Squamous odontogenic tumor: a case report and review of literatures

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Abstract

A squamous odontogenic tumor (SOT) is an epithelial originated benign tumor. It has been rarely reported and most was intramural type. We observed a case of SOT in the mandible. It was associated with the odontogenic cyst. It was shown positive to pancytokeratin and p53. Considering that the case was free from recurrence for 5 years after surgery, p53 positive did not seem to be related to the prognosis of the disease.

Squamous odontogenic tumor (SOT) is a rare disease which is believed to originate from Malassez' epithelial rests of the periodontal membrane[7]. Gender or site predilection of SOT has not been established. Some lesions can be shown in juxtaposition to tooth roots. Most lesions remain smaller than 2 cm[6]. In our case study, however, half of the left mandibular ramus was involved. The exact pathogenesis is still unknown.

Because of the rarity of SOT, little is known about the immunolocalization of pancytokeratin and p53. In particular, the expression of p53 has been reported as a prognostic indicator[4]. In this paper we are reporting the following case of SOT, including results of an immunohistochemical study.

CASE REPORT

Because of a swelling on his left mandible, an 18-year-old male was referred to the Department of Oral and Maxillofacial Surgery, Hallym University on November 12, 2001. The onset occurred 3 weeks prior, and the apparent cause was a blow from a fist. He visited a local clinic and they did not find any pathologic conditions. However, the swelling did not subside. A radiograph showed a radiolucent lesion in the left mandibular ramus and a fracture in the area of the parasymphysis (Fig. 1). The third molar in the lesion was congenitally missing. As the cystic lesion seemed to be multilocular, it was clinically diagnosed as ameloblastoma. The operation was done via intraoral incision. For the fracture, conventional rigid fixation was done with mini-plates and screws. The bony window (size: 5.0×3.0 cm) in the left mandibular ramus was made with a reciprocating saw. The mass was composed of thick lining tissues and was enucleated. The cortical bone was readapted and fixed with miniplates and screws. Post-operative care was taken as usual. Opening of the mouth was postoperatively restricted for 2 weeks in order to prevent re-fracture of the mandible. There was no recurrence after 60 months and the operation site had healed completely. Microscopically, the lesion was composed of numerous islands of squamous epithelium in a fibrous tissue (Fig. 2). In part, the epithelial islands showed microcystic degeneration. The size of some epithelial islands was large and they were separated from each other. We could not find any columnar cells that are typically seen in ameloblastoma. No calcified tissues were found. The final diagnosis was SOT. Monoclonal antibodies from a mouse were raised against a peptide mapping at the amino terminus of pancytokeratin (Clones AE1/

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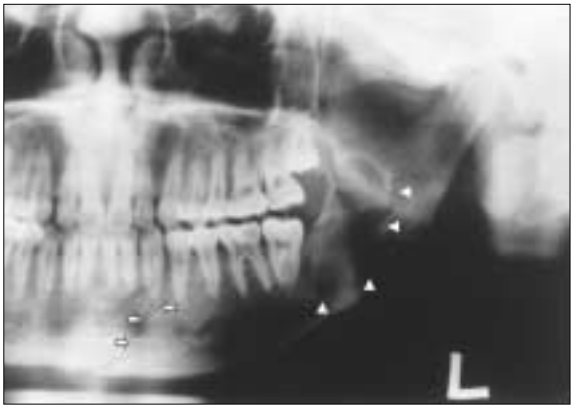

Fig. 1. Radiograph showing a well-defined multilocular radiolucency without the left mandibular third molar (arrow heads). The fracture line was visible in the parasymphysis area (arrow).

AE3/PCK26) and p53 of human origin and purchased from Ventana (Tucson, AZ, USA) and Zymed (San Francisco, CA, USA), respectively. The technique of immunohistochemical staining was well described in our previously published paper[9]. The sections were counterstained with Mayer's hematoxylin. Every epithelial component proved pancytokeratin positive (Fig. 3). Cells positive to p53 were also frequently observed. The squamous epithelial island showed many p53 positive cells which were localized in the suprabasal layer (Fig. 4).

