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A Study of Ameloblastoma in Children and Adolescents

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Ⅰ. Introduction

Ameloblastoma is a benign neoplasm of the jaws with locally aggressive capacity. The lesion is relatively rare, accounting for about 1 % of all tumors[1,2]. Ameloblastoma occurs at all ages with a peak incidence in the third and fourth decades of life. Small and Waldron[2] reported that the average age at the time of treatment was 38.9 years. In Korean studies by Choi et al.[3] and Park et al.[4], the average age was 30.7 years and 34 years, respectively. It is known that ameloblastoma is rare in childhood. Bhaskar[5] and Blackwood[6] reviewed odontogenic tumors in children and failed to find a single case. However, Daramola[7] et al. found that 16(22.9%) of 70 diagnosed cases were in patients under 18 years of age and Chidzonga[8] noted that 20(17.1%) out of 117 patients were 18 years of age or younger. Also Choi et al.[3] and Park et al.[4] reported that second decade revealed the highest rate. Therefore, there is need to study the appearance and frequency of ameloblastoma in children and adolescence.

The purpose of this study was to report cases of ameloblastoma arising in children and adolescents, and to detail the clinical, radiographic and histologic features.

Ⅱ. Materials and Methods

The records of 46 patients treated for ameloblastoma at Pusan Natioanl University Hospital, during the period from August 1984 to May 1998 were retrieved and the cases of those patients, who were 18 years of age or younger were selected. Sex, location, clinical and radiographic findings were reviewed and histologic type was recorded.

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Ⅲ. Results

The clinical, radiographic findings and histologic types are summerized in Table 1.

Table 1. Clinical, Radiographic findings and Histologic types in 15 cases of ameloblastoma in children and adolescents

Case No.	Sex	Age (Years)	Location	Clinical Findings	Radiographic Findings	Histologic Types
1	F	5	Symphysis	Swelling, Tenderness	Moderated-defined margin Unilocular (scalloped) Bucco-lingual cortex expansion No root resorption	Plexiform unicystic type
2	M	9	#75 to #37	Swelling	Well-defined margin Unilocular (scalloped) Bocco-lingual cortex expansion #75, 36 : Root resorption	Plexiform unicystic type
3	F	11	#36 to Lt. horizontal ramus	Swelling Tenderness	Moderate-defined margin Unilocular (scalloped) Bucco-lingual cortex expansion No root resorption	Plexiform type
4	M	11	#35 to Lt. ascending ramus	Swelling	Moderate-defined margin Unilocular (scalloped) Bucco-lingual cortex expansion No root resorption	Plexiform unicystic type
5	M	15	#46, 47	Swelling Tenderness	Well-defined margin Unilocular (scalloped) Bucco-lingual cortex expansion #36, 37 : Root resorption	Follicular type
6	M	15	Symphysis	Swelling Gingival ulceration	Moderate-defined margin Unilocular (smooth) No cortical expansion No root resorption	Follicular type
7	F	16	#43 to Rt. ascending ramus	Swelling Pain	Moderate-defined margin Multilocular (soap-bubble) Bucco-linguo-inferior cortex expansion #45, 46 : Root resorption	Plexiform type

(continued)

Table 1(cont.)

8	M	16	#35 to Lt. ascending ramus	Swelling Gingical ulceration	Moderate-defined margin Unilocular (scalloped) Bucco-linguo-inferior cortex expansion and partial perforation #35-37 : Root resorption	Plexiform unicystic type
9	M	16	#36 to Lt. horizontal ramus	Swelling	Well-defined margin Unilocular (scalloped) Bucco-lingual cortex expansion #36 ; Root resorption	Plexiform unicystic type
10	F	16	#45 to Rt. ascending ramus	Swelling Pain Pus discharge	Well-defined margin Multilocular (soap-bubble) Bucco-linguo-inferior cortex expansion # 45, 46 : Root resorption	Plexiform unicystic type
11	M	17	#32 to Lt. coronoid process	Swelling	Well-defined margin Unilocular (scalloped) Bucco-linguo-inferior cortex expansion #32-35 : Root resorption	Plexiform type Lymphnode metastasis
12	F	17	#36 to Lt. ascending ramus	Swelling	Well-defined margin Unilocular (scalloped) Bucco-linguo-inferior cortex expansion #36 : Root resorption	Plexiform type
13	M	18	#45 to Rt. ascending ramus	Swelling	Well-defined margin Unilocular (scalloped) Bucco-linguo-inferior cortex expansion #46,47 : Root resorption	Plexiform type
14	M	18	#45 to Rt. horizontal ramus	Swelling Tenderness	Well-defined margin Multilocular (soap-bubble) Bucco-lingual cortex expansion #46, 47 : Root resorption	Plexiform type
15	F	18	#35 to Lt. ascending ramus	Swelling Paralysis	Moderate-defined margin Multilocular (soap-bubble) Bucco-lingual cortex expansion #35 : Root resorption	Plexiform type

Sex and Age

15(32.6%) of 46 patients were 18 years of age or younger and the average age at the time of presentation was 14.5 years with a range between 5 and 18 years. There was male preponderance: nine(60%) patients were males and six(40%) were females, the male/female ratio being 1.5:1.

Site Distribution

All the lesions located in the mandible. Of the 15 cases, 11(73.3%) cases including one, extended to coronoid process occured in molar-ramus region; two(13.3%) cases were confined to the symphysis; two(13.3%) were found in the premolar-molar region.

Clinical Signs and Symptoms

Swelling was the main complaint in all patients. Tenderness was presented in four patients. A few patients showed other findings; pain, gingival ulceration, pus discharge and paralysis.

