

# A CLINICAL AND RADIOLOGIC CONSIDERATION OF AMELOBLASTOMA OF THE JAWS

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## CONTENT

- I. INTRODUCTION
- II. MATERIALS AND METHODS
- III. RESULTS
- IV. DISCUSSIONS
- V. CONCLUSION
- REFERENCES
- 국문초록

## I. INTRODUCTION

Ameloblastomas of the oromaxillary area make up 1% of all tumors of the jaws<sup>1-3</sup>, and 11% of odontogenic tumors<sup>1,2</sup>. They occur primarily in middle-aged adults, with a predilection for the posterior mandible<sup>4</sup>. The male-to-female ratio is roughly 1 : 1<sup>1,3,5-7</sup>. The most common symptom is a slow-growing painless swelling<sup>8</sup>. The exact origin of ameloblastomas is still essentially unknown. They generally are considered to originate from the enamel organ, epithelium of dentigerous cysts (or other odontogenic cysts, surface epithelial basal cells or remnants of the dental lamina, or Hertwigs sheath<sup>9</sup>). Grossly, these tumors can be solid,

cystic, or solid and cystic<sup>7,10</sup>. There are follicular, plexiform, mixed, unicystic, and desmoplastic histologic patterns<sup>1,3,4,5,11-15</sup>, with 50% of ameloblastomas being mixed<sup>5,16</sup>.

Radiographically, it exhibits an expansile unilocular or, more often, multilocular pattern with discrete margins, and association with an impacted tooth is commonly observed<sup>4</sup>.

Ameloblastoma of the jaw behaves differently from a simple cyst or tumor because it is locally invasive and tends to recur after removal<sup>17</sup>. The pre-operative diagnosis is important as the removal of this tumor is a more extensive procedure than excision of a simple cyst or tumor of similar size<sup>17</sup>. Ueno reported that the most recurrent ameloblastomas were of the follicular type that radiographically had a multilocular or soap bubble appearance<sup>18</sup>. On the other hand, most authors think that the behavior of the ameloblastoma cannot be predicted from histologic studies<sup>5,6,19</sup>. In this context, radiographic examination is important in evaluating the biologic behavior of the tumor<sup>18</sup>.

The purpose of this study was to clarify the clinical and radiologic parameters that can be used to predict the biologic behavior of ameloblastoma.

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## II. MATERIALS AND METHODS

Forty-six cases of ameloblastomas presented in this study were treated in the Department of Oral and Maxillofacial Surgery, Dental Hospital, Yonsei University, during the period from January 1979 to December 1993.

The clinical features were analyzed with respect to sex, age, site, signs and symptoms.

Radiologic type of the lesion was determined from the radiographs that were taken at the time of initial diagnosis. Unilocular or multilocular appearance was recognized on the radiographic examination of the lesions. All patients were surgically treated. Surgical specimens of all 46 cases were reviewed and also classified histopathologically according to the criteria generally used by most oral pathologists<sup>1,3,5,12-15</sup>. Recurrence of the tumor after surgery was determined based on examination of radiographs and generally, but not in all cases, on histologic examination of biopsy specimens<sup>18</sup>.

Statistical evaluation was performed with chi-square analysis to assess whether the observed frequencies differed significantly from those predicted in contingency tables.

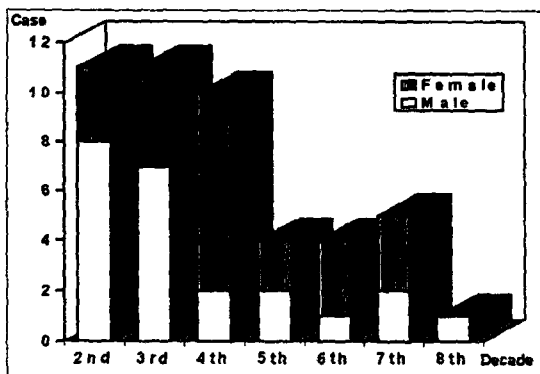


Fig 1. Sex and age distribution of ameloblastoma at time of diagnosis.