DISCUSSION

The first report of SOT was by Pullon et al. in 1975[7]. In their report, it was uncertain whether the tumor was a

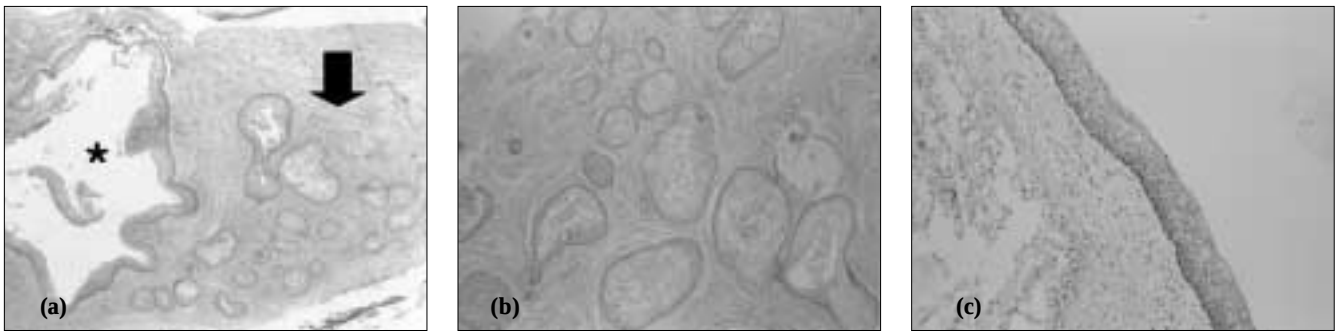

Fig. 2. (a) Simultaneous occurrence of an odontogenic keratocyst (*) and squamous odontogenic tumor (arrow) were observed. (Hematoxylin-eosin stain; original magnification ×10). (b) Epithelial islands showing typical features of the squamous odontogenic tumor. (Hematoxylin-eosin stain; original magnification ×40). (c) Some lining epithelium in large cystic cavity showing even thickness and parakeartinization like the odontogenic keartocyst. (Hematoxylin-eosin stain; original magnification ×100).

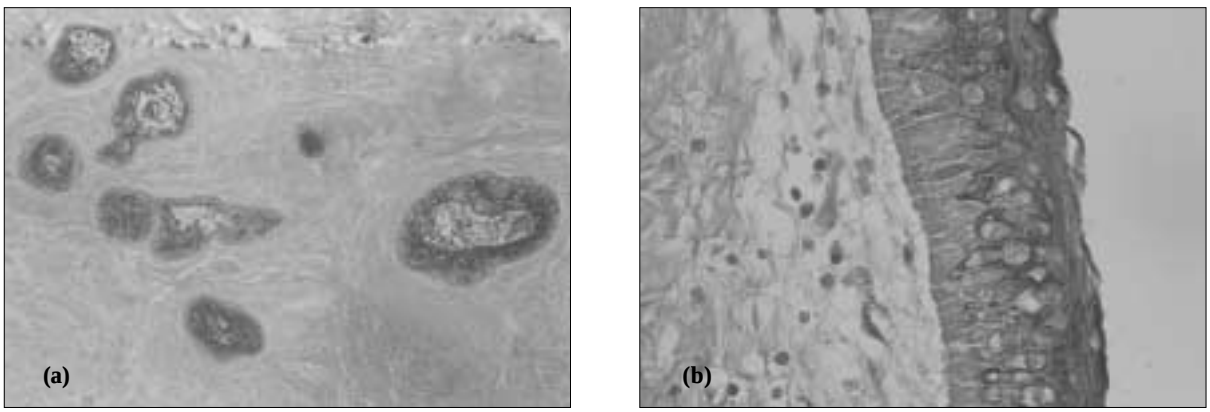

Fig. 3. Immunohistochemical findings to pancytokeratin. (a) All epithelial island showed pancytokeratin positive. (Mayer hematoxylin counterstained; original magnification ×40). (b) The lining epithelium of large cystic cavity also showing the same pattern. (Mayer hematoxylin counterstained; original magnification ×400).

