

CLINICAL AND RADIOLOGIC STUDY OF ODONTOGENIC KERATOCYST

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The term odontogenic keratocyst for all odontogenic cysts, showing keratinization of epithelium, regardless of type, was first suggested by Philipsen in 1956. These odontogenic keratocyst can be distinguished from other odontogenic cysts in clinical behaviors and histologic features.

Pindborg and Hansen (1963) discovered 30 odontogenic keratocysts from 792 cysts and presented their clinical, radiographic and histologic characteristics¹⁸⁾

Payne (1972) clinically classified 103 odontogenic keratocysts which corresponded with the criteria of odontogenic keratocyst that had been suggested by Pindborg, and was especially interested in recurrence after surgical treatment. He did follow-up check (6 months to 18 years) on 20 patients and got 45 per cent of recurrence rate.¹⁷⁾

Donoff and his colleagues (1972) detected 16 odontogenic keratocysts among 326 cysts (48 per cent incidence) through microscopic examination.¹⁰⁾

There was no odontogenic keratocyst in 241 radicular cysts and all the 4 dentigerous cysts were odontogenic keratocyst, which conformed to the results in 1967 Sockolene and Shear had advocated that the origin of all odontogenic keratocyst was primordial. Referring to "daughter" cysts in their microscopic study, they observed the isolated cystic epithelial lining from the underlying mesenchyme, and suggested the role for its cause.²¹⁾ Donoff (1972) also reported the enzymatic mechanism of collagenase in the growth of odontogenic keratocyst.¹¹⁾

Brannon (1976) closely analyzed the clinical features and microscopic appearance of 321 odontogenic keratocyst.⁷⁾ According to this study, 5.1 per cent of all odontogenic keratocysts appeared in basal cell nevus syndrome and 5.8 per cent developed in the patient with multiple cysts, from which he suggested the correlation of odontogenic keratocyst with multiple cysts.

In summary, odontogenic keratocyst occurs most frequently in the second and third decades and at the mandibular third molar region, and shows slight predilection in male. It has high recurrence rate after surgical treatment, which is the important reason why we must differentiate it from other lesions before surgical procedures.

The radiographic appearance of odontogenic keratocyst cannot be distinguished from that of other intrabony cysts. Frequently its lumen, densely filled with keratin, will cause the usual

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radiolucent cyst-like image to have hazy appearance.

Sometimes, it may have scalloped borders and even occur with a multilocular appearance.²⁶⁾

The purpose of this article is to define further clinical behaviors and radiographic appearances of odontogenic keratocyst developed in jaws, with special interest in recurrence rate which is generally high.

I. Materials and Methods

Materials

32 patients whose microscopic examinations were verified as odontogenic keratocyst were examined in this study. Among them 22 patients were from Seoul National University Hospital, 5 from Kyung Hee University Hospital and 5 from Yon Sei University Hospital.

Method

Under the radiographs of 21 males and 11 females, the following details were examined, and in all possible cases, patient's records were also investigated. In order to confirm recurrence after surgical treatment, follow-up was done in all possible cases.

1. Distribution by sex and age
2. Location
3. Distribution of cysts according to type
4. Radiographic appearances
 - a. resorption of dental roots
 - b. types: unilocular and multilocular
 - c. border: smooth round and scalloped
 - d. haziness and expansion of cortical plates
5. Recurrence
6. Clinical findings

II. Results

1. Distribution by sex and age

Of the 32 patients, odontogenic keratocysts occurred in 21 males and 11 females. From the second to the fifth decades, most of the patients were fairly distributed and a gradual decline through the later years was shown. At the time of first diagnosis, almost all lesions were very large. Judging from this observation, cysts must have developed earlier in life than detected first. The ages and sex are shown in Table 1.

