Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology 27 (2015) 693–697

Contents lists available at ScienceDirect

Journal of Oral and Maxillofacial Surgery, Medicine, and Pathology

journal homepage: www.elsevier.com/locate/jomsmp

Case Report

Primary intraosseous squamous cell carcinoma arising from ameloblastoma of the mandible: A case report

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ARTICLE INFO

Article history:
Received 2 September 2014
Received in revised form 26 January 2015
Accepted 20 February 2015
Available online 20 March 2015

Keywords:
Primary intraosseous squamous cell carcinoma
Odontogenic tumor
Ameloblastoma

ABSTRACT

Primary intraosseous squamous cell carcinoma (PIOSCC) is a rare odontogenic malignant tumor, and the origin is considered remnant odontogenic epithelium, odontogenic cyst, and rarely benign odontogenic tumors. We report an extremely rare case of PIOSCC arising from ameloblastoma.

A 62-year-old man complained a large hard mass at the sublingual to the submental region. The computed tomography images revealed a large multilocular soft tissue mass extending into the bilateral molar areas with expansion of the mandible and also into the subcutaneous layer at the submental and the submandibular regions. He had received a curettage treatment of unicystic ameloblastoma in the right mandible 23 years ago. Malignant odontogenic tumor was suspected, and sectional mandibulectomy following reconstruction using a titanium plate and pedicle latissimus dorsi flap were performed. Histological examination disclosed a close correlation of intraosseous growth of squamous cell carcinoma and ameloblastoma. PIOSCC arising and dedifferentiated from long term existed ameloblastoma was mostly considered. There has been no recurrence and no metastasis for more than five years after the surgical treatment.

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1. Introduction

Primary intraosseous squamous cell carcinoma (PIOSCC) is a rare odontogenic carcinoma which is considered to be arising from odontogenic epithelial remnants during odontogenesis, epithelial lining of odontogenic cysts, and benign odontogenic tumors [1–3], although the pathogenesis has been still controversial. PIOSCC arising from ameloblastoma is extremely rare, and there have been three reported cases [4–6]. There was only one report showing coexistence of squamous cell carcinoma (SCC) and ameloblastoma in the same histological specimen [6].

The present case showed intraosseous growth of well-differentiated SCC in the mandible where ameloblastoma had been existing consecutively more than 20 years, and the coexisting of SCC and ameloblastoma with histological close correlation. There was no histological evidence of malignant transformation of the ameloblastoma such as secondary ameloblastic carcinoma in the specimen. The pathogenesis of PIOSCC arising from ameloblastoma is discussed by comparing with previously reported cases.

2. Case report

A 62-year-old man visited our hospital with a complaint of a large painless swelling at the lower face. He had medical history of curettage of ameloblastoma limited in the molar to the premolar region of the right mandible 23 years ago (Fig. 1A), and the specimen revealed findings of unicystic ameloblastoma histologically (Fig. 1B). Periodical follow-up was ceased more than 20 years ago after the curettage treatment. He had noticed the swelling of the anterior alveolar area of the mandible for three years. There was no symptom of pain and hypoesthesia. A hard

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http://dx.doi.org/10.1016/j.ajoms.2015.02.004
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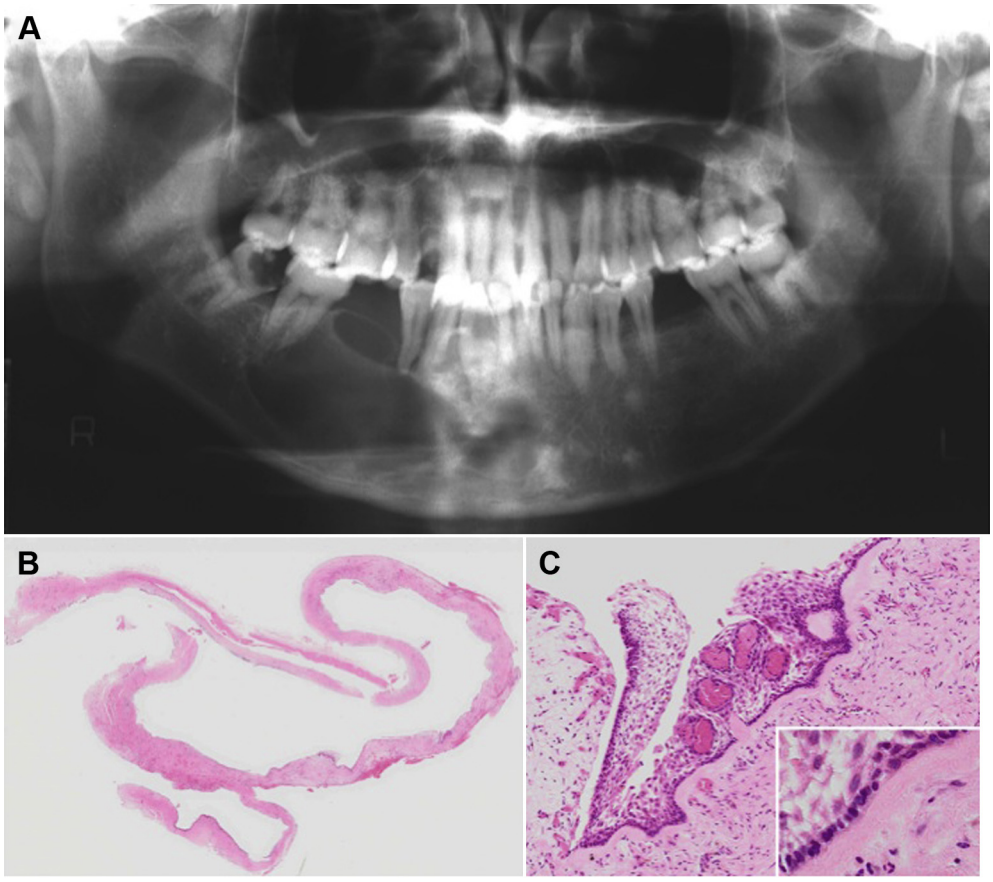

