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內文：

### Abstract

1. Gingival tumors of what appear to be peripherally located **intraosseous ameloblastoma(IA)** arising from the **alveolar bone surface** have often been confused with **peripheral ameloblastoma(PA)**causing **resorption of the underlying bone**
2. A series of **five cases** of ameloblastoma that demonstrated a **combined PA and IA architecture**
3. The tumor commonly involved **the anterior-premolar area**, mostly in the **maxilla** and mainly in **middle-aged men**
4. The clinical presentation was **an exophytic gingival mass** inferior to which was a **small bone defect**
5. The predominant extraosseous component showed a **papillary gross surface**, reflecting the histologic proof of fusion between the **submucosal tumor** and **the surface epithelium**
6. In two cases, **recurrence was observed deep in the alveolar bone** with no involvement of the gingiva
7. From diagnostic, therapeutic and prognostic points of view, this type of IA should not be confused with PA

## Introduction

1. Ameloblastoma can be encountered in **any area of the jaws** from the body of bone through the alveolar crest to the gingiva, and between **1 and 10%** of cases are reported to occur peripherally
2. By definition, peripheral ameloblastoma does not spread beyond the **gingival submucosa** into the alveolar bone
3. The final diagnosis always rests with exclusion of a recognizable **intraony lesion** ; However, the bone defect may occur to varying degrees, when large enough
4. It is often confusing whether such lesions are primarily PA that erode into the underlying bone or if superficial intraosseous ameloblastoma that expand out into the overlying giniva

## Materials and Methods

1. **Five cases** of ameloblastoma with a **combination of PA and IA growth pattern** were retrieved from the archives of Tsurumi and Meikai University Hospital, one of which was previously reported as PA by us
2. Available clinical records were **retrospectively reviewd** and **the newly prepared hematoxylin and eosin-stained slides** were thoroughly examined through sectioning at many levels
- 3.This work was approved by the Research Ethics Committees

## Results

## 1. Clinical and Radiographic Findings

**Table 1** Clinical information

Case number	Sex/age	Location	Size (cm)	Radiographic pattern <sup>a</sup>	Treatment	Recurrence	Year of diagnosis
1	M/48	Mx (A)	2.0	Cup	SE + Cu	Yes <sup>b</sup>	1995
2	M/75	Mx (P)	1.5	SR	SE + Cu	Yes <sup>b</sup>	2000
3 <sup>d</sup>	M/28	Mn (A-P)	2.5	Cup	BR	No	2000
4	M/54	Mx (A)	1.4	Cup	BR	No	2001
5	M/57	Mx (M)	3.0	SR	BR	No	2009

(1) The patients ranged in age from **28 to 75** years with an average of **52 years** and all were **men**

(2) Four cases involved the maxilla and the remaining one was found in the mandible

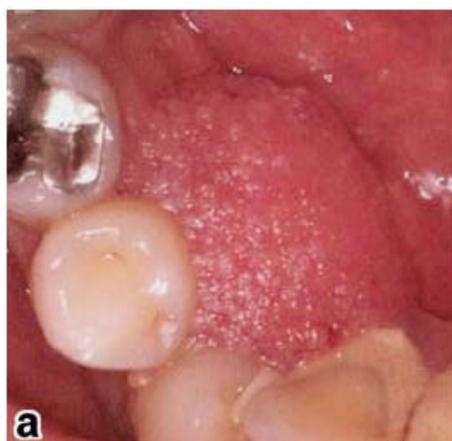
(3) With the exception of cases 5, they had a distinct predilection for **the anterior-premolar region**, 2 of which involved the **incisor area**

(4) The average size of tumors was **2.0cm**, ranging from **1.4 to 3.0 cm**

(5) The most common sign was **a painless gingival swelling** of several months' duration

(6) The lesion appeared as **a broad-based, exophytic soft-tissue mass with a granular surface**