Table 1. Age and sex distribution of OKC

Age (Years)	Sex		Total
	M	F	
0-10	0	0	
11-20	6	3	9
21-30	7	2	9
31-40	1	1	2
41-50	5	4	9
51-60	1	1	2
61-70	1	0	1
Total	21	11	32

2. Location

10 cysts occurred in maxilla and 22 cysts in mandible.

Of 22 mandibular lesions, 10 cysts were located in molar-ramus area, 8 cysts crossed over the symphysis of mandible and 1 case showed multiple cysts.

However, in maxilla, even though all the area of the upper jaw was almost equally involved, 9 out of 10 cases were associated with antrum. 1 cyst only involved nasal cavity. The locations of odontogenic keratocysts are shown in Table 2.

Table 2. Location of 32 OKCs in present study

Location	No.	Total	Location	No.	Total
Maxilla			Mandible		
Anterior teeth	2	10	anterior teeth	0	22
premolar	3		premolar	2	
molar	2		molar	1	
* unstated	2		molar-ramus	10	
**antrum involved	9		cross symphysis	8	
			# unstated	1	

*: involved more than two above described area

** : Of 10 maxillary cysts, 1 cyst had no more information than the involved antrum. 8 cysts of remaining 9 associated with antrum at any manner.

#: recurred type multiple cyst.

3. Distribution of cysts according to type

The types of cysts that were preliminary diagnosed on the clinical and radiographic basis before microscopic examination are shown in Table 3.

Without microscopic confirmation, 12 cases were diagnosed as primordial cyst (37.5%), 6 cases as dentigerous cyst (18.8%), 2 cases as mural ameloblastoma and 2 cases as residual cyst.

Table 3. Distribution of cysts according to type

Initial diagnosis	No.	Per cent
primordial cyst	12	37.5
dentigerous cyst	6	18.8
residual cyst	3	9.4
recurred cyst	3	9.4
mural ameloblastoma	2	6.3
radicular cyst	2	6.3
globulomaxillary cyst	1	3.1
periodontal cyst	1	3.1
ameloblastoma	1	3.1
unknown	1	3.1
Total	32	

4. Radiographic appearances

(1) root resorption

The degree of the dental roots resorption are divided into the 4 groups;

—: present intact dental root (no evidence of root resorption)

—: can recognize the root resorption but slight

+: less than one third

++: more than one third

With the above classification, the following results can be obtained

—: 12 cases

—: 9 cases

+: 2 cases

++: 2 cases

(2) type – divided as unilocular and multilocular type in radiographic appearances.

In mandibular lesions, unilocular and multilocular types were equally distributed but,

in maxillary lesions, almost all cysts appeared as unilocular type (7 cases of 9) (Fig. 1).

(3) border--divided as scalloped and smooth rounded.

In mandibular cysts, 16 cysts had the scalloped border while the remaining 6 cysts had smooth rounded border.

In maxillary cysts on the contrary, 7 out of the 9 cysts had the smooth rounded border (Fig. 1, Table 4).

Table 4. Type and border of OKC

	Type		Border	
	unilocular	multilocular	scalloped	smooth-round
Mand.	11	11	16	6
Max.	7	2	2	7

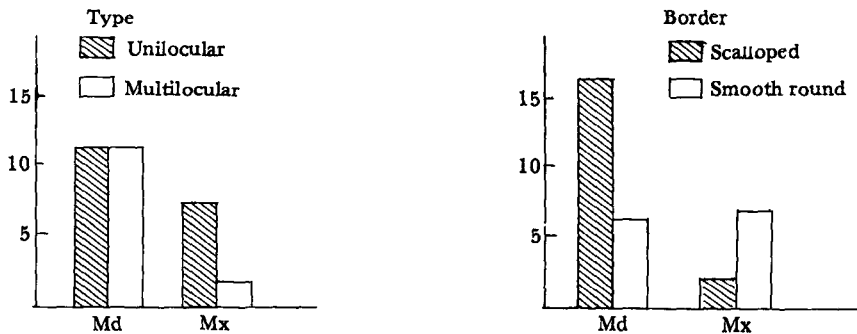


Fig-1. Type and Border of OKC in present study.