Fig. 1. Panoramic tomography image shows a radiolucent lesion at the right mandible (A), and histological feature of unicystic ameloblastoma showing cystic wall (B; HE, ×1.5) and a part of the lining epithelium shows biphasic cuboidal basal cells with nuclear palisading and stellate inner cells with thick hyalinized basement membrane (C; HE, ×10, insert, ×20).

mass of 80 mm × 70 mm × 50 mm was noted at the anterior alveolar to the mental region, and covering alveolar mucosa showed normal color with tooth indentation (Fig. 2A). There was pus and keratin debris discharge from skin of the submental region (Fig. 2B). Panoramic tomography revealed multilocular radiolucent area in the right mandible and extending into the left side with expansion and destruction of the cortical bone (Fig. 2C). The computed tomography (CT) images showed the mass with low density as soft tissue involving the subcutaneous layer at the submental to the submandibular region (Fig. 2D).

The biopsy specimen from the anterior alveolar region revealed an epithelial tumor forming follicular nests composed of columnar and cuboidal-shaped basal cells and acanthomatous inner cells regarded as ameloblastoma. Although there was no cellular atypia histologically, rapid growth of the tumor after the biopsy suggested a possibility of malignant odontogenic tumor (Fig. 3A and B). Sectional mandibulectomy between the bilateral mandibular angles, resection of the submental and the sublingual tissues, and the bilateral submandibular neck dissection were performed under the general anesthesia. The mandible and the submental skin were reconstructed using a titanium plate and the pedicle latissimus dorsi flap (Fig. 3C and D).

Histologically, the resected specimen revealed invasive growth of well-differentiated SCC forming nests and sheets with marked keratinization in the mandibular bone extending into the submucosal and the subcutaneous layers (Fig. 4A and B). There was no histological evidence suggesting mucosal origin of SCC in the specimen. The carcinoma cells show marked nuclear pleomorphism with conspicuous nucleoli and atypical mitotic figures (Fig. 4B). There was follicular proliferation of ameloblastoma closely located to the SCC nests and fused each other in parts (Fig. 4C and E). The ameloblastoma nests were composed of columnar basal cells and inner stellate cells with occasional granular and acanthomatous changes, and without cellular atypia and mitotic figure (Fig. 4D). Immunohistochemically, Ki-67 positive cells were in the most of peripheral portion of the SCC nests, while a few in the ameloblastoma (Fig. 4F). According to the histological findings, PIOSCC arising from ameloblastoma was diagnosed, most likely. There was no metastatic tumor in the neck lymph nodes. Postoperatively, there has been no complication, recurrence, and metastasis for more than five years after the surgery.

3. Discussion

PIOSCC is defined as an intraosseous carcinoma, but it is difficult to distinguish from mucosal SCC in a case of advanced stage showing destruction of cortical bone and fusion to oral mucosa [1]. Although the present case is regarded as an advanced stage of PIOSCC since evidence of destruction of the mandibular cortical bone and the tumor extension into the subcutaneous layer beyond the mandibular bone, the covering alveolar mucosa showed normal color and no histological finding suggesting mucosal origin in spite of marked expansive swelling of the mass.

PIOSCC is subcategorized into three groups according to the origin; (1) solid type, considered to be derived from odontogenic epithelial remnants such as the periradicular epithelial rests of

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