UNICYSTIC AMELOBLASTOMA: A NEW ENTITY?

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SUMMARY

This article consists of two selected case reports of a recently named odontogenic tumour, unicystic ameloblastoma. The clinical and radiographic findings of the two cases mimic that of odontogenic cysts but not dentigerous cysts as in most reported cases. Histologically, either a normal or ameloblastomatous cyst lining is evident. Other features of ameloblastoma are present within the cyst wall or as luminal nodules within the cystic space. A review of the literature indicates that this is a non-aggressive tumour with a low recurrence rate.

INTRODUCTION

Since 1925, many have reported the development of ameloblastoma within the walls of odontogenic cysts and the most commonly cited is the dentigerous cyst. A variety of terms have been given to them including mural ameloblastoma,[1] luminal ameloblastoma, ameloblastoma arising from dentigerous cyst as well as those arising from radicular cyst. The term unicystic ameloblastoma was coined because this is thought to be a distinct entity with a completely different behaviour from conventional ameloblastoma (solid/multicystic).[2]

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By definition, this is a unilocular or multilocular radioluscency which mimics an odontogenic cyst, in many instances a dentigerous cyst clinically. Histologically, there is evidence of a cyst lining epithelium with either ameloblastomatous proliferations within the cyst wall or as epithelial intraluminal projections.[2] These lesions are mainly located in the mandible in most cases although a few have been reported to be in the maxilla.

The purpose of this report is to describe two additional cases of unicystic ameloblastoma which do not mimic a dentigerous cyst.

CASE REPORTS

Case 1

A 17-year-old Chinese female patient presented at the Dental Faculty in December 1982 with pain and swelling around the lower left quadrant of two months duration. No facial deformity was observed but intraorally, bucco-lingual expansion of bone was evident.

Radiographically, there was a unilocular radiolucent lesion measuring 4 cm x 2 cm within the lower left quadrant with evidence of root resorption of both teeth 45 and 47 (Fig. 1). Aspiration biopsy revealed a straw-coloured fluid. A

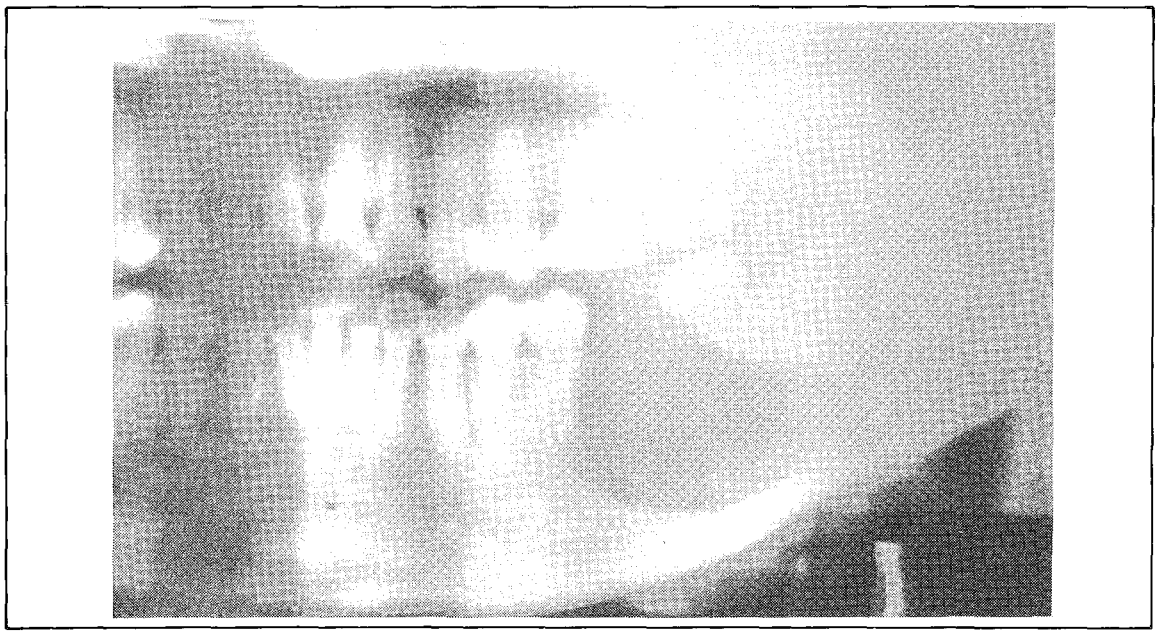

Fig. 1 Radiographic appearance of Case 1 resembling a residual cyst.

