RESEARCH

Keratocystic odontogenic tumour in a Hong Kong community: the clinical and radiological features

DS MacDonald-Jankowski*,¹ and TK Li²

¹Division of Oral & Maxillofacial Radiology, Faculty of Dentistry, University of British Columbia, Vancouver, Canada; ²Head of Oral Radiology, Faculty of Dentistry, University of Hong Kong, Hong Kong, China

Objectives: The aim of this study was to evaluate the clinical and conventional radiological features of a consecutive series of cases of “keratocystic odontogenic tumour” (KCOT) affecting a Hong Kong Chinese community and to determine their outcome by follow-up.

Methods: All cases were accompanied by appropriate radiography and were histopathologically confirmed.

Results: 33 consecutive KCOTs were reviewed. 18 patients were male. The mean age at first presentation was 30.6 years. Swelling was the most frequent presenting symptom. Those patients first presenting with pain were significantly older, whereas those first presenting with a maxillary lesion were significantly younger. The maxilla and mandible were affected in 13 and 20 cases, respectively. KCOTs were most frequently confined to the posterior sextants of both jaws. KCOTs affecting the maxilla were mainly unilocular, whereas those affecting the mandible were multilocular. Patients with multilocular KCOTs were significantly older. Patients with KCOTs associated with root resorption were significantly older, whereas patients associated with unerupted teeth were significantly younger. 69% displaced teeth, 41% resorbed them and 56% were associated with unerupted teeth. All but two were followed up for at least 2 years. Three lesions recurred.

Conclusions: KCOTs in this community displayed some differences from those reported in the literature.

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Introduction

The “odontogenic keratocyst” (OKC), first named by Philipsen, has recently been renamed by him as the “keratocystic odontogenic tumour” (KCOT) and reclassified as an odontogenic neoplasm by the World Health Organization (WHO).¹ According to the WHO’s 2005 edition of the histological classification of odontogenic tumours, the KCOT has been defined as “A benign uni- or multicystic intraosseous tumour of odontogenic origin, with a characteristic lining of parakeratinized stratified squamous epithelium and potentially aggressive, infiltrative behaviour. It may be solitary or multiple. The latter is usually one of the stigmata of the inherited naevoid basal cell carcinoma

syndrome (NBCCS).”¹ Furthermore, to emphasize the essential parakeratotic feature of this new tumour, Philipsen adds, “Cystic jaw lesions that are lined by orthokeratinizing epithelium do not form part of the spectrum of a ... KCOT.”¹

The ethnic and environmental background of the Hong Kong Chinese and some of the pathology affecting their jaws have recently already been outlined.² Although Lam and Chan³ published a report on keratocysts affecting the Hong Kong Chinese in 2000, this was essentially a histopathological report and did not consider the clinical and radiological presentations. The details of those clinical features it reported (gender, age and site) were compromised by the authors’ failure to separate them out according to histopathological type. Furthermore, although their cases were derived from otorhinolaryngology and oral maxillofacial units over an unknown period, it was clear that the whole

*Correspondence to: Dr DS MacDonald, Division of Oral & Maxillofacial Radiology, Faculty of Dentistry, UBC, 2199 Wesbrook Mall, Vancouver V6T 1Z3, BC, Canada; E-mail: dmacdon@interchange.ubc.ca

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KCOT caseload diagnosed and treated at the Prince Philip Dental Hospital (PPHD, which also houses the University of Hong Kong's Faculty of Dentistry) and its associated Oral and Maxillofacial Surgery Unit (in Queen Mary hospital) had not been included.

Aims and research question

It is the aim of this study to report a consecutive series of solitary KCOTs so that their full clinical and conventional radiological picture can be reviewed. This is important as Hong Kong Chinese are already known to display features in three benign odontogenic neoplasms⁴⁻⁷ that differ from other communities. Therefore, a review of this newly recognized odontogenic neoplasm affecting this community is merited. The research question is "What are the clinical and radiological characteristics of solitary KCOTs observed in a Hong Kong Chinese community?"

This requires a detailed analysis of the clinical and radiological features observed in a continuous series of cases of solitary KCOT lesions in a Hong Kong Chinese community admitted to PPHD.