(4) Haziness and expansion of cortical plates

4 cysts of the mandible and 1 cyst of the maxilla showed increased radiopacity, but these observation can not be accepted objectively due to lack of standardization.

9 cases arising in mandible showed cortical plate expansion in which 1 case had buccal and lingual expansion. In maxillary lesions, because there were no available dental radiographs showing cortical expansion, we could not examine it.

5. Recurrence

15 patients who were able to be recalled, were examined radiographically from 2 months to 4 years after operation (Table 5).

Table 5. Comparison of recurrence rate of OKC in this study with other reports.

	Present study	Toller	Rud	Pind borg
recurrence	4 of 15	9 of 33	7 of 21	10 of 16
per cent	26.7	57	33	62

4 patients showed recurrence after surgery (26.7%). Of these one had two times recurred history. The first recurrence occurred 3 years and 3 months after surgery, and the second recurrence occurred 1 year and 6 months later after the second operation.

6. Clinical findings

11 cysts of 21, of which dental records were available, combined with infection, 8 cysts showed only bony swelling and 2 cysts were found in the routine radiographic examination.

III. Discussion

The range of patients in this study was 12 to 65 years, with the mean age being 31 years 7 months. A peak incidence was in the second and third decades with gradual decline thereafter.^{5,6,7,9,10,17,19} Brannon found the peak incidence to be in the second and third decades of life with the mean age being somewhat higher at 37 years 9 months.⁷⁾

There was an apparent sex predilection,^{5,6,7)} with twenty-one males and eleven females (male-to-female rate of 2:1). A comparison with Brannon's findings shows the sex ratio to be similar (1.35:1).⁷⁾

Of the 32 cases, 22 (68.8%) occurred in the mandible and 10 (31.3%) in maxilla.^{5,6,7,10,15,17,19)} The most common site in this study was the mandibular molar-ramus regions (31.3%, 10 out of 32 cases). The second most common site was the mandibular anterior region in which all cysts crossed the symphysis (25%, 8 out of 32 cases).⁷⁾ Of these, one involved the mandible from the first molar to the opposing coronoid process, (Fig. 3), another one from the second molar to the opposing second molar and the others at least from the first premolar to the opposing first molar (Fig. 8). This surprisingly large destruction of mandible suggests that the OKC's have more powerful destructive potential than other cysts. Similar findings were shown on maxillary lesions. In 2 cases, cysts tended to erode the ANS that is known too hard to be perforated by a common cysts may be due to the compact intermaxillary suture (Fig-9). The rate of growth of the OKC is also greater and the rate of expansion of the OKC is the most rapid, because keratinized epithelial lining would be expected to act as a more efficient semi-permeable membranes than the epithelium in other odontogenic cysts (Brady), and the osmolarity of the cystic fluid in

OKC's is greater than that in the other odontogenic cysts (Toller).^{6,24)} In maxilla, though the whole area of the upper jaw was almost equally involved, interestingly 9 out of 10 cases associated with antrum, in which two cases combined with maxillary sinusitis, one case involved a nasal cavity and others encroached the antrum inferiorly or laterally¹⁰⁾ (Fig-7). Thus, almost maxillary OKC's more or less contributed to the abnormal feature or function of maxillary sinus.

Only one multiple cyst occurred in mandible as recurrence type.

The incidence by location in this study is in close agreement with the findings of Branon (65.4% in mandible, 34.6% in maxilla). The most common site was the mandibular third molar and/or ramus region (28.8%).⁷⁾ Payne also showed similar results with this study. In his large series, 65.5% occurred in the mandible, with 31% occurring in the area of the mandibular third molar, angle and ramus and 28.4% occurred in maxilla.¹⁷⁾

Of 25 cases which could be observed the degree of the dental roots resorption, 12 cases showed no evidence of the root resorption.