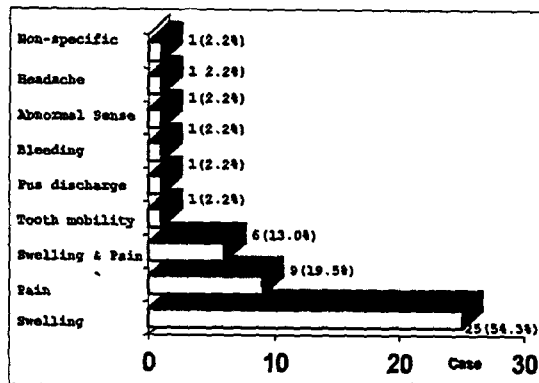


Fig 2. Clinical Signs and Symptoms

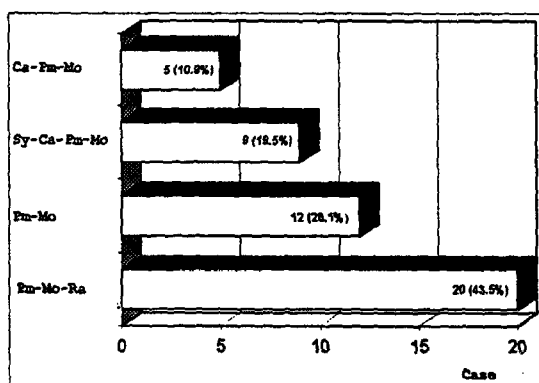


Fig 3. Sites of ameloblastoma.

## III. RESULTS

### Clinical Findings

#### Sex and Age (Fig. 1)

There was no predilection in sex distribution; sex ratio of 1 : 1 is noted. The ages of patients ranged from 11 to 71 years, with a peak in the second decade. The average age was 34 years.

#### Clinical Signs and Symptoms (Fig. 2)

Clinical records for 46 cases of intraosseous ameloblastoma were available. The most frequent sign was swelling of the face or jaw.

This sign occurred in 25 cases (54.3%). Pain occurred in 15 cases (32.5%).



**Fig. 4.** Panoramic radiograph showing multilocular lesion in the left mandibular body and ramus.



**Fig. 5.** Panoramic radiograph showing unilocular lesion in the right mandibular body and ramus area.

**Table 1.** Radiologic types and its correlation with age of patients

Radiologic type	Patient's Age(yr)							n	(%)	M.A
	10-19	20-29	30-39	40-49	50-59	60-69	70-80			
U	5	6	6	1	0	2	0	20	( 43.5)	30
M	6	5	4	3	4	3	1	26	( 56.5)	38
Total	11	11	16	4	4	5	1	46	(100.0)	34

U:Unilocular, M:Multilocular, M.A:Mean Age

### Radiologic Findings

#### Sites of Tumor(Fig. 3)

Of the 46 intraosseous ameloblastomas, 45 (97.8 %) involved mandible and one(2.2 %) the maxilla. Twenty cases(43.5 %) involved the premolar-molar-ramus area. Maxillary lesion arose in the canine-premolar-molar area.

Radiologic type of the lesion was determined from the radiographs that were taken at the time of initial diagnosis. Multilocular appearance(Fig. 4) was found in 26 cases (56.5%) and unilocular appearances(Fig. 5) in 20 cases (43.5%).

The radiologic types were assessed with respect to the age of patients(Table 1). The mean age for the patients with multilocular lesions was 38 years, and that for the patients with unilocular lesions was 30 years.

Generally, there was no correlation between radiologic types and age of patients.

### Histologic Findings

Histologic type of each tumor was subclassified according to the predominant histologic pattern into following categories: follicular, plexiform, mixed, unicystic, and desmoplastic<sup>1,3-5,11-15</sup>. If both characteristics were observed in the same case, it was classified as mixed type.