(7) A focal ulceration was recorded in case 4



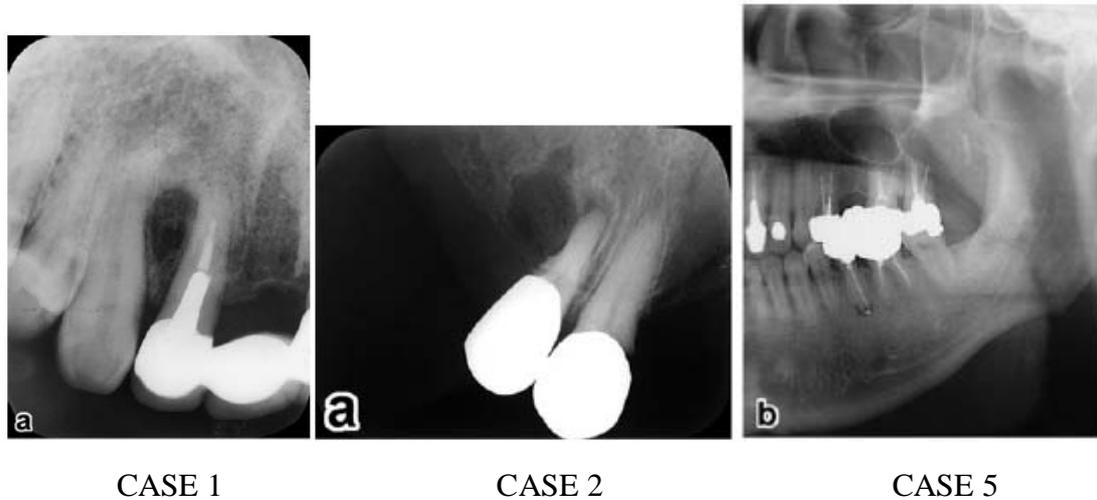
CASE 3



CASE 5

(8) On plain radiographs, cases 1, 2 and 5 showed **a deep cup-like defect** in the

marginal alveolar bone, and **an ill-defined, small radiolucency** was found in cases 2 and 5



(9) Neither **displacement** nor **resorption** was visible in the teeth roots

(10) In CT and MRI, **a true invading defect** was clearly demonstrated in cases 3 and 5



## 2. Treatment and Follow-up

(1) Cases 3, 4 and 5 were diagnosed preoperatively as ameloblastoma by **incisional biopsy** and managed by **en bloc resection**

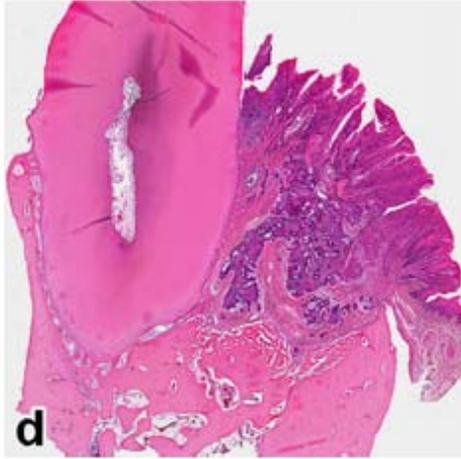
(2) Although a follow-up period is variable, ranging from **6 months to 8 years**, **none of them recurred**

(3) In cases 1 and 2 that were treated by a combination of **simple excision and curettage of tumor bed** under the clinical diagnosis of **papilloma**, **recurrence** was evident within 2 years.

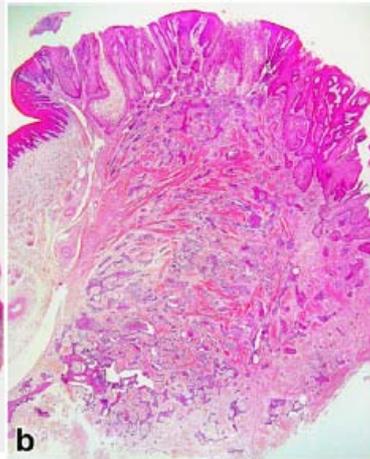
(4) After further **marginal resection** of the alveolar bone with the removal of lesional teeth, both patients were locally free of tumor for 7 years

### 3. Pathologic Finding

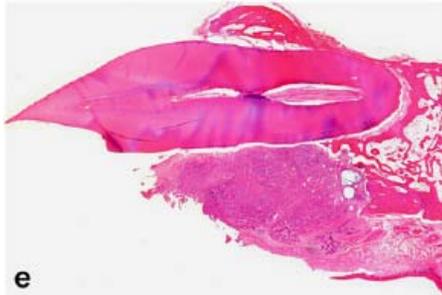
(1) In a tissue section whole mount, **extraosseous proliferations** of ameloblastoma in the gingiva were predominant.



