PERIPHERAL AMELOBLASTOMA OF THE
Buccal Mucosa: WITH A REVIEW
OF THE LITERATURE

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Received for publication, June 25, 1990

SUMMARY: A peripheral ameloblastoma arising on the buccal mucosa is
exceedingly rare. Only three document cases have been reported, and we present one
additional case of this tumor. In this case, the lesion repeatedly occurred in the same
location in spite of twice local excisions. The first lesion was diagnosed as basal cell
carcinoma with ameloblastomatous features. The pathological findings of the lesion were
palisading of the peripheral cells of epithelial masses and structures resembling stellate
reticulum, which could also be observed also in peripheral ameloblastomas. The second
lesion was diagnosed as basal cell adenoma, and the third as peripheral ameloblastoma.
The histogenesis of peripheral ameloblastomas and intraoral basal cell carcinomas are
discussed, especially with reference to the lesions of the buccal mucosa.

INTRODUCTION

Ameloblastoma is a odontogenic neoplasm
that most often occurs in bone, and its origin
has been accepted to be one of the following
sources: (1) the developing enamel organ, (2) cell
rests of the enamel organ, (3) epithelium of
odontogenic cysts, (4) basal cells of the surface
epithelium, or (5) heterotopic epithelium, as in
lesions that occur at extraoral sites (10).

The histogenesis of the peripheral ameloblastoma is controversial. The peripheral amelo-
blastoma is described as a neoplasm arising in soft tissue overlying tooth bearing region, not
including the buccal mucosa, the lips, the
craniofaryngeal duct, or the tibia (1). On the
other hand, Shiba emphasized (12) that
ameloblastomas arising in buccal mucosa or
labial mucosa should come within the same
category of peripheral ameloblastoma, and
indeed three cases of ameloblastoma arising in
the buccal mucosa has been described (2, 7, 13).
And it is generally accepted that peripheral
ameloblastoma arises from the basal cells of the
oral mucosa of from remnants of the dental
lamina (3).

We herein report a fourth case of a peripheral
ameloblastoma arising in the buccal mucosa of
which histological feature was confused with
basal cell carcinoma, and review literatures
concerning the histogenesis of the peripheral
ameloblastoma and the differentiation from
basal cell carcinoma.

CASE REPORT

A 46-year-old Japanese man was referred to
the Otolaryngology Clinic at our hospital for the evaluation of a raised, tender, friable mass on the right buccal mucosa. The lesion had been present for a few months, and except for the tenderness it was asymptomatic. The clinical diagnosis was basal cell carcinoma. After surgical excision, the microscopic examination revealed that the lesion was mainly composed of a superficial proliferation of epithelial islands arising from overlying surface epithelium (Fig. 1), and individual islands were sometimes attached to the basilar layer of the overlying epithelium (Fig. 2). The outer layers of the islands consisted of palisaded columnar cells with their nuclei polarized away from the basement membrane. In the central area of the lesion, there was a stellate reticulum arrangement with occasional acanthomatous areas. Based on these histological findings, a diagnosis of basal cell carcinoma with ameloblastomatous features, was made.

Seven years after the initial treatment, a tumor occurred at the right buccal mucosa near the maxilla. The second surgical excision was performed, and the lesion was diagnosed as basal cell adenoma (Fig. 3) by another pathologist.

Six years after the second operation, a third lesion was noted at the same location as the previous second lesion. The third surgical excision of the tumor was done. A diagnosis of peripheral ameloblastoma, follicular type, was made (Fig. 4).

These tumors were immunohistochemically examined in paraffin-embedded tissues by the ABC methods previously described (6). Lectins such as Con A, UEA-I, WGA, PNA, and SBA (EY Lab. INC) and antibody for keratin (DAKO) were used. Immunohistochemical results are shown in Table 1. The presence of Lectin

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**Fig. 1.** Islands and nest of tumor epithelium lying below the surface epithelium (Hematoxylin and Eosin stain ×100).

**Fig. 2.** Island of ameloblastomatous tissue in continuity with the surface mucosa (Hematoxylin and Eosin stain ×300).

**Fig. 3.** Lesion diagnosed as basal cell adenoma: Similarity to the ameloblastoma (Hematoxylin and Eosin stain ×30).

**Fig. 4.** Peripheral ameloblastoma, follicular type. The epithelium is arranged in discrete islets (Hematoxylin and Eosin stain ×30).
binding and keratin was slightly more remarkable in the inner layers than in the outer layers of the lesion (Fig. 5).

**DISCUSSION**

The peripheral ameloblastoma is the rare tumor which is usually found in the mandibular molar area (14) and in the maxillary gingiva (11). Only three cases of the peripheral ameloblastoma arising from buccal mucosa have been reported (2, 10, 13) (Table 1).