The ameloblastomas were of the follicular or mixed type in each 11 cases(23.9 %), the plexiform or unicystic type in each 10 cases (21.7 %), and desmoplastic and non-defined type in each 2 cases(4.4%). There was no case of malignant ameloblastoma. Radiologic types were also compared with histologic types(Table 2).

**Table 2.** Radiologic types and its correlation with histologic types

Radiologic type	Histologic Type						n( % )
	Follicular	Plexiform	Mixed	Unicystic	Desmoplastic	Non-Defined	
U	7	5	3	4	1	0	20 (43.5)
M	4	5	8	6	1	2	26 (56.5)
Total	11(23.9)	10(21.7)	11(23.9)	10(21.7)	2(4.4)	2(4.4)	46(100.0)

**Table 3.** Histologic types and its correlation with age of patients

Histologic Type	Patient's Age(yr)							n	M.A
	10-19	20-29	30-39	40-49	50-59	60-69	70-80		
Follicular	2	2	3	0	2	1	1	11	39
Plexiform	2	4	2	1	0	1	0	10	30
Unicystic	5*	4	1	0	0	0	0	10	23
Mixed	1	1	2	3	2	2	0	11	46
Desmoplastic	0	0	1	0	0	1	0	2	48
Non-defined	1	0	1	0	0	0	0	2	25
Total	11	11	10	4	4	5	1	46	34

\* A correlation between the histologic type and age was found by  $X^2$  test( $P<0.05$ ).

**Table 4.** Results of Therapy for ameloblastomas by radiologic type

Radiologic Type	Result of Therapy		
	Tumor-Free	Recurrence	n ( % )
U	5(71.4)	2(28.6)	7(100.0)
M	7(63.6)	4(36.4)	11(100.0)
Total	12(66.7)	6(33.3)	18(100.0)

Generally, there was no correlation between histologic and radiologic types.

The histologic types were also analyzed with respect to the age of patients(table 3).

The unicystic type was commonly found in the younger patients and the mean age for the patients with unicystic lesions was 23 years. A correlation between the unicystic type and second decade of age was found by  $X^2$  test ( $P<0.05$ ).

#### Recurrent Cases of Ameloblastoma

All patients were surgically treated. Recurrence of the tumor after surgery was

determined based on examination of radiographs and generally, but not in all cases, on histologic examination of biopsy specimens.

There were 18 patients qualified for the study only if they had been followed for more than 1 year after initial treatment(Table 4).

Follow-up ranged from 1 to 8 years, with an average age of 4 years. There were 6 cases of recurrent ameloblastoma(33.3 %) and the mean age of these patients was 32 years. Radiologically, only 2(28.6%) of 7 patients with a unilocular lesion experienced recurrence, whereas 4(36.4%) of 11 patients with a multilocular lesion had recurrence.

#### IV. DISCUSSION

Ameloblastoma occurs with equal frequencies in the two sexes. In the previous literature, more than half of the studies<sup>3,5,6,20-27</sup> reported that the ameloblastoma occurred with almost equal frequency in males and females.

The male and female ratio in the study in Korea<sup>28</sup> was 1.4 : 1. In the present series(Fig. 1), the male and female ratio was 1 : 1. In the majority<sup>3,5,20-23,26,27,29-33</sup> of studies, the average age of the patients at the time of diagnosis was between 30 and 40 years of age. In the previous Korea investigation<sup>28</sup>, the average age of the patients was 30.7 years of age and present study 34 years of age.

The ameloblastomas grow slowly, without clinical signs in the early stages<sup>18</sup>. The lesion develops gradually, and bone expansion or facial deformity appears. The majority of our patients(54.3%) initially noticed swelling(Fig. 2). Although it is generally said that pain is seldom a complaint<sup>34</sup>, 32.5% of the patient in our series complained of pain. It is not known whether the cause of the pain is the tumor or secondary infection<sup>18</sup>.