Radiographic Findings

Radiographically, 11(73.3%) cases manifested unilocular lesions with smooth or scalloped border. 4(27.7%) cases showed multilocular radiolucency with soap bubble appearance. The margin of the lesions were well-defined in 8(53.3%) and moderate-defined in 7(46.7%) cases. All the cases except one(93.3%) showed bucco-lingual or bucco-linguo-inferior cortical expansion, and cortical perforation was shown in only one(6.7%) case Root resorption was presented in 11(73.3%) cases.

Histologic Findings

Histologically, 15 cases were composed of 7(46.7%) cases of plexiform type, 6(40%) cases of pexiform unicystic type and 2(13.3%) cases of follicular variant.

Ⅳ. Discussion

Ameloblastoma is an aggressive but benign epithelial neoplasm that presenting 11% of odontogenic tumors.[1,9]

They may occur over a wide age range, with a mean age in the mid 30s. The appearance of ameloblastoma in childhood is rare. Small and Waldron[2] estimated the frequency of ameloblastoma in children younger than 9 years of age to be 2%. Ramanathan and Lee Sung Guan[10] also demonstrated the paucity of childhood ameloblastoma. Recent studies.[7,8,11-13] reported much higher incidence of ameloblastmas developed in children and adolescents.

Keszler et al.[8] noted that eight(8.7%) of 92 cases were in patients younger than 16 years of age. Daramola[7] at al., Chidzonga[8] and Olaitan et al. reported the frequency of ameloblastoma under 18 years of age to 22.9%, 17.1% and 14.6%, respectively. Kahn[13] reviewed 132,884 ameloblastomas and found 38(12.2%) cases in persons less than 20-years-old.

In this study the incidence was rather higher than other stydies[7-13], 32.6% of the histologically confirmed ameloblastoma were in patients of 18 years of age or younger. 4.3% incidence in the first decade is also higher than the study of Small and Waldron[2].

The site and clinical features were very similar to previous reports of ameloblastomas[3,4,13,14]

In the present study, unilocular radiolucency was more frequent than multilocular lesion, in accordance with previous studies[11,15] for childhood, but it is different from the results of other studies[3,4,16-18] for all age group. The fact that young patient show umilocular preponderance is also supported by the studies[14,19] of cytogenic ameloblastoma, relatively common in early age group. Although cortical bone expansion and root resorption were also the radiographic characteristics of ameloblastoma in young persons like other studies for all age group[16,20], cortical perforation was rarely present.

The two most common histologic types of ameloblastoma are known as plexiform and follicular patterns[4,15,17,21,22]. However pexiform and plexiform unicystic types were two predominant forms in this study like the report of Choi et al[3]. The result that plexiform and plexiform unicystic types were found with almost equal frequencies was different from the anticipation of much more unicystic type, on the basis of many previous reports[13,14,19,23,24-28] that unicystic type was common in young persons. Actually plexifom type was the most common in this study and almost all the plexiform types were manifested in late teens. On the contrary to this, Kahn[15] reported only three plexiform unicystic types among 38 cases in young persons.

Histologic types were not related with root resorption in the current study.

In this study, there were one metastatic case and one recurred case.

Metastasis to cervical lymph nodes was shown in the largest one of all the 15 cases.

The recurred case presented the past history of having been enucleated of the lesion under the impression of odontogenic keratocyst two years ago.

It was impossible to confirm whether the previous lesion was real odontogenic keratocyst or misdiagnosis of unicystic ameloblastoma, but epithelial remants with proliferative potential might have been left after cyst enucleation and might caused recurrence.

V. Summary

A retrospective study of 15 cases of ameloblastoma in children and adolescents was performed. During the period of evaluation, 46 patients with ameloblastoma were seen, of which 15(32.6%) cases were in the patients aged 18 years or younger with the mean age of 14.5 years. There was more prevalence in male (1.5:1). All the cases occured in the mandible, the molar and ramus area was the most frequent location(66.7%), and the most frequent sign was swelling of the face or jaw. Radiographically, 11(73.3%) cases manifested unilocular leion. Cortical expansion and root resorption were presented in 14(93.3%) and 11(73.3%) cases respectively. Pexiform and plexiform unicystic types were common in the ameloblastoma occured in young patients.

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-국문초록-

아동 및 청소년에 발생한 법랑모세포종

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법랑모세포종은 흔히 중년에 호발하는 질환으로 알려져 있지만 아동 및 청소년에서도 그 발생이 보고되고 있다. 그러나 어린 환자들에 발생한 법랑모세포종의 임상적, 방사선학적 및 조직학적 분석은 드물다. 본 연구는 1984년 8월에서 1998년 5월사이에 부산대학교병원에서 법랑모세포종으로 진단받은 46명의 환자들중에서 18세 이하 환자 15명에 대한 후향적연구로서 아동 및 청소년에서의 법랑모세포종의 특징을 기술하고자하였다.

그 결과는 다음과 같다.

1. 46례의 법랑모세포종중에서 15(32.6%)례가 18세 이하의 아동에서 발생하였으며, 남자 9(60%)명, 여자 6(40%)명으로 1.5:1의 남녀비를 나타내었다.
2. 15례 모두 하악에 발생하였으며, 이 중 11(66.7%)례가 대구치부와 하악지에 나타났다.
3. 15례 모두 하악 종창의 주소를 보였다.
4. 방사선학적으로, 11(73.3%)례는 단방성 병소를, 4(27.7%)례는 다방성 병소를 나타내었다. 피질골 종창은 14(93.3%)례에서, 치근흡수는 11(73.3%)례에서 나타났다.
5. 조직학적으로 총상형이 7(46.7%)례, 단낭성형이 6(40%)례, 그리고 여포형이 2(13.3%)례 였다.

Key words : ameloblastoma, children