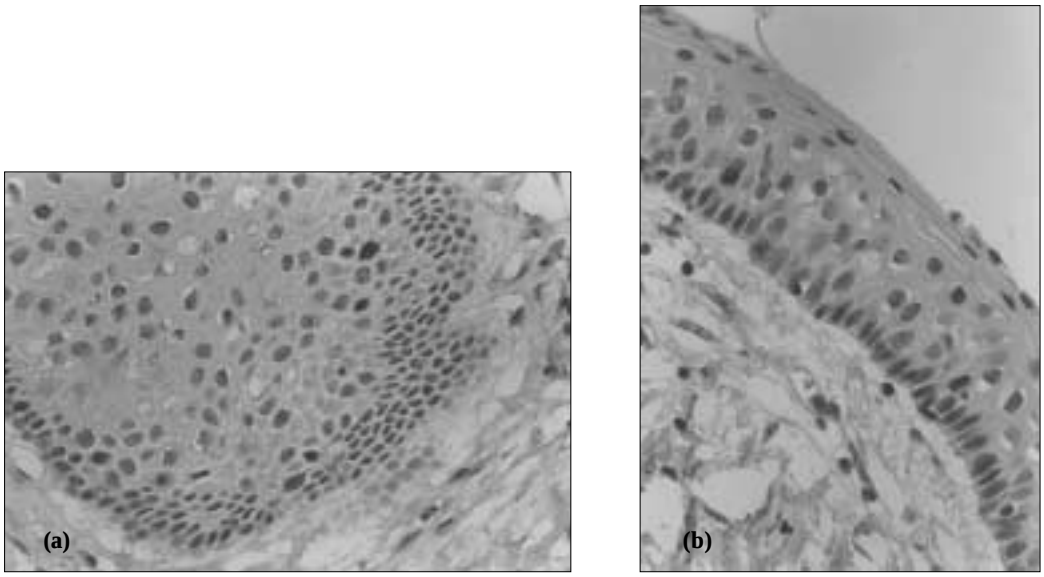

Fig. 4. Immunohistochemical findings to p53. (a) Some epithelial cells have p53 positive nucleus. (Mayer hematoxylin counterstained; original magnification ×400). (b) The immunopositive to p53 was also observed in odontogenic keratocyst-like area. (Mayer hematoxylin counterstained; original magnification ×400).

benign neoplasm or a harmatoma. Our samples showed several large cystic lesions and multiple epithelial islands. This is well correlated with the mural type of SOT. Some SOTs have been reported as mural growths within the walls of odontogenic cysts and are regarded as examples of in situ SOTs[11]. Some islands of squamous epithelium have shown cystic degeneration. The radiographic findings were similar to benign epithelial odontogenic tumors or odontogenic cysts. First, a diagnosis of odontogenic cyst could be considered. However, odontogenic cyst does not have multiple epithelial islands as found in our case study (Fig. 1). Odontogenic cyst may include proliferations of cells like squamous islands but are less dominant[8]. Second, ameloblastoma could be considered. The radiographic pattern had a "soap-bubble" appearance, which was similar to ameloblastoma. However, the multiple epithelial islands did not show the peripheral polarization of epithelium seen in ameloblastoma. Third, calcifying epithelial odontogenic tumor (CEOT) could be considered. Most CEOT is found in the posterior mandible and shows multiloculated radiolucency[6]. Our case study, however, did not show the calcification that is frequently found in CEOT. Other lesions that must be considered for differential diagnosis are adenomatoid odontogenic tumor, epithelial odontogenic ghost cell tumor, ameloblastic fibroma, and keratoameloblastoma. Slootweg reported that the immunopositive to p53 was commonly detected in ameloblastoma, odontogenic keratocyst, and odontogenic carcinoma[10]. However, Carvalhais et al reported that it was not detected in odontogenic tumors and cysts[1]. Thus, immunopositive to p53 may be controversial in odontogenic tumors. Cytokeratin has been used for differential diagnosis of epithelial origin tumor from mesenchymal origin tumor. The detection of specific cytokeratin may be helpful for the diagnosis of odontogenic tumor. Cytokeartin 13 is positive in the ameloblastoma, and cytokeratin 14 is positive in the adenomatoid odontogenic tumor[2]. However, the epithelial components of CEOT are positive to vimentin[2]. The treatment of choice was reported as a conservative surgical method and recurrence was quite rare[6,11]. However, it might infrequently precede aggressive behavior. A case of SOT was reported to have transformed into intraosseous squamous cell carcinoma[4]. In this case, p53 positive was reported as a prognostic indicator[4]. Our case study also showed p53 immunopositive (Fig. 4). Expression of p53 in many reactive lesions is unlikely to be the consequence of gene mutations[5]. It may indicate physiological expression[3]. Our case has been free from recurrence until now. No evidence of malignant transformation has been discovered. Therefore, p53 positive in this case may not be of prognostic significance. As the clinical data related to p53 expression in SOT are limited, our conclusion is a

preliminary one. Close follow-up and further study will be required.

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