In 9 cases, only slight resorbing evidence was recognized (Fig-4). An apparent root resorption could be found in 4 cases of which 2 cases showed two plus resorption.

Unilocular and multilocular types were equally distributed in mandible but, most cysts appeared as unilocular type (7 out of 9 cases) in maxilla^{1,5,7,10,18,26)} (Fig. 3, 4, 5, 6, 8). In this study, some of cysts categorized as multilocular type probably were unilocular type in nature, but this is impossible to determine it due to character of plane radiographs. This is similar to ameloblastoma in which almost of maxillary lesions appear as unilocular type.^{9,18,19)}

In mandible, 16 out of 22 cysts showed a scalloped border while in maxilla 7 out of 9 cysts a smooth rounded border (Fig. 2, 4). This finding does not appear in general odontogenic cyst, but in ameloblastomas.^{5,13,26)}

Pindborg and Hansen (1963) summarized the radiographic appearance of OKC as festooning margins and a multilocular structure.¹⁸⁾ Keith (1973) reported that radiographs showed loculation in adjacent bone and at operation, macroscopic satellite cysts were found at these sites.¹⁵⁾ These satellite cysts at first called daughter cyst or microcyst but, the former term is preferred to the latter term because there is no evidence that satellite cysts arise from the proliferation of the lining of the main cyst.

However, Thoma²³⁾ advocated that multilocular cyst develops from an epithelial sprout that branches and forms at each end of the branch a small cyst. These become larger and as they crowd together and fused the intercystic tissue is resorbed so that a single space results with a lobular outline and partial septa, which appear to subdivide the cavity.

Putting these various reports together, the scalloped margin of OKC apparently associates with satellite cysts.^{6,9,10,14,15,16,17,19)}

Of the 21 cases in which patient's chart could be available, 19 were clinically symptomatic before the patient sought treatment. The most common findings at the initial examination were drainage and/or pain (11 cases).^{5,7,9,15,19)} Painless swelling was the next common complaint (8 cases) of which two patients showed crepitus. In one patient neurologic involvement, was clinically evident.¹⁶⁾ This symptom was paralysis of chin area. 2 cases were found in the routine

examination.⁷⁾

The recurrence of odontogenic keratocysts has been emphasized by several investigators (Pindborg and Hansen (62%) 1963,¹⁸⁾ Payne (45%) 1972,¹⁷⁾ Donoff (15.4%) 1972)¹⁰⁾, and it's striking when compared with the relative rarity of recurring non-keratinized cysts. The recurrence rate in this series is not as high as that of approximately 60 per cent recorded by Pindborg and Hansen (1963) and Toller (1967). The information available in this article includes a number of patients with only short postoperative follow-up of two months to one year, and of the recurrence rate may yet turn out to be greater.

Browne said the longest time interval, between the removal of an odontogenic keratocyst and its subsequent recurrence is 30 years. However it seems likely that, if routine radiological follow-up had been employed in a number of those cysts in which the recurrence was not found until after 6 years or more, it would have been discovered much earlier. In view of these findings, it would seem to be advisable to carry out annual radiological checks for five years post-operatively.⁵⁾

The reason for the recurrence of these lesions remains speculative.⁴⁾ Fickling (1965)¹²⁾ enumerated the possible causes of recurrence; (1) ineffective original enucleation; (2) failure to ensure healing from the depth of the wound; (3) true budding or daughter cyst formation,^{6),8),17),25)} and (4) multiple center potential.^{14,15)} Browne postulated that the recurrence is because of the nature of the lesion itself and not related to its method of treatment.⁵⁾ Browne reviewed 65 patients and found no significant correlation between type of operation and recurrence, nor did the site or size of the lesion seem to matter.