According to the reports in the literature, approximately 80% of the tumors are found in the mandible<sup>3,5,35-37</sup>, but the incidence in the present study(Fig. 3) was 97.8%, in accordance with the finding in Korea<sup>28</sup> 94.3%. The molar-ramus area is most frequently involved<sup>18</sup>. Dental epithelium, enamel organ, remnants of the dental lamina, the rests of Malassez, and the epithelial lining of odontogenic cysts are the suspected sites of origin of the tumor<sup>38</sup>. In the present study, the most commonly affected site of the mandible was the premolar-molar-ramus region, in accordance with previous findings<sup>28</sup>.

Ameloblastoma appears macroscopically as a solid or cystic lesion<sup>39</sup>. It would appear that

ameloblastoma begins as a solid tumor and gradually becomes more cystic with age<sup>3,40</sup>, probably as a result of the degenerative transformation of the tumor<sup>41</sup>. On the other hand, ameloblastomas have developed in the walls of dentigerous cysts<sup>6,13,42-44</sup>. The radiographic extent of the lesions is deceptive, and invasion of bone occurs well beyond the apparent limits of the tumor, especially with the solid variety<sup>45</sup>. The prognoses for multicystic and well-circumscribed unicystic lesions differ markedly<sup>13,46,47</sup>. Radiographic findings are important in the predilection of the clinical course of the tumor, because the unicystic type exhibits less aggressive biologic behavior than does the multicystic type<sup>13</sup>. The radiologic types of ameloblastoma in the current study were assessed with respect to the age of patients(Table 1). The mean age for the patients with unilocular lesions was more higher than that for the patients with multilocular lesions, but there was no correlation between radiologic types and age of patients, in disparity with the result of Ueno et al<sup>39</sup>. A multilocular appearance is rarely seen in a simple cyst or tumor but was present in many of the presenting cases in this series(Table 2).

In many previous reports<sup>3,5,20,23-27,29,31,48-53</sup>, the plexiform and and follicular types accounted for the vast majority of cases, and the other types were rare. The present findings that most of the tumors fell within the four categories of follicular, plexiform, unicystic and mixed types agreed well with most previous reports(Table 2). Ameloblastomas commonly contain areas with two or more different histologic patterns<sup>25,9</sup>. Most previous studies reported that the histologic typing of ameloblastomas is not clinically important<sup>5,6,19</sup>. But Ueno insisted that more clinical data on tumor behavior and histologic types need to be accumulated<sup>39</sup>. The histologic types of amelo-

blastoma in the current study were analyzed with respect to the radiologic types and the age of the patients. The histologic types had no correlation with the radiologic types (Table 2). The histologic types were also analyzed with respect to the age of patients (Table 3).

The unicystic ameloblastoma was most commonly found in the younger patients and the mean age for the patients with unicystic lesions was 23 years. The mean age for the patients with follicular, mixed and desmoplastic lesions was significantly higher than that of patients with unicystic lesions. A correlation between the unicystic type and second decade of age was found by  $X^2$  test ( $P < 0.05$ ). Our results confirm previous observations<sup>10,13,54</sup> that unicystic ameloblastomas tend to occur at an earlier age than the multicystic or solid counterparts. Possible reasons for this remain obscure. The occurrence of cases in the 6th and 7th decades has not previously been reported. Again reasons for this are not clear, but smaller, non-expansile lesions may possibly remain undiagnosed for some years<sup>55</sup>. Perhaps the most important consideration regarding unicystic ameloblastomas is that of biologic behaviour. It has been widely stated<sup>10,13,14,56</sup> that these lesions are less aggressive than their solid or multicystic counterparts and should be treated by enucleation or curettage. However, Gardner<sup>56</sup> has pointed out that there is a difference in biological behaviour between those lesions that are simply cystic (Group 1) or show intraluminal proliferation (Group 2) and those in which the epithelium penetrates and breaches the fibrous wall, therefore having the capacity to invade cancellous bone (Group 3)<sup>55</sup>. Unfortunately we have inadequate follow up in our series of cases and are not able to support this contention with convincing data regarding recurrences. In any event such follow up data

might be of little value in answering the question of whether some unicystic ameloblastomas may be handled conservatively since most of the lesions in this series were treated by radical surgical excision before they become recognised as a distinct entity<sup>55</sup>.

