The Journal of Korean Academy of Maxillofacial Plastic and Reconstructive Surgery
Vol. 13, No2 : 160 - 166

UNICYSTIC AMELOBLASTOMA
— Case Report —

Eui - Wung Lee, Hyung - Sik Park, In - Ho Cha, Jin Kim*

*Dept. of Oral & Maxillofacial Surgery, Dept. of Oral Pathology**

College of Dentistry, Yonsei University

In 1977, Robinson & Martinez described a distinct varient of ameloblastomas in which the response to curettage was found to be favorable, with a recurrence rate of 25%. They referred to this varient as unicystic ameloblastoma.

Unicystic ameloblastoma occur most commonly in the second and third decades of life, which is considerablly younger than the average age of discovery for the classical ameloblastoma. For the accurate histopathological diagnosis of the unicystic ameloblastoma, the specimen obtained the excisional biopsy, complete enucleation or incisional biopsy from the multiple site of the lesion.

The purpose of this report is to review of the literature and to present three cases in which an unicystic ameloblastoma appear to be arising in the wall of a dentigerous cyst.

I. INTRODUCTION

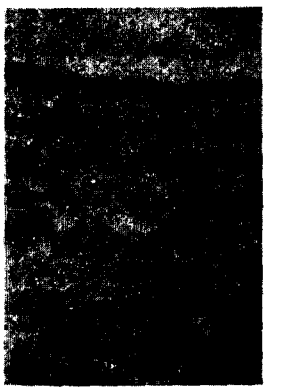
Ameloblastoma is the most aggresive of the odotogenic tumors and it arises from the dental lamina or a derivatives of the lamina[1](enamel organ, epithelial rests, follicular cysts). The apparent development of ameloblastoma in the wall of a dentigerous cyst was first described by Cahn in 1933. In 1965, Stanley & Diehl reviewed the English-language publication and pointed out that the evidence for 641 cases of ameloblastoma was somewhat equivocal, as in many of the reports no definite evidence is offered to show that a neoplasm has developed from that was previously a simple cyst. In 5% to 6% of cysts, however, the lining of the connective tissue wall shows ameloblastic proliferation.[1,4,5] These lesions are the precursors of ameloblastomas and require careful removal & follow-up. Generally, ameloblastoma arising from the dentigerous cysts, was called mural ameloblatoma. In 1977, Robinson & Martinez described a distinct varient of ameloblastoma in which the response to curettage was found to be favorable, with a recurrence rate of 25%, They referred to this varient as unicystic ameloblastoma. In 1984, Gardner reported a series of 35 instances of unicystic ameloblastoma, which was termed as plexiform unicystic ameloblastoma, refering to a pattern of epithelial proliferation that has been described in dentigerous cysts. The term unicystic implies that ameloblastomas associated with odontogenic cysts are either unilocular radiographically or arise from a grossly unilocular cystic cavity. Leider indicated that some of these neoplasms are multilocular radiographically and may exhibit pseudopod-like cystic extensions grossly and microscopically. In addition, since an ordinary cyst lining can be identified in most of these lesions, They prefer to simplify the terminology and refer to this variant as cystic ameloblastoma.[3,6,7]

A number of important factors must be considered in planning the treatment of ameloblastoma. It is essential to distinguish among the three clinical types of ameloblastoma –the intraosseous solid or multicystic lesion, the well-circumscribed unicystic type, and the rare peripheral (extraossous) ameloblastoma– because they require different forms of treatment. Unicystic ameloblastomas in which the tumor extends into the lumen of the cyst