clinical diagnosis of a residual cyst (radicular cyst) was made and the lesion was enucleated.

Histologically, there was an ameloblastomatous lining epithelium. Odontogenic epithelial islands were present within the cyst wall. Luminal projections of ameloblastomatous proliferations were also evident (Fig. 2).

No recurrence was observed approximately 18 months after surgery. Evidence of bone filling up the defect was present.

Case 2

A 30-year-old Chinese female patient presented with pain and a fluctuant swelling of the right alveolar region intraorally. She had undergone surgery on three previous occasions, the details of which are not known.

The radiographic examination revealed a multilocular radioluscency with clear radiopaque margins. Root resorption of tooth 43 was evident (Fig. 3). Tooth 43 was non-vital. A straw-coloured fluid was obtained from the lesion on aspiration

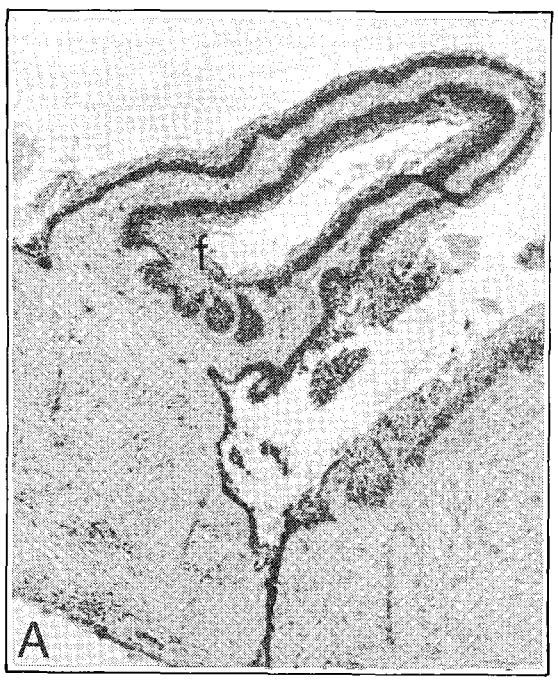

Fig. 2 Photomicrograph of case 1 showing:
A: part of a cyst lining with ameloblastomatous features. An island of ameloblastomatous epithelium (f) is also evident (original magnification A-13x).

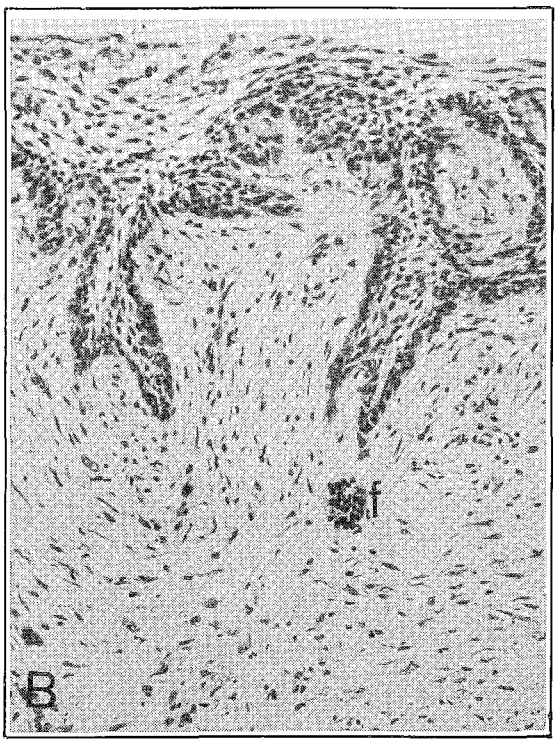

Fig. 2B: a higher magnification of the ameloblastomatous epithelium proliferating into the connective tissue (original magnifications: B--33X).