Considering the treatment of the ameloblastoma as well as other tumors, the suggestion that selection of the most effective treatments must be determined by various factors such as the patient's general health, specific anatomical location of the tumor, extent of the lesion, and aesthetic considerations<sup>16</sup>, is noteworthy. All patients in the present study were surgically treated.

Recurrence of the tumor after surgery was determined based on examination of radiographs and generally, but not in all cases, on histologic examination of biopsy specimens.

The diagnosis of recurrent ameloblastoma usually depends upon the presence of a corticated translucent area which increases in size seen on serial radiographs taken over a period of years<sup>17</sup>. There were 18 patients qualified for the study only if they had been followed for more than 1 year after initial treatment (Table 4). Follow-up ranged from 1 to 8 years, with an average age of 4 years.

There were 6 cases of recurrent ameloblastoma. The mean age of these patients was 32 years. Radiologically, the recurrent lesions of multilocular appearance were 4 cases (36,4%) of total 11 lesions. Irrespective of treatment modality, Robinson and Martinez (1977) found an overall 67% recurrence rate; Gardner and colleagues (1987) reported a 71% recurrence rate. When planning a definitive operation for ameloblastoma, the surgeon must not only consider the clinical subtype of the lesion but also its extent, including soft-tissue involvement<sup>57</sup>. Large lesions of the ascending ramus of the mandible are particularly difficult

to manage, and, when inadequately treated, have a high rate of recurrence<sup>58</sup>. CT can be used in these cases to clearly demonstrate tumor extension and thus facilitate adequate treatment planning and management<sup>57</sup>.

## V. CONCLUSIONS

A clinical and radiologic study of 46 intrabony ameloblastomas were undertaken from the files of the Department Oral and Maxillofacial Surgery, Dental Hospital, Yonsei University for the years January 1979 through December 1993. The results were as follows:

1. In the clinical findings, the mean age of patients was 34 years, with no sex predilection noted and the most frequent sign was swelling of the face or jaw.
2. In the radiological findings, the majority of tumors, 97.8%, involved the mandible with the posterior regions favored, and 56.5% of the lesions were multilocular type. Radiologic types had no correlation with the age of patients.
3. In the histologic findings, 23.9% of the lesions were follicular or mixed type. Histologic types had no correlation with the radiologic types, but a correlation between the unicystic type and first decade of age was found by  $X^2$  test ( $P < 0.05$ ).
4. Among the qualified 18 patients treated with radical or conservative surgery, the recurrence rate was 33.3%. The difference of recurrent rate between the multilocular type (36.4%) and the unilocular type (28.6%) was not much.

Conclusively, more detailed clinical and radiologic parameters should be added to clearly predict the biologic behavior of ameloblastoma.

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## 악골에 발생한 범랑모세포종의 임상 및 방사선학적 고찰

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하악에 발생한 45예, 상악에 발생한 1예로 총 46예의 범랑모세포종이 연구되었다. 환자의 평균연령은 34세였으며 성별에 따른 차이는 없었다. 가장 흔한 징후는 악골 또는 안면의 종창이었다. 소구치-대구치-하악지 부위를 침범한 예가 20예(43.5%)였다. 다방성 병소가 26예(56.5%)에서 발견되었다. 방사선학적 형태는 환자의 연령이나 조직학적 형태와는 상관관계가 없었다. 단방형 형태는 주로 젊은 환자에서 발견되었으며 10대에 호발하는 상관관계를 나타내었다( $P < 0.05$ ). 근치 또는 보존적 수술로 치료를 받은 분석가능한 18명의 환자에서 재발율은 33.3%를 나타내었다. 다방성 형태(36.4%)와 단방성 형태(28.6%) 간에 재발율의 차이는 크게 나타나지 않았다. 결론적으로 범랑모세포종의 생물학적 행태를 명확하게 예측하기 위해서는 좀 더 상세한 임상 및 방사선학적 변수가 첨가되어야 할 것으로 사료되었다.