Stoeltinga has demonstrated the presence of many foci of dental lamina cells between keratocyst membrane and the overlying oral mucosa. He thus recommends excision of the overlying mucosa to prevent recurrence.^{9,22)}

Satellite cysts and epithelial islands are often seen in association with keratinizing cyst. Satellite cysts may arise from proliferation of the epithelium of the main cyst, but there is evidence suggesting origin from the adjacent rests of epithelium, the remnants of the dental lamina.^{6,8)} Several authors showed that there was no significant correlation between the presence of satellite cysts or epithelial rests and subsequent recurrence.

Other factors contributing to recurrence of the keratocyst suggested by Chuong et al.⁹⁾ include the following; (1) Keratocysts have a relatively thin wall, which is easily ruptured, thereby presumably enhancing in complete removal.⁴⁾ (2) Keratocyst epithelium has a tendency to separate from the underlying mesenchyme, which may contribute to scattering of epithelial fragments into the surrounding tissues.^{5,10)} (3) Keratocysts are frequently found at operation to have perforated buccal or lingual cortical plates, thereby involving the surrounding soft tissue and making total removal less than certain.²⁰⁾

However, there is no clear-cut cause of recurrence, so further study was recommended.

Summary

1. There was an apparent sex predilection for male (male to female ratio is 2:1).
2. The peak incidence was the second and third decades with gradual decline thereafter with a mean age of 31 years 7 months.
3. The most common site was a mandibular molar-ramus region, 22 cases (68.8%) occurred in the mandible and 10 cases (31.3%) in the maxilla.
4. The largest number of keratocysts was initially diagnosed as primordial cyst (37.5%), followed by dentigerous cyst (38.8%) and residual cyst (9.4%).
5. Radiograph showed various degree of dental root resorption in 13 out of 25 cases (52%).
6. Maxillary keratocysts tended to exhibit a unilocular type and smooth round border, while mandibular keratocyst have scalloped border.
7. 11 out of 21 cysts clinically combined with infection, and 8 cysts showed only bony swelling and 2 cysts were found in the routine radiographic examination.

REFERENCES

1. Tae Won Park: Clinico-radiological study of cyst of the jaw, J. Kor. Acad. Maxillo. Rad., 13:161-166, 1983.
2. Rhee, C.G., Kim, K.S., Nam, I.W., and Shin, S.C.: The clinical and statistical studies on cysts in oral cavity, Kor. Med. J., 11, 1966.
3. Anneroth, G. and Hansen, L.S.: Variations in keratinizing odontogenic cysts and tumors, Oral Surg., 54:530-546, 1982.
4. Browne, R.M., and Miller, W.A.: Rupture strength of capsules of odontogenic cysts in man, Arch. Oral Biol., 14:1351, 1969.
5. Browne, R.M.: The odontogenic keratocyst, clinical aspect, Brit. Dent. J., 128:225, 1970.
6. Browne, R.M.: The odontogenic keratocyst, Brit. Dent. J., 131:249-1971.
7. Brannon, R.B.: The odontogenic keratocyst, A clinicopathologic study of 312 cases. Part I. Clinical features, Oral Surg., 42: 54-72, 1976.
8. Brannon, R.B.: The odontogenic keratocyst, A clinicopathologic study of 312 cases. Part II. Histologic features, Oral Surg., 43: 233-255, 1977.
9. Chuong, R., Donoff R.B., and Guralnick, W.C.: The odontogenic keratocyst, J. Oral Maxillofac. Surg., 40:797-802, 1982.
10. Donoff, R.B., Guralnick, W.C., and Claymann, L.: Keratocyst of the jaws, J. Oral Surg., 30:800-804, 1972.
11. Donoff, R.B., Harper, E. and Guralnick, W.C.: Collagenolytic activity in keratocysts, J. Oral Surg., 30:879-884, 1972.
12. Fickling, B.W.: Cysts of the jaw: A long-term survey of types and treatment, Proc. R. Soc. Med., 58:847-854, 1965.
13. Goaz, P.W., and White, S.C.: Oral Radiology, The C.V. Mosby Co., 1982.
14. Harris, M.: A review of recent experimental work on the dental cyst, Proc. R. Soc. Med., 67:1259-1263, 1973.
15. Keith, D.A.: Macroscopic satellite cyst formation in the odontogenic keratocyst,