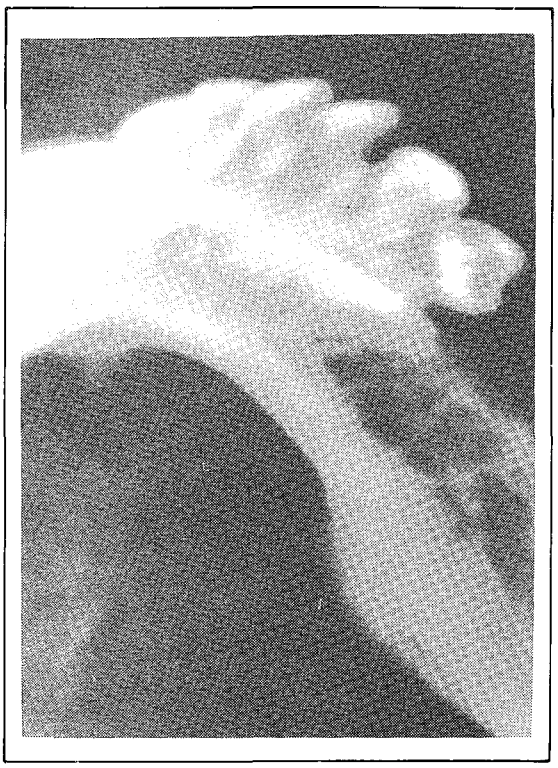

Fig. 3 Radiographic appearance of Case 2 with root resorption of tooth 43.

biopsy. The contents of the lesion consisted of protein – 6.4 g/100 ml, cholesterol – 170g/100ml and albumin – 2.9g /100 ml.

A clinical differential diagnosis of a radicular cyst and an odontogenic keratocyst was made. An incisional biopsy was done which revealed the lesion to be suggestive of a unicystic ameloblastoma. A final enucleation was done.

Histologically, a normal cyst lining consisting of stratified squamous epithelium was evident. Islands of odontogenic epithelium were present within the cyst wall.

A follow-up period of two years showed no recurrence. Radiographically, bone regeneration was evident at the surgical site.

DISCUSSION

In both cases, the radiographic presentations are that of benign cysts most suitably residual (radicular) cyst and odontogenic keratocyst. The contents of these cyst-like lesions composing of a straw-coloured fluid, protein, albumin and cholesterol are more in keeping with that of radicular cysts (residual cysts depending on radiographic presentation).[2]

The diagnosis of conventional ameloblastoma most often would alert the clinician into a radical treatment approach due to its reportedly high recurrence rate. The histological features of conventional ameloblastoma would not be a guide to treatment. In unicystic ameloblastoma, the histological picture appeared to be quite significant when considering the mode of treatment.

Gardner[3] proposed two methods of treatment namely enucleation or enucleation with a marginal resection/close follow-up. The histological findings should aid in the choice of treatment. In most instances, enucleation only would have been carried because a clinical diagnosis of a benign cyst had been made. However, if the ameloblastic epithelium had penetrated the periphery of the cyst wall, the second approach would be preferred to prevent a recurrence.

In both cases reported here, the enucleated tissue did not show the penetration of the tumour into the periphery of the cyst wall. These lesions had been followed up for a period of at least 18 months and there were no evidence of any re-currences. Although there is a general acceptance that this is a non-aggressive tumour with a low recurrence rate,[1,2,3] it is still too early to be confident of such behaviour. It is thus very important that clinicians be aware of the existence of unicystic ameloblastomas which do not warrant radical and mutilating treatment as have been done for the conventional ameloblastomas. A close follow-up of at least five years is mandatory.

REFERENCES

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