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Case report

Sclerosing odontogenic carcinoma in the mandible

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ABSTRACT

We report a case of an asymptomatic sclerosing odontogenic carcinoma in a 68-year-old woman. A bulge was noted in the anterior mandibular region, and it increased rapidly without clinical symptoms. On computed tomography, a radiolucent osteolytic lesion was identified in the central incisor apex of the mandible, and the labial cortical bone disappeared. Magnetic resonance imaging showed an internal heterogeneous circular mass outside the alveolar region. Pathological assessment of a biopsy specimen did not lead to a definitive diagnosis, but a benign odontogenic tumor was suspected. The tumor was resected en bloc with 4 incisors. The stem of the tumor was attached to the alveolar bone, but it was mainly outside the bone. Histopathological examination of the surgical specimen showed a capsule under the epithelium and no cellular atypia with bland cytoplasmic cuboidal or polygonal epithelial cells forming a small circle or polygon in the sclerosing stroma. A similar tumor was scattered in the alveolar bone. Immunohistologically, the tumor was positive for cytokeratin (AE1/AE3) and p63, and only a small number of Ki-67-positive cells were noted (approximately 1%). The tumor in our case was not consistent with odontogenic tumors in previous classifications. Based on previous findings, clinical behavior and immunohistological findings, the final diagnosis was a sclerosing odontogenic carcinoma. There has been no recurrence or metastasis 5 years after surgery.

1. Introduction

Koutlas et al. named sclerosing odontogenic carcinoma (SOC) as a primary intraosseous carcinoma of the jaw with histologic features which have not been reported previously [1]. It is an extremely rare tumor with only 9 reports since the initial report by Koutlas et al. As SOC is considered to be a locally aggressive odontogenic neoplasm, which is characterized by a diffuse sclerotic stroma and infiltration of numerous thin lamellar or small nests similar to epithelial cells. Mitosis is rare in this tumor. The features of this tumor include skeletal muscle invasion and perineural infiltration, as seen in malignant tumors, but cellular atypia is poor. This tumor was classified as an independent odontogenic carcinoma in the most recent World Health Organization classification [2]. However, it is unclear whether this is a malignant tumor or a morphological variation of an odontogenic carcinoma.

We experienced a tumor that showed invasion of the mandibular anterior region. The features of this tumor were not compatible with the features of any of the epithelial odontogenic tumors described in both the World Health Organization classification [2,3] and the AFIP Atlas [4].The tumor was eventually diagnosed as SOC. Here, we present this rare SOC in the midline of the mandible.

2. Case report

A 68-year-old woman showed rapid swelling of the mandibular anterior region without pain since the last 3 months. She had no paresthesia in the lower lip and the mental skin. Clinical findings included mild bulging of the mental region and obvious bulging of the mandibular anterior gingiva. The gingival mass measured 25????????25???mm with ulceration of the overlying mucosa. The tumor was fixed to the alveolus (Fig. 1). All 6 incisors were considered vital, and none of the teeth showed displacement and mobility. There was no bulging of the lingual alveolar gingiva, and the area appeared normal. Cervical lymphadenopathy was not observed. There were no radiography findings consistent with the clinical findings in orthopanoramic images. Computed tomography showed a radiolucent lesion around the root of the central incisor, which was accompanied by labial cortical bone resorption. There was no absorption of the root. The lingual cortical bone appeared healthy (Fig. 2A and B). On magnetic resonance imaging (MRI), a well-defined internal heterogeneous circular mass was noted in the mandibular anterior region. Most of the tumor showed swelling outside the bone (Fig. 2C). Based on these clinical findings, we initially suspected a malignant tumor (e.g., a squamous cell carcinoma, a malignant tumor derived from the salivary

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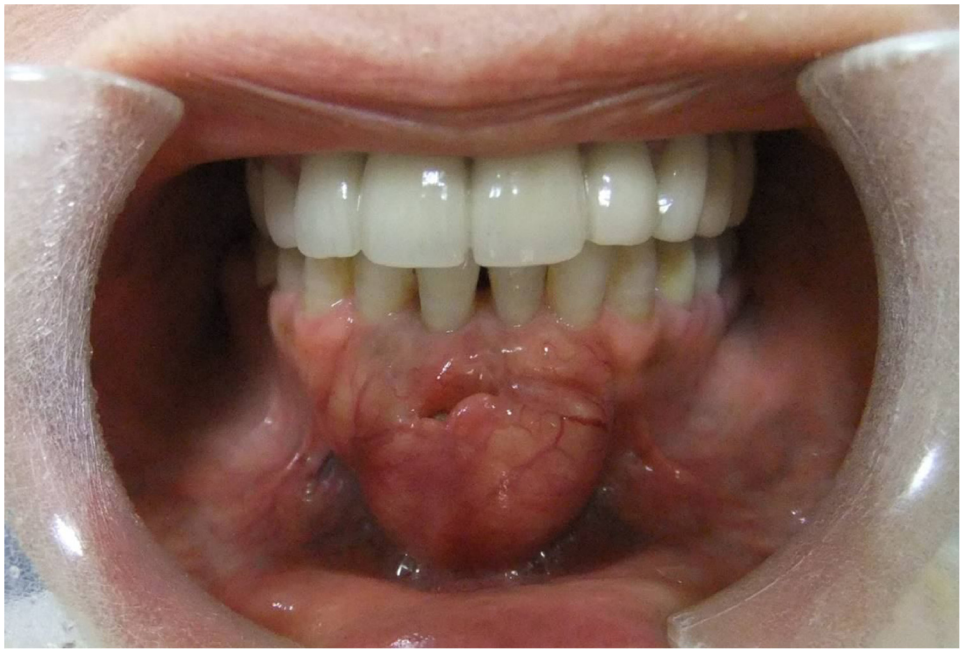

