

PERIPHERAL AMELOBLASTOMA OF THE BUCCAL MUCOSA: WITH A REVIEW OF THE LITERATURE

Takashi SHINZATO¹, Takayoshi TODA¹, Masaya KIYUNA¹
Yushi HAMADA¹, Seitetsu HOKAMA¹ and Katsuji AKAMINE²

*Department of Clinical Laboratory and Laboratory Medicine¹ and Department of
Otolaryngology², School of Medicine, University of the Ryukyus, Okinawa, Japan*

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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 ameloblastoma is described as a neoplasm arising in soft tissue overlying tooth bearing region, not including the buccal mucosa, the lips, the craniopharyngeal 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 or 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



Fig. 1. Islands and nest of tumor epithelium lying below the surface epithelium (Hematoxylin and Eosin stain $\times 100$).

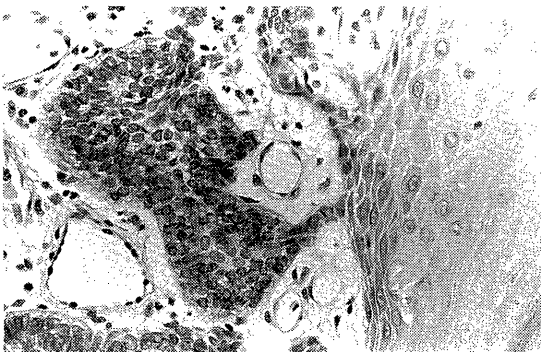


Fig. 2. Island of ameloblastomatous tissue in continuity with the surface mucosa (Hematoxylin and Eosin stain $\times 300$).

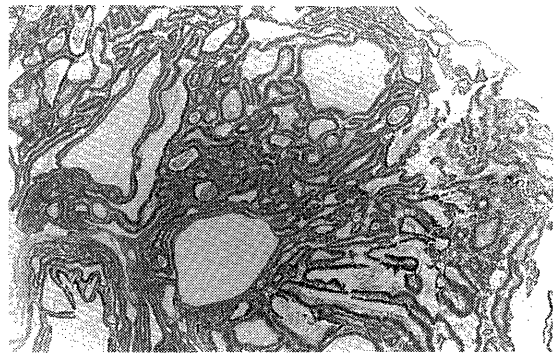


Fig. 3. Lesion diagnosed as basal cell adenoma: Similarity to the ameloblastoma (Hematoxylin and Eosin stain $\times 30$).



Fig. 4. Peripheral ameloblastoma, follicular type. The epithelium is arranged in discrete islets (Hematoxylin and Eosin stain $\times 30$).

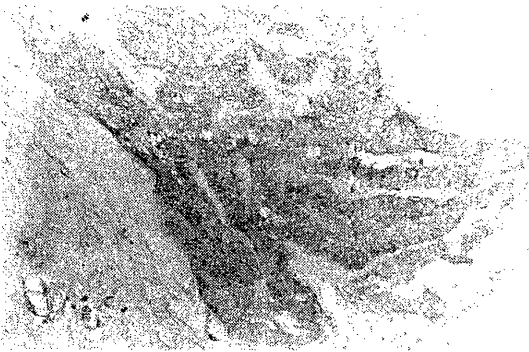


Fig. 5. Immunohistochemical staining for Con A binding. Inner cells of the lesion are more positive than outer layer (ABC Method $\times 300$).

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 | Keratin |
|-------------|-------|-------|-----|-----|-----|---------|
| Outer layer | - | ± | - | - | - | + |
| Inner layer | + | ± | ± | ± | ± | ± |

Table 2. Cases of peripheral ameloblastoma of the buccal mucosa

| Reference | Age, sex, race | Location | Continuity with basal layer | Recurrence |
|--------------|----------------------------|----------|-----------------------------|--------------------|
| 2 | 63 yr, male, white | Left | Unstated | — |
| 6 | 68 yr, male, Oriental | Right | + | — |
| 13 | 52 yr, female, East Indian | Right | + | — |
| Present case | 46 yr, male, Japanese | Right | + | + (7 yrs later) |

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.

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