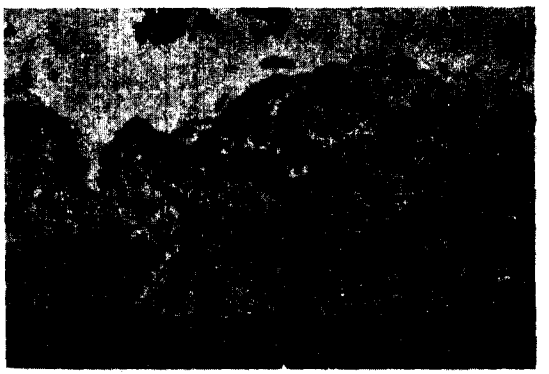
〈CASE 1〉

pre-op photo

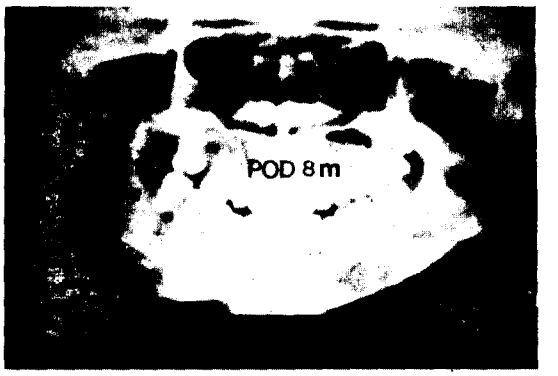
pre-op PANEX



incisional biopsy result.



total enucleation biopsy result.



post-op PANEX

or involves only the cystic lining can be expected to be removed completely by enucleation. Alought endorsing the principle of total excision as the ideal treatment of ameloblastoma, some members of this group make an allowance for age, treating the young in a more conservative manner.[9] Tiecke and associates consider enucleation to be a more realistic attitude toward the problem of large cystic lesion in young patients.[10]

The purpose of this report is to review the literature and to present three cases in which an unicystic ameloblastoma appears to be arising in the wall of a dentigerous cyst.

II. CASE REPORT

〈CASE 1〉

A 19-year-old girl was referred to our dental hospital for the treatment of asymptomatic swelling on Rt. buccal cheek. The patient recognized the swelling on Rt. madibular angle area about 1.5 years ago. The swelling was rapidly enlarged after a traumatic injury by first-blow on the swelling site about 6 months ago. Recently, pus discharged from Rt. lower 2nd molar crevice. So, and she visited a local dental clinic.

Oral examination revealed a large amount of bony expansion around the Rt. mandibular molar area, ascending ramus and angle area, resulting in a facial asymmetry(photo). And there were also submandibular lymphadenopathy.

Radiologic examination revealed a multilocular radiolucency extending from the Rt. lower 1st molar to the coronoid process, condylar neck and angle area. The Rt. 3rd molar was displaced toward cornoid process and mandibular canal was also displaced inferiorly. The root of the Rt. 2nd molar showed external root resorption(X-ray).

Under the local anesthesia, incisional biopsy was perfo-rmed. The result of the biopsy was dentigerous cyst

(slide).

The patient was admitted to our hospital. Routine laboratory results were within normal limits. Under the general anesthesia, the lesion was complete enucleation using intraoral approach. The Rt. 3rd molar was removed and mandibular canal was preserved.

The result of the excisional biopsy was unicystic ameloblastoma(Ackermann Group 1).

Postoperative progress was good. Follow-up radiographs showed a rapid filling of bone up to a year postoperatively. Today, facial asymmetry was improved and the lesion has no recurrence evidence(X-ray).

〈CASE 2〉

A 10-year-old girl was referred to our dental hopital for the treatment of painless swelling on Rt. buccal cheek.

Oral examination revealed a unilocular radiolucency including the Rt. lower 2nd molar, The Rt. lower 3rd molar tooth bud was modrate displaced and the mandibular canal was intact(X-ray).

Under the local anesthesia, incisional biopsy was performed. The result of the biopsy was unicystic ameloblastoma(Ackermann Group 2) (Slide).

The patient was admitted to our hospital. Routine laboratory results were within normal limits. Under the general anesthesia, the lesion was complete enucleation and the Rt. lower 2nd & 3rd molar were extracted. The tooth bud of the Rt. lower 3rd molar was not involved in the lesion and the mandibular canal could not found.

Postoperative progress was good. Today, there is no sign of recurrence (photo, X-ray).

