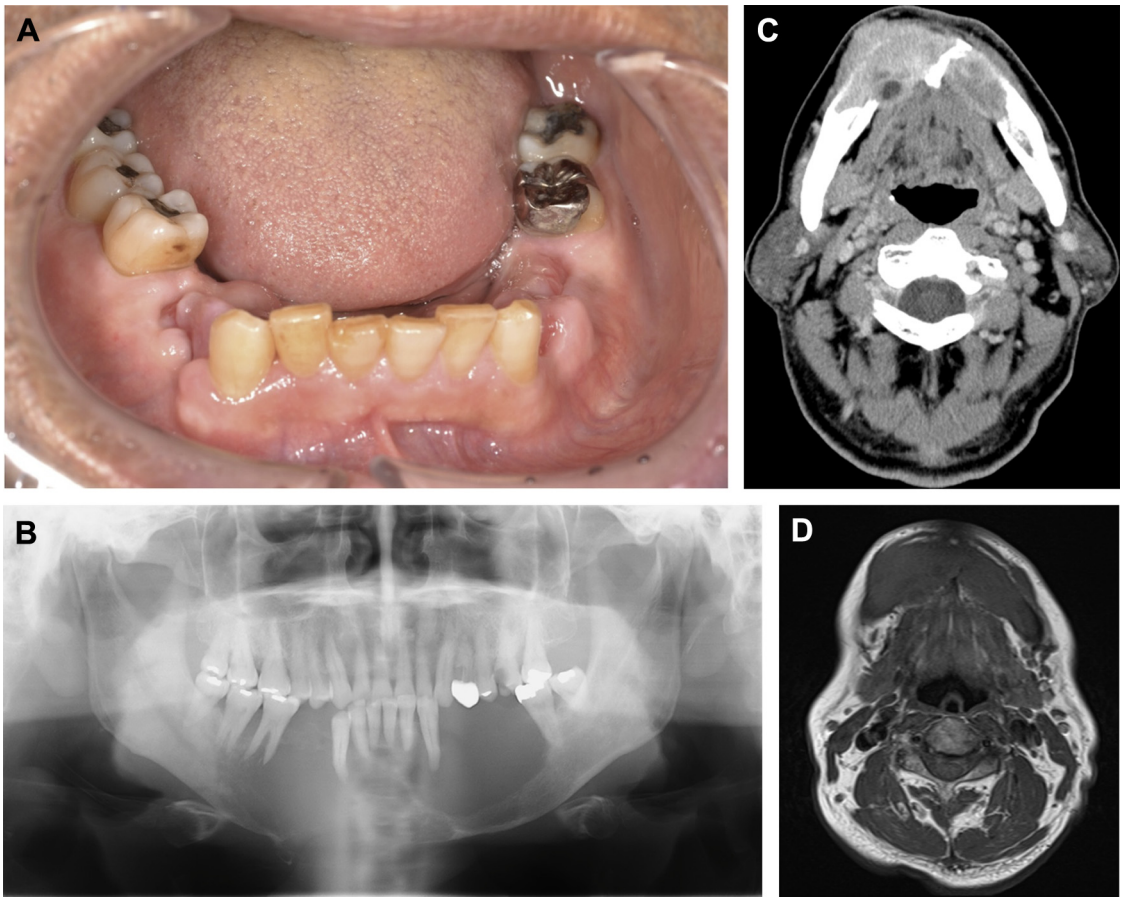

Fig. 1. Preoperative clinical and imaging results. **A**, Swelling on buccal side covered with intact mucosa, including bilateral extracted sockets between the gingiva. **B**, The panoramic radiographic image revealed widespread bone destruction in the mandible. **C**, Computed tomography (CT) showed a poorly marginated, multilocular lesion that extended bilaterally along the mandible. The lesion infiltrated just under the skin. **D**, Magnetic resonance imaging (MRI) yielded observations that were similar to those of CT.

those of the CT (Figure 1D). The time−intensity curve derived from the dynamic study exhibited a mixture of rapid increase, gradual decrease, and gradual increase patterns. CT/MRI/ultrasonography showed no evidence of cervical lymph node metastasis. On the basis of the clinical and imaging findings, the lesion was suspected to be a malignant tumor arising from the mandibular bone.

Incisional biopsy specimen showed neoplastic appearance comprising a sarcomatoid component consisting of spindle cells and a carcinomatous component exhibiting dense and solid proliferation of polyhedral cells (Figures 2A and 2B). The tumor cells of both areas had deeply stained atypical nuclei and scattered mitotic figures. Spindle cells of the sarcomatoid area showed a fascicled arrangement and storiform pattern, accompanied by collagen bundles−like pleomorphic undifferentiated sarcoma (Figure 2C). Some borders of the carcinomatous nests were indistinct. In some areas, the tumor cells emerged from the carcinomatous nests and blended with the sarcomatoid spindle cells (Figure 2D). The immunohistochemical study revealed strong reactivity for cytokeratin AE1/AE3 (Dako, Glostrup, Denmark) in the carcinomatous and sarcomatoid areas (Figure 2E), and positive results for vimentin (Dako) in the spindle cells. The Ki-67 (Dako, clone: MIB-1) labeling index was 34.2%. On the basis of histologic and immunohistologic findings, we concluded that both types of neoplastic cells had an epithelial origin. Although the mucosal epithelium was not included in the specimen, we made a pathologic diagnosis of spindle cell carcinoma because we could not identify obvious ameloblastic patterns in the biopsy specimen.

After the diagnosis of spindle cell carcinoma was established, we performed extended surgery. The tumor was surgically removed under general anesthesia. Bilateral modified radical neck dissection, segmental resection of mandible, and fibula flap reconstruction were performed (Figure 3). For surgery, a three-dimensional replica model of the mandible was made, with the help of preoperative CT images (see Figure 3A), and preoperative model surgery was performed. Ideal reconstructed mandible shape made by fibula (see Figure 3B) was preshaped using silicon putty impression material as reference (see Figure 3E).[11] The tumor was resected, along with mandibular bone, using segmental resection.