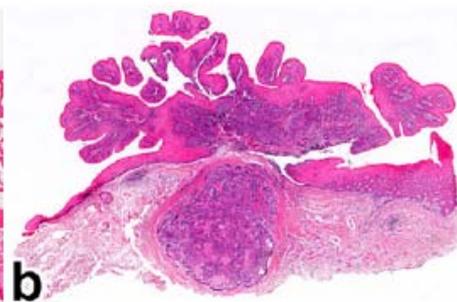
CASE 3



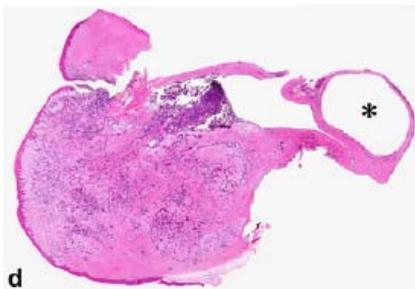
CASE 1



CASE 4

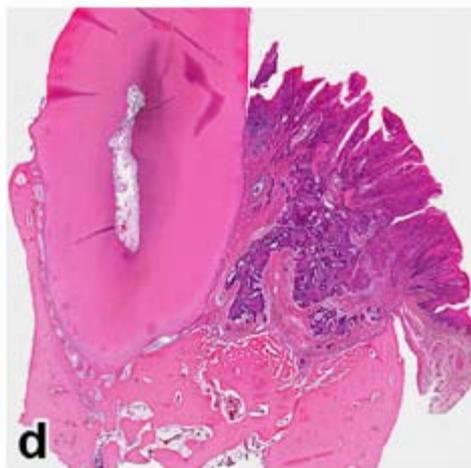


CASE 2

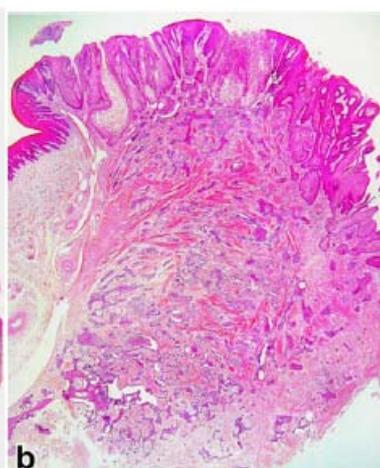


CASE 5

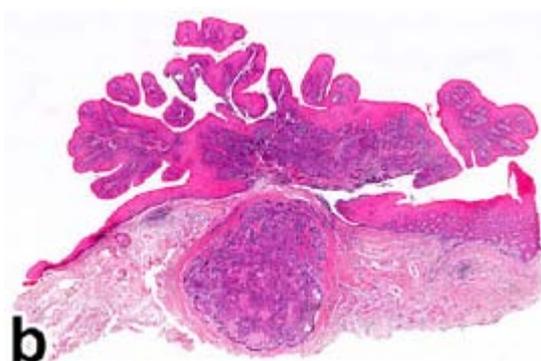
(2) All tumors fused with the surface epithelium over a wide area.