〈CASE 3〉

A 28-year-old male, with discomfort during mastication in the Lt. posterior teeth area, was referred to our dental hospital.(duration : 1 month)

Oral examination revealed a slight expansion in the

〈CASE 2〉

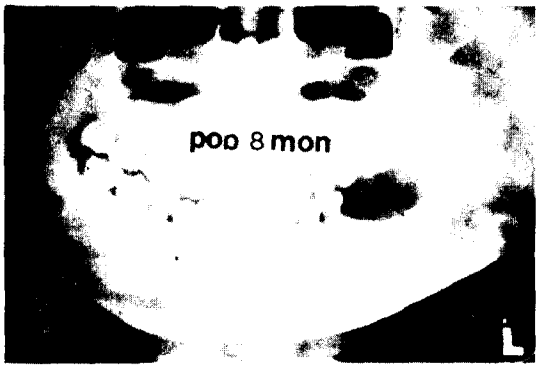
pre-op photo

pre-op intraoral photo

post-op histopathologic features

〈CASE 3〉

pre-op PANEX



post-op PANEX

post-op histopathologic features

Lt. mandibular angle area. No limitation of mouth opening and no submandibular lymphadenopathy were presented. Radiologic examination revealed a large unilocular radiolucency of the mandibular body, angle and coronoid process. Root resorption, tooth displacement and canal displacement was not present(X-ray).

The patient was admitted to our hospital. Routine laboratory results were within normal limits. Under the general anesthesia, the lesion was complete enucleation. The result of the biopsy was unicystic ameloblastoma (Ackermann Group 3b) (slide).

Postoperative progress was good. The enucleated cavity was rapidly filled with bone. Today, there is no sign of recurrence(X-ray).

III. DISCUSSION

The origin of the unicystic ameloblastoma remains controversial with some authors favouring origin from pre-existing odontogenic cysts, usually dentigerous cyst, but including odontogenic keratocysts and residual cysts[7] while others regard to this lesion as a cystic neoplasm arising de novo.[6]

The question of whether ameloblastoma originates in dental cysts or whether ameloblastoma becomes cystic would appear unresolvable without delineation of precise diagnostic criteria concerning ameloblastoma and dental cysts. In 1970, Vickers and Gorlin have published specific histologic criteria that are helpful in the diagnosis of early lesions. 1, hyperchromatism of basal cell nuclei of the epithelium lining the cystic cavities 2, palisading with polarization of basal cell nuclei of the epithelial lining the cystic cavities 3, cytoplasmic vacuolization of basal & basilar cell of the epithelial lining the cystic cavities.[11,12]

The lesion was divided into 3 groups according to the following criteria which have been slightly modified from those first described by Robinson & Martinez and

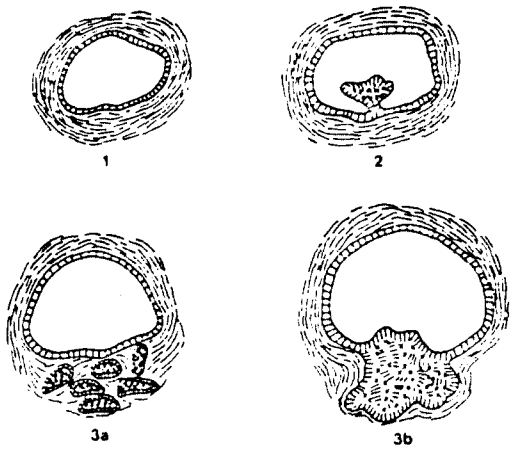


Fig. 1. Histologic types 1. Group 1-cyst lined by variable. often nondescipt, epithelium with no infiltration into fibrous cyst wall. 2. group 2-cyst showing intraluminal plexiform epithelial proliferation with no infiltration. 3. Group 3-cyst with invasion of epithelium into the cyst wall in either(a) a follicular of (b) a plexiform pattern. (Cited from Ackermann, G.L ; The unicystic ameloblastoma. J. Oral Pathol. 17 : 541, 1988)

later expanded by Gardner.[6,12](figure 1)

It is estimated that unicytic ameloblastoma constiue 5~15% of all ameloblastomas.[3,13] Most unicystic ameloblastomas resemble dentigerous cysts clinically and radiologically, but a few are not associated with embedded teeth and therefore resemble premordial or residual cysts. They appear to occur exculusively in the mandible, with a distinct predilection for the third molar region.[3,6,12]

