Ameloblastoma in Hong Kong Chinese

CSP Poon, PC Wu, MKP So

A total of 50 ameloblastomas in Hong Kong Chinese were received from January 1980 through December 1992 by the Oral Pathology Unit, Department of Pathology, Prince Philip Dental Hospital, Hong Kong. There were eight cases (16%) of unicystic and 42 cases (84%) of classical ameloblastoma. The mean age of the former group (22.1 years) was one decade younger than that of the latter (32.1 years). Otherwise, the slight male predominance, mostly mandibular location, size variation, and histological patterns were similar to those found in other series of ameloblastoma. In the unicystic group, fibrous cyst wall invasion by epithelial strands was observed in all cases. The implications of these findings in diagnosis and management are discussed.

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Key words: Chinese; Ameloblastoma; Mandible; Odontogenic tumours

Introduction

Ameloblastoma is a benign but locally invasive neoplasm derived from odontogenic epithelium. The WHO histological typing of odontogenic tumours classifies ameloblastomas as intra-osseous central, and extra-osseous peripheral types.[1] The central type involves predominantly the mandible, with reported frequencies[2,3] ranging from 71% to 80%. The small number of ameloblastomas arising directly from the surface epithelium or from residues of the dental lamina lying outside the bone constitute the peripheral type. Within the central type, the unicystic variant is recognised as a clinically, radiologically, and pathologically distinct entity with prognostic significance that warrants alternative management to the classical central type.[4] Our previous study[5] shows that odontogenic tumours comprise 72% of all jaw bone tumours in Hong Kong Chinese and central ameloblastoma constitute 62% of all odontogenic tumours. In this study, we analysed data for the period January 1980 through December 1992. The clinicopathological characteristics and the differences between the classical and unicystic groups are emphasised.

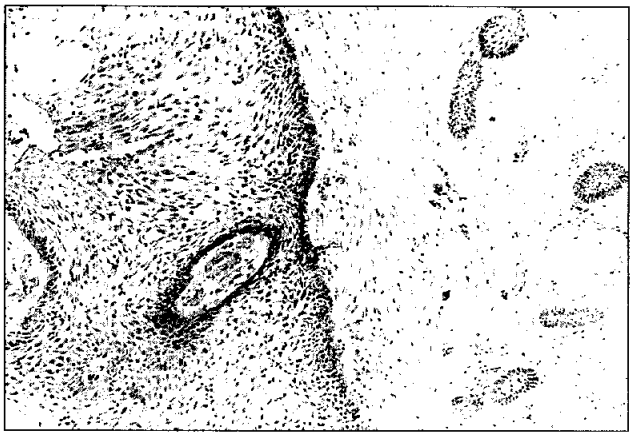

Fig 1. Unicystic ameloblastoma showing invasion of the fibrous cyst wall by epithelial strands (H&E, x 75)

Subjects and methods

The pathology of all ameloblastomas received by the Oral Pathology Unit, Prince Philip Dental Hospital, Hong Kong, during the study period were reviewed. Haematoxylin and eosin and relevant special stains of paraffin sections were independently studied by the first two authors with final overall consensus. The pathological features were analysed by SPSS (statistical package for social sciences).

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Table 1. Age of patients according to sex, tumour subgroup, and site

	Sex		Tumour type		Tumour site		
	Male	Female	Unicystic	Classical	Mandible	Maxilla	Total
No.	29	21	8	42	8	42	50
Mean age (y)	31.3	29.3	22.1	32.1	27.9	44.0	30.5
95% CI*	23.9-38.7	23.0-35.6	16.6-27.6	26.4-37.8	22.8-33.0	30.1-57.9	25.6-35.4
Median age (y)	22.0	29.0	20.0	26.0	23.0	45.0	24.0
Age range (y)	3-73	11-68	13-37	3-73	3-73	19-70	3-73

*95% confidence interval for the mean

The criteria used in the diagnosis of ameloblastoma followed those of Vickers and Gorlin,[6] namely, peripheral palisading of antipodal nuclei, nuclear hyperchromatism, and cytoplasmic vacuolation. The unicystic variant was diagnosed when a unicystic lesion was confirmed on pathological examination (Fig 1).[7] No extra-osseous (peripheral variant) ameloblastomas were diagnosed in this series.

Results

There were a total of 50 ameloblastomas. Eight of the 50 (16%) were unicystic (group U). The other 42 (84%) were of the classical type (group C) with solid, multilocular, or combination solid/multilocular forms.