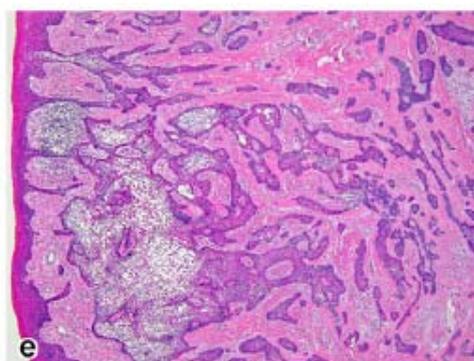
CASE 3



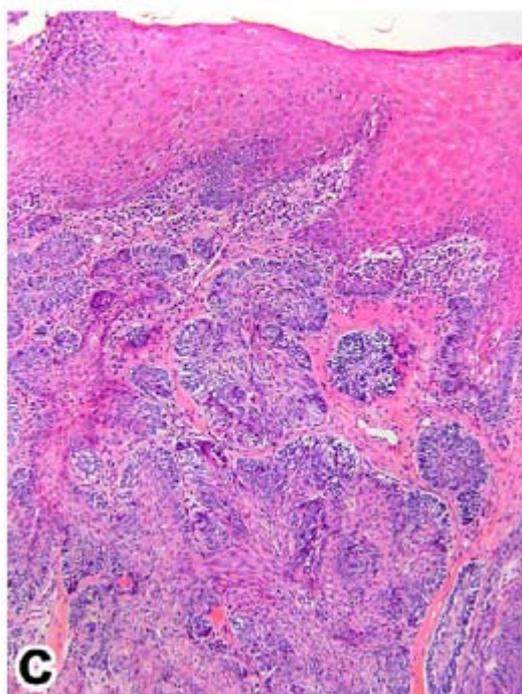
CASE1



CASE 2



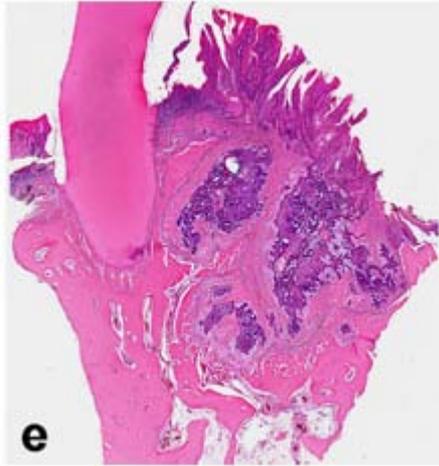
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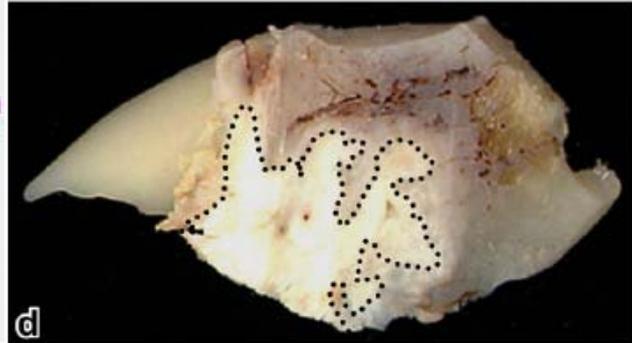
CASE 2

(3)Further examinations of the leading edge of tumor on multiple sections at different

levels revealed destructive intraosseous proliferations, mostly at the apical root part of lesional teeth.

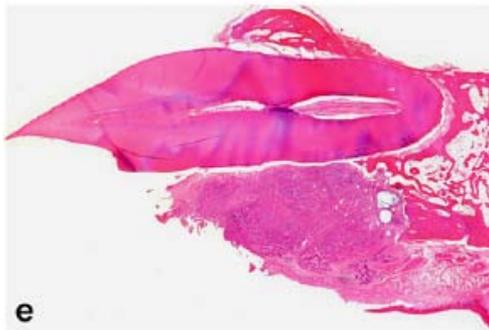


CASE 3



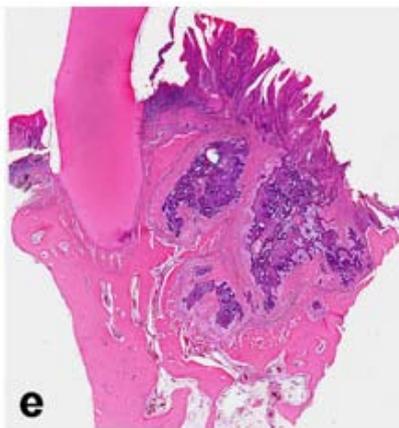
CASE 4

2

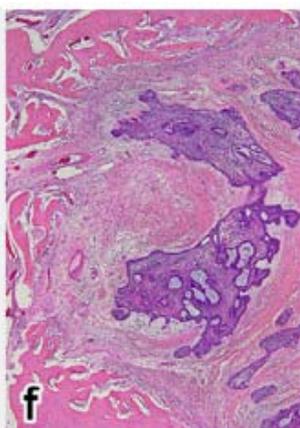


CASE 4

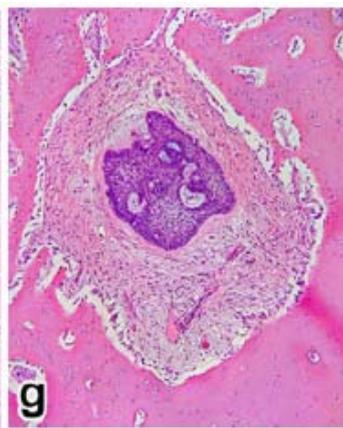
(4) Spread far beyond the apex in the surrounding alveolar bone was found in cases 3, and decalcified bone contained a few outlying tumor nests