- Oral Surg., 35:21-27, 1973.
16. Panders, A.K., and Hadders, H.N.: Solitary keratocysts of the jaws, *J. Oral Surg.*, 27: 931-938, 1969.
 17. Payne, T.F.: An analysis of the clinical and histopathologic parameters of the odontogenic keratocyst, *Oral Surg.*, 33:538-546, 1972.
 18. Pindborg J.J., and Hansen, J.: Studies on odontogenic cyst epithelium, *Acta Path. Microbiol. Scand.*, 58:283-294, 1963.
 19. Rud, J., and Pindborg, J.J.: Odontogenic keratocysts: A follow-up study of 21 cases, *J. Oral Surg.*, 27:323-330, 1969.
 20. Schofield, J.J.: Unusual recurrence of an Odontogenic keratocyst, *Brit. Dent. J.*, 130:487, 1971.
 21. Soskolne, W.A., and Shear, M.: Observation on the pathogenesis of primordial cysts, *Brit. Dent. J.*, 123:321, 1967.
 22. Stoeltinga, P.T.W.: Studies on the dental lamina as related to its role in the etiology of cysts and tumors, *J. Oral Pathol.*, 5:65, 1976.
 23. Thoma, K.H.: *Oral Surgery*, 5th Ed., The C.V. Mosby Co., 1969.
 24. Toller, P.A.: The osmolality of fluids from cysts of the jaws, *Brit. Dent. J.*, 129:275-278, 1970.
 25. Wilson D.F., and Ross, A.S.: Ultrastructure of odontogenic keratocyst, *Oral Surg.*, 45:887-893, 1978.
 26. Wood, N.K., and Goaz, P.W.: *Differential Diagnosis of Oral Lesions*, Second Ed., The C.V. Mosby Co., 1980.
 27. Wright, J.M.: The odontogenic keratocyst: orthokeratinized variant, *Oral Surg.*, 45:887-893, 1978.

(국 문 초 록)

— 치원성 각화 낭종의 임상 방사선학적 연구 —

서울대학교 치과대학 치과방사선학교실

김 성 래 · 박 태 원

치원성 각화 낭종의 임상적 방사선학적 양상을 연구하였다. 32명의 환자(21명의 남자, 11명의 여자)를 이 연구를 위하여 사용하였다. 환자들의 기록과 방사선사진을 통하여 성별 및 연령의 분포, 호발부위, 방사선사진의 양상, 재발율 임상적 소견들을 조사하였던바 아래와 같은 결과를 얻었다.

1. 남성이 여성에 비해 약 2 배 호발하였고, 10대와 20대에서 가장 많이 발생하였다.
2. 호발부위는 하악의 구치에서 하악지에 이르는 부위였고 하악골이 상악골에 비해 호발하였다.
3. 원기성 낭종에서 각화성 변화를 일으킨 것이 가장 많았다.
4. 치근의 흡수를 많이 보였으며, 하악골에서 경계는 원형과 부채형 (scalloped) 이 거의 등등히 나타났고 상악골에서는 대부분 원형의 단방형 낭종이었다.
5. 높은 재발율을 보였다.
6. 감염이 되어 있는 경우가 많았다.