Sex and age

Overall, there was a slight male predominance with a male to female ratio of 1.4:1. The mean age at presentation was 30.5 years (range, 3-73 years). No significant difference in presenting age was found between the sexes (Table 1). Group U had an equal male to female ratio while group C had a ratio of 1.5:1. In group U, the mean incident age (22 years; range, 13-37 years) was one decade younger than that of group C (32 years; range, 3-73 years).

Tumour site

Twenty-four tumours (52%) occurred on the right side and 22 (48%) occurred on the left. In four cases, the pathology specimen received did not disclose the exact site or the side where the tumour came from. Forty-two (84%) came from the mandible and eight (16%) from the maxilla (Table 2). Thirty-two (76.1%) of the mandibular tumours were posteriorly located in the premolar and molar regions; six cases (14.3%) occupied the anterior incisor region and two tumours (4.8%) extended across the midline symphysis, with one having replaced almost the entire mandible. The eight maxillary tumours also demonstrated a predominantly posterior location with five tumours (62%) occupying the pre-molar and molar regions, including an extensive one that straddled the symphysis. All of the unicystic ameloblastomas were located in the premolar and molar sites, with seven (87.5%) in the posterior mandible and one (12.5%) in the posterior and anterior maxilla.

Tumour size

Sizes ranged from 1.5 cm to 13.0 cm, with a mean of 4.2 cm. Tumour size was not related to sex, age, or tumour site. No significant difference in size was observed between the classical and unicystic types.

Table 2. Distribution of ameloblastomas according to tumour subgroup

Site	Unicystic No. (%)	Classical No. (%)	Total No. (%)
Maxilla	1(12.5)	7(16.7)	8(16.0)
- Anterior*	0	2	2
- Posterior†	0	5	5
- Extensive	1	0	1
Mandible	7(87.5)	35(83.3)	42(84.0)
- Anterior	0	6	6
- Posterior	7	25	32
- Extensive	0	2	2
- Unknown	0	2	2
Total	**8(100)**	**42(100)**	**50(100)**

* Anterior incisor and canine regions
† Posterior premolar and molar regions

Table 3. Histopathology of 50 cases of ameloblastoma and relation to margin

Histology	Classical (Group C) No. (%)	Unicystic (Group U) No. (%)	Exp*	Inf†
Follicular	12(28.6)	3(37.5)	5	6
Plexiform	15(35.7)	4(50)	14	5
Mixed	13(31.0)	1(12.5)	5	8
Desmoplastic	2 (4.7)	0	0	2
Total (%)	42(100)	8(100)	‡24(53)	21(47)

* Exp expansile margin
† Inf infiltrative margin
‡ Information for five cases was not available

Form

Eight cases were of the unicystic type. In group C, 11 appeared solid, 8 were multicystic, and 15 showed a mixture of cystic and solid areas. In eight cases, only small biopsy samples were available for pathological examination and the macroscopic picture of the tumour could not be ascertained.

Histopathology

The distribution of various histological patterns is shown (Table 3). Of the 42 classical cases, pure follicular, pure plexiform, and mixed follicular/plexiform types formed the majority (94%). Two tumours exhibited a desmoplastic pattern with mixed follicular and plexiform features. Acanthomatous change in the form of intercellular bridges and keratinisation were observed in 17 cases (34%). Basaloid cells were prominent in seven cases (14%). Densely hyalinised stroma was present in eight cases (16%). The latter differed from the pure desmoplastic variant[8] in that abundant stroma and stromal hyalinisation occurred only focally in parts of the tumour and the epithelial element was not characteristically compressed by the fibrous stroma. These eight cases were excluded from the desmoplastic group, although they may be regarded as hybrids.

In the unicystic tumours, a pure follicular pattern (3 cases, 37.5%) and a pure plexiform pattern (4 cases, 50%) predominated. The remaining case (12.5%) had a mixed follicular and plexiform pattern. In all cases, focal areas of the epithelial lining comprised nondescript epithelium only, often with focal inflammation, ulceration, and granulation tissue formation. The true nature of these cysts may therefore be obscured as a result of the sampling error inherent in small biopsies. The unicystic group was more frequently associated with root resorption (51%) and impaction (37.5%) of teeth than was the classical group (7.1% and 11.9%, respectively).