CASE 3



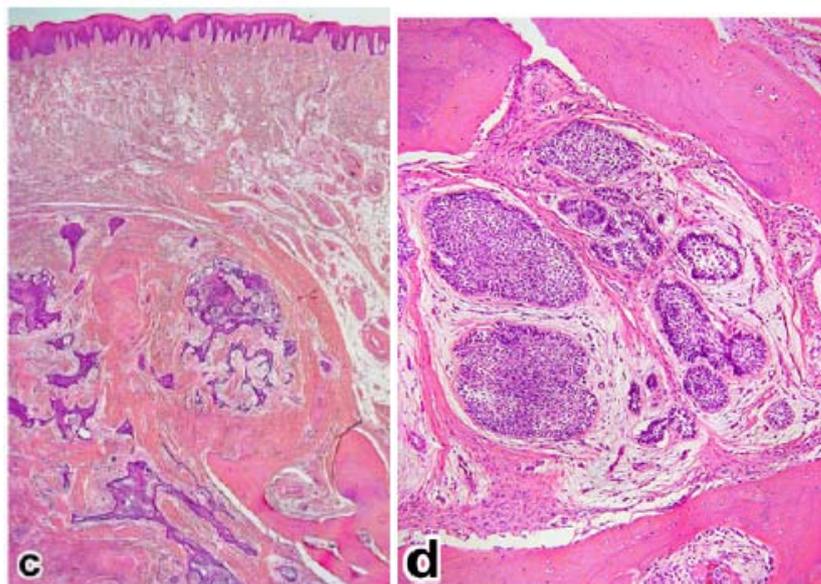
CASE 3



CASE 3

(5) Cases 1 and 2 recurred in the bone, but the surface epithelium was intact with no

neoplastic change



CASE 1

CASE 2

### Discussion

1. The surrounding alveolar bone provides more resistance to tumor growth than does the gingiva, infiltrating IA has a tendency to extend into the overlying mucosa, either directly or along the periodontal ligament, once tumor nests penetrate the bony confine.
2. As a result, such IA sometimes appears as a gingival mass lesion without causing any clinically visible expansion of the affected bone, thus producing the illusion of PA
3. Theoretically, IA would be within the bone whereas PA would be against the buccal or lingual bone. Critical to differentiating IA from PA is the identification of an intact cortical bone covering the tumor
4. Unfortunately, this decisive finding may be obscured with time through complete loss of thinner cortical plates. As with the cases described here, new imaging modalities are more successful than plain radiographs in evaluating the actual extent of the tumor in the alveolar bone. The root divergence, one of the most characteristic radiographic signs of interradicular IA, is not marked in our cases, probably reflecting their unobtrusive intraosseous growth. Since recurrence is so likely to follow too conservative an operation, the removal of unaffected bone and involved teeth as a bloc is consequently advisable.

5. With regard to the histogenesis, the rests of Malassez inside or outside the periodontal ligament space may be a potential candidate. Such epithelial residues, either in the alveolar bone surface or in the cervical or middle portion of the periodontal ligament, seem likely starting points of our cases.

6. The previous observation that ameloblastomatoid rests appeared later in adult life and tended to increase with age lend credence to the occurrence in an older age range

7. The diagnosis cannot be determined by its extraosseous/intraosseous proportion. The practical approach for the surgeons is that an identifiable intrabony lesion is curious in PA, and if observed, either radiologically or clinically during surgery, should prompt consideration of IA.

9. They also draw attention to the co-existence of insidious IA in what seems at first glance to be PA, when larger than 2 cm. If, on histologic diagnosis of clinically presumed PA, there is any doubt that the tumor has proliferated from the gingiva into the underlying bone, the pathologists should re-examine the tumor-bone interface by extensive sampling of the entire specimen.