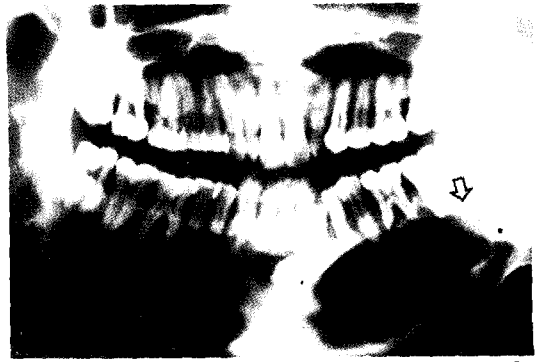
Explanation of Figures

- Fig-2.** This radiograph demonstrates an example of the dentigerous cyst occurring at the mandibular pre-molar region, histologically proved to be odontogenic keratocyst. It is multilocular type with scalloped border. Lower right canine and first premolar showed slight root resorption.
- Fig-3.** This case shows an example of a huge cyst involving whole mandible except the right mandibular ramus area. Impacted lower left canine is seen within the cystic cavity, but this cyst isn't likely to be a dentigerous cyst, considering the direction of impacted tooth crown. It also a multilocular appearance with scalloped border.
- Fig-4.** This case is a residual cyst occurring at the upper anterior region. It showed unilocular appearance with smooth round border.
- Fig-5.** This case shows an unilocular radiolucent cystic lesion with smooth round border at the lower pre-molar-molar area. Slight root resorption is seen on the lateral aspect of mesial root of lower left first molar.
- Fig-6.** Radiograph shows a relatively small cystic cavity of unilocular appearance with smooth round border, posterior to the lower left second molar.
- Fig-7.** This case shows a relatively large radiolucent cystic cavity of unilocular appearance with scalloped border, extending from the upper canine to the upper left maxillary tuberosity area. Also we could find the destruction of the lateral wall of the left maxillary sinus.
- Fig-8.** This is an example of a very large cyst, extending from the lower left second molar to the right second molar. It shows an unilocular appearance with scalloped border and lateral root resorption on both lower first molar.
- Fig-9.** This radiogram shows a relatively large cyst in the maxilla, crossing the midline. It is multilocular type with scalloped border and shows tooth displacement of upper right lateral incisor, canine, first pre-molar and second premolar.

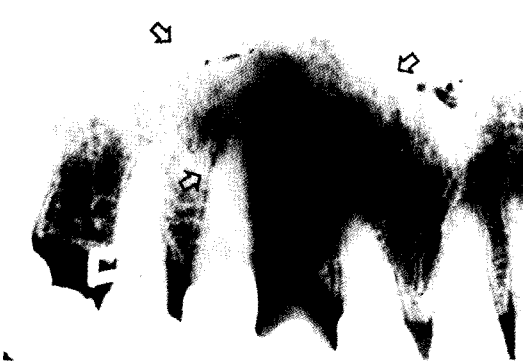
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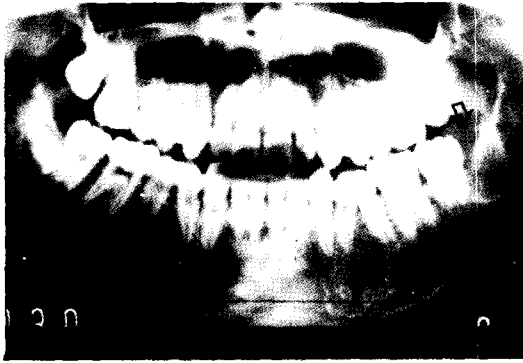
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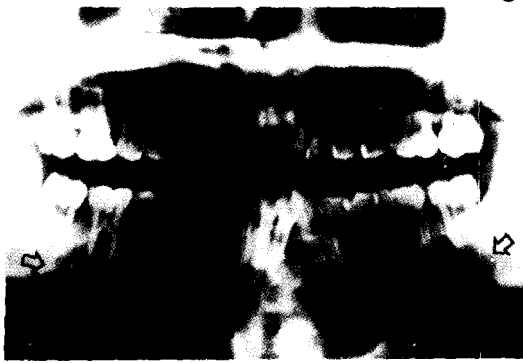
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