The histologic appearance of the peripheral ameloblastoma is almost the same as that of the central ameloblastoma (12, 14). The basal cell adenoma may resemble the ameloblastoma (13). The histologic features of the second lesion of the present case were similar to that of peripheral ameloblastoma, follicular type, such as the third lesion. The first pathological diagnosis of the present case was reported as basal cell carcinoma with ameloblastomatous features. We reviewed the histology of the first tumor and we could diagnose it as peripheral ameloblastoma, basal cell type. Simpson reported (11) a case of basal cell carcinoma with ameloblastomatous lesion as our case, describing that the histologic appearance was not exclusive to ameloblastoma. Thus, the differentiation between the peripheral ameloblastoma, especially acanthomatous or basal type, and the intraoral basal cell carcinoma is sometimes confused.

Greer and Hammond (5) believed that the peripheral ameloblastoma exhibits central polarization of the nuclei of the peripheral cells, while the true intraoral basal cell carcinoma shows a sharp demarcation between the peripheral cells and the central reticular cells. On the other hand, Gardner (4) considered these two lesions to be indistinguishable. The ultrastructural findings of previously reported cases (5) are rather nonspecific. The immunohistochemical examination of this present case showed almost the same findings as those of the case that was reported by Yamamoto et al. (14). They stated that immunohistochemical study disclosed no significant differences between peripheral ameloblastoma and basal cell carcinoma. We suppose that most of the authors (4, 9, 11, 13) consider the peripheral ameloblastoma and the oral basal cell carcinoma as the same lesion.

Lucas indicated that the possible origin of the tumor was the oral mucosa or cell rests (8). In many cases including our case (Fig. 2), there is an epithelial continuity between the tumor cells and the overlying basal epithelial cells. Therefore, it is not surprising that peripheral ameloblastoma or intraoral basal cell carcinoma arise from the basal cell layer of the oral mucosa overlying the alveolar processes (3, 8, 13).

However, it is difficult to explain the origin of peripheral ameloblastoma arising in the buccal mucosa on the basis of dental lamina rest proliferation, because of the rare occurrence of dental lamina rest in such a location. Although the possibility of salivary gland (rests

| Table 1. Lectin binding and keratin in peripheral ameloblastoma |
|----------------------|------|------|------|------|------|
|                      | Con A | UEA-I | WGA | PNA | SBA |
| Outer layer          | -     | ±     | -   | -   | -   |
| Inner layer          | +     | ±     | ±   | ±   | ±   |

Fig. 5. Immunohistochemical staining for Con A binding. Inner cells of the lesion are more positive than outer layer (ABC Method × 300).
Table 2. Cases of peripheral ameloblastoma of the buccal mucosa

<table>
<thead>
<tr>
<th>Reference</th>
<th>Age, sex, race</th>
<th>Location</th>
<th>Continuity with basal layer</th>
<th>Recurrence</th>
</tr>
</thead>
<tbody>
<tr>
<td>2</td>
<td>63 yr, male, white</td>
<td>Left</td>
<td>Unstated</td>
<td>–</td>
</tr>
<tr>
<td>6</td>
<td>68 yr, male, Oriental</td>
<td>Right</td>
<td>+</td>
<td>–</td>
</tr>
<tr>
<td>13</td>
<td>52 yr, female, East Indian</td>
<td>Right</td>
<td>+</td>
<td>–</td>
</tr>
<tr>
<td>Present case</td>
<td>46 yr, male, Japanese</td>
<td>Right</td>
<td>+ (7 yrs later)</td>
<td>+</td>
</tr>
</tbody>
</table>

of Serres) origin or remnants of the vestibular lamina cannot be ruled out (4, 7), Woo et al. (13) consider that its origin would most probably be pluripotent cells in the overlying basal layer. Besides, they refer to the origin of the both lesions in the oral cavity: it is either from the pluripotent cells as above described or from native or ectopic epithelial rests. If this is the case, it is easy to explain the phenomenon of our case that peripheral ameloblastomas was observed in the same location after the complete local excision of basal cell carcinoma. That is to say, peripheral ameloblastoma and basal cell carcinoma of the oral cavity would be identical in origin, and the pluripotent cells could differentiate into peripheral ameloblastoma or basal cell carcinoma. Controversial matter about these lesions seem to be of semantics, since the same lesion may be observed from different aspects. We propose that pathological diagnoses should be peripheral ameloblastoma when more than half area of the lesion include follicular or plexiform pattern which resembles the enamel organ of the developing tooth germ.

Most authors have reported that peripheral ameloblastomas possess innocuous nature, and that conservative surgery is recommended. Nevertheless, Woo et al stated (13) that recurrences were observed in all cases that were treated by simple local excision of the lesion. The tumors repeatedly occurred in the same lesion of the present case in spite of twice local excision. Whether recurrences of the tumor frequently occur or not, patients should be kept under periodic observation.

REFERENCES

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