Fig. 1. Clinical findings (intraoral photograph).
Involvement of 4 incisors with bulging of the alveolar bone. The tumor measures 25????????25???mm with ulceration of the overlying mucosa. It shows stickiness and hard elasticity with ulceration of the overlying mucosa.

gland, and a malignant fibrous histiocytoma). Pathological examination of a biopsy specimen showed small rounded tumor nests in the fibrous stroma with lymphocytes. The lesional cells showed a flat eosinophil cytoplasm and a similar cyclic nucleus. Overall, cellular heteromorphism was mild, and there was no mitosis (Fig. 3A). Immunohistologically, approximately 99% of the tumor cells were positive for cytokeratin (AE1/AE3) (Fig. 3B) and were strongly positive for epithelial membrane antigen, with good staining for p63. There were no cytokeratin (CK)-5/6-positive cells, and Ki-67 was negative (1% or less). Based on these results, a benign epithelial odontogenic tumor was suspected, but biopsy findings did not lead to a definitive histological diagnosis. As the tumor was infiltrating into the jawbone, en-bloc resection of the teeth and alveolar bone was performed.

Under general anesthesia, 4 incisors and the alveolar bone were resected en bloc, whereas the lingual periosteum was preserved (Fig. 4A). The surgical site was primarily closed through suturing of the labial cavity. The basal part of the tumor originated from the alveolar bone and mainly expanded under the epithelium (Fig. 4B). On assessment of the soft tissue surgical specimen, it was found that the tumor had a capsule and expanded under the epithelium, and there was no continuity with the epithelium (Fig. 5A). Necrosis was not observed. Eosinophilic polyhedral tumor cells similar to squamous epithelial cells were scattered under the epithelium, and proliferation of various sizes and shapes of tumor nests was noted. In the deep region, tumor nests with a similar circular appearance were dispersed, and they were pressed by the sclerosing fibrous stroma. There were not observed perineural and vascular infiltration, invasion of skeletal muscle. There was no evidence of peripheral columnar cells, palisading nuclei, or a stellate reticulum (Fig. 5B and C). Cellular atypia was poor, and there was no mitosis or amyloid deposit. In the bone specimen (decalcified specimen), the same tumor nests were in the vicinity of the periodontal ligament, and they infiltrated the alveolar bone destructively (Fig. 5E and F). On immunohistological examination, the tumor cells were positive for CK-19 (Fig. 5D), and few Ki-67-positive cells were noted (approximately 2%). CK-7 positivity was not noted. Based on previous findings, clinical behavior, and immunohistological findings, the final diagnosis was SOC. As the surgical margin was negative, we performed careful follow-up without postoperative treatment. The patient has not demonstrated clinical evidence of recurrence or metastasis more than 5 years after surgery.

3. Discussion

In 2008, Koutlas et al. [1] reported 3 cases of a previously unreported histopathologic variant of odontogenic carcinoma. The tumors in these previous cases showed thin cords and small nests of cuboidal or polygonal cells, as well as prominent sclerosis of the stroma. They were called SOCs. SOC is extremely rare and only 9 cases have been reported till date. This tumor was categorized in the World Health Organization classification as an odontogenic tumor for the first time in 2017 [2]. Although it has been classified as a carcinoma, there has been no report of metastasis. Therefore, it is controversial whether this tumor is a malignant tumor, a distinct entity, or merely a morphological variation of an odontogenic carcinoma. The cytological characteristics of a malignant tumor, including the lack of heterotypic mitosis and presence of muscle and perineural invasion, are noted in this tumor. The histopathologic differential diagnosis includes normal odontogenic arrest, odontogenic fibroma, desmoplastic ameloblastoma, squamous odontogenic tumor (SOT), calcifying epithelial odontogenic tumor (CEOT), epithelial-rich variant of central odontogenic fibroma (COF), and clear cell odontogenic carcinoma.

A typical odontogenic tumor with local invasiveness is an ameloblastoma. The ameloblastomatous feature is tall columnar cells showing nuclear polarization, peripheral palisading, stellate reticulum, cystic degeneration, or squamous differentiation. In a CEOT, epithelial cells with an eosinophil cytoplasm exist as small islets, and the additional

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