Invasiveness

a) Mode of growth

Tumour growth was categorised as being either expansile or infiltrative. A tumour was considered expansile if its margin showed a broad and smooth pushing front. Those tumours that permeated into adjacent tissue for significant distances were termed infiltrative. The distribution of histological types and mode of growth are shown in Table 3. Follicular and plexiform histological types were found in similar proportions of expansile and infiltrative lesions in group C. The two desmoplastic variants showed an infiltrative front. The unicystic group, however, mostly displayed an expansile pattern (75%). An infiltrative margin was found in two cases (25%), one with a pure follicular and the other with a mixed histological pattern.

b) Bone invasion

Twenty-six cases (52.0%) showed intertrabecular bone infiltration into the adjacent cancellous bone, subperiosteal invasion, or extra-osseous spread to the submucosa and mucosa. Co-existence of these features was frequent. Of the 10 cases (20%) that showed intertrabecular growth, five were associated with extra-osseous submucosal and mucosal invasion and three had breached the bone cortex, being limited only by periosteal new bone. Two cases in the unicystic group showed breaching of the alveolar plate with reactive new bone formation and mucosal invasion. Intertrabecular infiltration was not identified in this

Table 4. Clinicopathological features of four recurrent ameloblastomas

Patient (Sex)	Age 1* (y)	Age 2†	OT1‡	Site	Histology	Tumour margin	Bone invasion
1 (M)	nk	70	nk	Maxilla	Mixed follicular, plexiform squamous	Expansile	Nil
2 (F)	9	26	Curettage	Mandible -anterior	Follicular basaloid	nk	nk
3 (F)	11	32	Curettage	Mandible -anterior	Follicular squamous§	Infiltrative	Breached cortex
4 (M)	18	22	Enucleation	Mandible -posterior	Follicular squamous	Infiltrative	Intertrabecular+ breached cortex

* Age 1 age at first presentation
† Age 2 age at recurrence
‡ OT 1 type of first operation
§ Squamous squamous metaplasia
nk not known

group, but all cases showed invasion of the cyst wall by islands and follicles of ameloblastoma.

Recurrence

Four tumours had recurred, two following curettage and one after a simple enucleation performed previously. The details of previous operation in one case were unknown. The clinicopathological features of these tumours are shown in Table 4.

Discussion

The slight male predominance in our series (1.4:1) concords with the results of a previous study by us (1.2:1).[5] The ratio has been relatively stable for the past 30 years. A similar sex distribution was observed in the series conducted by Waldron et al[8] (1.2:1) and by Gunhan et al[2] (1:1). In these two studies, the average patient age was 43.8 and 36.7 years, respectively. The mean age (32.1 years) of our patients with the classical type of ameloblastoma was considerably younger. Whether or not ethnic background has a role to play is not clear.

The age of a patient at tumour presentation is affected by both subjective and objective factors. The former is influenced by the patient's awareness of the lesion and tolerance of symptoms. These variables are difficult to quantify. More objective are the tumour incipient age, its growth rate, and complications. A tumour that starts growing at an early age, given the same growth rate, would definitely present earlier. Complications such as haemorrhage or fluid accumulation with cystic changes cause a rapid increase in tumour size and hence, an earlier presentation. The younger age (mean) of our patients with classical ameloblastoma (32.1 years) suggests that ameloblastomas in Chinese people start earlier in life.

A later occurrence of maxillary ameloblastomas (mean, 44 years) compared to mandibular tumours (mean, 30.9 years) was observed in this series. There was no significant difference in the size of the tumours in these locations. Since the onset of the tumour could not be reliably calculated from the duration of symptoms offered by patients, it is possible that maxillary tumours grow more slowly than do their mandibular counterparts, or, more likely, that they occur later in life.

Ueno et al[9] report the major prognostic factors of ameloblastomas to be: mode of initial operation, histology, age, and radiological appearance. Factors that favour recurrences include inadequate treatment, a follicular histological pattern, older age (older than 20 years) and the presence of multilocular lesions on radiology. In our recurrent cases, the initial operations had been curettage in two, enucleation in one, and one was unknown. A follicular pattern occurred in three and a mixed pattern in one; squamous metaplasia was present in three cases and basaloid cells in one. The age at first presentation was known in three cases, all being below the age of 20 years. Because of our find-

ing of infiltrative growth into adjacent soft tissue and bone invasion in approximately 50% of cases, the importance of adequate removal is obvious. In the series of unicystic ameloblastoma investigated by Wang et al,[10] a high recurrence rate of 57% after simple curettage was observed. Our finding of cyst wall invasion in all unicystic variants suggests that simple curettage of this variant is likely to be inadequate in at least a significant proportion of cases, and thus carries a high risk of recurrence, similar to the classical type of ameloblastoma.

In summary, ameloblastomas in Hong Kong Chinese are similar to other series as regards sex distribution, site predilection, size variation, and histological patterns. The reason for the younger age of our patients, both of the classical and unicystic type, is obscure. The frequent observation of cyst wall invasion in our unicystic ameloblastomas may warrant more aggressive treatment than curettage for this variant to prevent recurrences.

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