Unicystic ameloblastoma occur most commonly in the second & third decades of life, which is considerablelly younger than the average age of discovery for the classical ameloblastoma.[3,6,12,13] Although all ameloblastoma population resembles in terms of predilections for sex and site, the average age of the patients with unicystic ameloblastoma, in comparison with the total group, was 21.8 vs 38.9 years.[3] Unicystic ameloblastomas were either asymptomatic or discovered on routine radiographic examination or the patients noted an enlargement of the jaw without pain or paresthesia. The lesions were found to occur in younger patients when associated with an impacted tooth than in cases with no impaction radiographically. Unilocular, scalloped, and multilocular patterns with well-delineated margins were observed. Shteyer & Lustmann estimated the recurrence rate of unicystic ameloblastomas to be less than 10% after enucleation.[3,7,13]

If the ameloblastomatous epithelium simply lines the cystic lumen or projects into the lumen, without involving the connective tissue wall in both cases, the unicystic ameloblastoma can be expected to be removed completely by enucleation and recurrence is unlikely. On the other hand, if there is proliferation of the ameloblastoma lining the cystic lumen into the periphery of the connective tissue wall, or there are islands of ameloblastoma in the wall, then more ameloblastoma may be present in the cancellous bone surrounding the cystic lesion and recurrence is possible. The posterior maxilla represents a particularly dangerous location for potentially invasive tumor. Therefore, surgeons may understandably be reluctant to confine their treatment to enucleation in this site even when dealing with unicystic ameloblastomas. In such cases, marginal resection should be employed.[9]

Robinson and Martinez recognized 3 histological types. Later Gardner added a fouth histological sub-type and pointed out that there was a difference in biological behaviour between those lesion that were simply cystic or with intraluminal epithelial proliferation and those with the epithelium proliferating outward breaching the fibrous wall, having the capacity to invade adjacent cancellous bone.[6,12,13] While Group 1 and 2 lesions may be treated by enucleation, Group 3 lesions should be treated aggresively as for conventional ameloblastomas.[13]

The true nature of the lesion may become evident only when the entire specimen is availale for examination. In addition, incisional biopsies often does not allow correct classification of the lesion as one of the 3 subtypes. With this in mind, we recommend that incisional biopsies be avoided. Three histological types are recognized which have an important influence on biological behaviour, treatment and prognosis. The objectives of correct histological diagnosis, classification and appropriate therapy are best achieved by an enucleation biopsy. In

the case of Group 3, further surgery will be necessary.[12, 13]

As in case 1, clinically and radiographically ameloblastoma was suspected, the biopsy result showed dentigerous cyst and the pathologic report after complete enucleation was unicystic ameloblastoma. The receiving of the facial asymmetry, caused during the healing process, by mandible regaining its normal shape is considered to be good due to the young age of the patient and the histologic feature being unicystic ameloblastoma. In case 3, unicystic ameloblastoma was diagnosed and further aggresive surgery was in store, but considering the age of the patient, the possibility of recurrence must be explained to the patient and the progress is periodically observed.

There is one other point that requires emphasis concerning the treatment of apparent recurrences of ameloblastomas in general. A radiolucency in the jaws in the site of previous surgery for ameloblastoma, even several years after the operation, is not necessarily a recurrent tumor. Replacement by bone may have been slow, and the radiolucency may simply represent fibrous connective tissue. Atraumatic neuroma may also occur. It is essential that biopsies be performed in these apparent recurrences to avoid unnecessarily extensive surgery for lesions that are shown subsequently on histopathologic examination to contain no ameloblastoma.[18, 19] Intermaxillary fixation was frequently employed postoperatively in the treatment of large lesions in order to prevent pathologic fracture.[7]

Ⅳ. SUMMARY

An accurate diagnosis is essential because of the difference in choice of treatment in ameloblastoma. Also the surgeon must be considered the anatomic location of the lesion and patient age. For the accurate histopathological diagnosis of unciystic ameloblastoma, the specimen obtained from the excisional biopsy, complete enucleation or incisional biopsy from the multiple site of the lesions.

When we have been treated the unicystic ameloblastoma, the operators must be close long period follow-up check because of the recurrence possibility of the lesion eventhough the unicystic ameloblastoma has been known as low recurrence rate rather than solid or multicystic ameloblastoma.

REFERENCES

1. Bhaskar, S.N. : Synopsis of oral pathology, 6th ed. : 224, Mosby. St. Louis, 1981.
2. Kruger, G.O. : Textbook of oral & maxillofacial surgery, 6th ed. : 645~648, Mosby. St. Louis, 1984.
3. Shteyer, A., Lustmann, J. : The mural ameloblastoma. J Oral Surg. 36 : 866, 1978.
4. Stanley, H.R., Dieh., D.L. : Ameloblastoma potential of follicuar cysts. Oral Surg. 20 : 260, 1965.
5. McMillan, M.D. : Ameloblastomas associated with dentigerous cysts. oral Surg. 51 : 489, 1981.
6. Robinson, L., Martinez, M.G. : Unicystic ameloblastoma. Cancer. 40 : 2278, 1977.
7. Leider, A.S., Eversole, E.R. : Cystic ameloblastoma. Oral Surg. 60 : 624, 1985
8. Gardner, D.G. : Plexiform unicystic ameloblastoma. Cancer. 47 : 1358, 1981.
9. Gardner, D.G., Pecak, A.M.J. : The treatment of ameloblastoma based on pathologic and anatomic priciples. Cancer. 46 : 2514, 1980.
10. Caster, D.V., McCully, A.C. : Intracystic ameloblastoma in the young patient Oral Surg. 23 : 127, 1967.
11. Vicker, R.A., Gorlin, R.J. : Ameloblastoma : Delineation of pathologic features of neoplasia. Cancer. 26 : 699,1970.
12. Gardner, D.G. : A pathologist's approach to the treatment of ameloblastoma. J Oral Maxillofac Surg. 42 : 161, 1984.
13. Ackermann, G.L., Altin, M. : The unicytic ameloblastoma. J Oral Pathol 17 : 541, 1988.
14. Dresser, W.J., Segal, E. : Ameloblastoma associated with a dentigerous cyst in a 6-year-old child. Oral Surg. 24 : 388, 1967.
15. Wilson, D.L., Roche, W.C. : Dentigerous cyst with a ameloblastomatous change, J Oral Surg. 18 : 173, 1960.
16. Hutton, C.E. : Occurence of ameloblastoma within

a dentigerous cyst. Oral Surg. 24 : 147, 1967.
17. Waldron. C.A. : Ameloblastoma in perspective. j Oral Surg. 24 : 331, 1984.
18. Gardner, D.E., Corio, R.L. : Plexiform unicystic ameloblastoma. Cancer. 53 : 1730, 1984.
19. Kim, J., Choi, I.J. : Ameloblastoma arising in odontogenic cysts. K J Path. 20 : 435, 1986.

국문초록

낭종성 법랑아세포종

- 문헌고찰 및 증례 -

연세대학교 치과대학 구강악안면외과학교실, 구강병리학교실*
이의웅 · 박형식 · 차인호 · 김 진*

치성낭종과 법랑아세포종과의 조직학적 기준이 모호한 경우에 과거에는 병리학자들이 진단하는데 어려움이 많았으나 1970년 Robinson & Martinez가 조직학적 기준을 제시하면서 unicystic ameloblastoma라 명명하였다. Unicystic ameloblastoma는 conventional ameloblastoma 보다 젊은 연령인 10대, 20대에서, 하악 제3대구치부에서 호발하며 방사선학적으로 단방성 혹은 다방성의 비교적 경계가 뚜렷한 방사선 투과성 병소를 보인다. 이 병소는 적출술만으로 치료하였을 때 solid or multicystic ameloblastoma보다 현저히 낮은 재발율을 보인다.

임상적, 방사선학적으로 unicystic ameloblastoma가 의심되면 병소를 완전히 적출하여 정확한 병리조직학적 진단을 하여 solid or multicystic ameloblastoma일 때는 2차적으로 광범위한 수술 등을 고려하는 것이 타당할 것으로 사료되어 문헌고찰과 함께 3증례의 치료 경험을 보고하는 바이다.