Materials and methods

The histopathological files of PPHD between January 1989 and December 2004 were reviewed for KCOTs. The clinical notes and radiographs of each case were retrospectively reviewed. In order to diminish the effects of "expectation bias", which is intrinsic to a retrospective review of cases, the radiographs were read prior to the clinical notes. Each patient's gender, age, clinical history and findings on examination and the differential or provisional diagnosis were obtained from the clinical records. The definitive diagnosis of a KCOT was made on the basis of the histopathology. Each KCOT was radiographed in two planes. Panoramic and periapical radiographs were available for all cases. Every case affecting the body of the mandible was accompanied by a true occlusal radiograph. Those cases, which substantially affected the vertical ramus, were investigated by anterior-posterior projections. Periapical or occlusal radiographs were available for all lesions in the anterior sextants of both jaws. Occipitomental and lateral sinus views had been obtained for those cases involving the maxillary antrum. The radiographs were viewed on a standard illuminated screen, under reduced ambient lighting. The influence of the KCOT on adjacent structures, such as the teeth, the buccal and lingual cortices, lower border of the mandible and the maxillary antrum, was noted.

Measurements on panoramic radiographs are subject to "unequal magnification and geometric distortion across the image."⁸ In order to produce an indication of mesiodistal extent of the KCOT, the authors used "units"

based on whole and half dental units. Each tooth counts as one dental unit, except that the two lower incisors counted as one unit. The distance between the third molar and mandibular foramen, and between the mandibular foramen and the sigmoid notch, each counted as one unit. The condylar and coronoid processes counted as one unit each. Although these "dental units" are arbitrary, they are based on anatomical features that are common to almost all individuals. Therefore, this may make for a better comparison between individuals than measurements from a conventional panoramic radiograph which may vary widely not only between patients and units, but at different times for the same patient. This was particularly important, as geometric distortion is a significant phenomenon in panoramic radiography.⁹

Parameters such as marginal definition, the boundaries between the alveolar and basal processes of the jaws, and anterior and posterior sextants have already been defined.²

Significant differences in frequencies were tested by the χ^2 test with $P < 0.05$. Significant differences in age and size were tested by a Student's *t*-test with $P < 0.05$.

Results

33 cases of solitary KCOTs affecting ethnic Chinese were identified for which histopathology reports, radiographs and clinical notes were available. This amounted to 2.1 new KCOTs on average for each year of the study. The number of new patients registering at PPHD from January 1989 to December 2004 was 62537. Therefore, the annual average number of new patients was 4169. The population of Hong Kong over the period including the above time interval increased from 5.5 million in 1985 (by census) to almost 7 million in 2005 (by census).

Details of the gender, age, clinical presentation or complaint, size and distribution, and radiological features of the 33 solitary KCOTs are shown in Table 1. Their provisional and differential diagnoses are set out in Table 2.

The mean age of the 33 patients with solitary KCOTs was 30.58 (SD 16.33) years. The statistical analyses of the mean patient ages for the main clinical and radiological features have been set out in Table 3.

15 cases were referred by general dental practitioners and one by a general medical practitioner; the rest presented themselves to PPHD in the first instance. 26 patients were at least aware of their lesions for a period of time before presentation. The mean period of prior awareness was 0.77 years (SD 1.28 years).

The 12 patients who presented with pain were significantly older than the 21 patients who did not. Pain as a presenting complaint was significantly more frequent in KCOTs affecting the mandible; $\chi^2 = 9.40$: 1 degree of freedom (df); $0.01 > P > 0.001$. Although swelling occurred as a presenting complaint more frequently in the case of KCOTs affecting the maxilla, this was not significant; $\chi^2 = 0.12$: 1df: $P > 0.05$.

Table 1 Horizontal extent of keratocystic odontogenic tumours in the present study

Case number, presenting complaint, period of prior awareness (years); Sex (M/F) and age (years)																			Associated unerupted teeth	
		Right									Left								I	I
Maxilla		Retro-molar	8	7	6	5	4	3	2	1	1	2	3	4	5	6	7	8	Retro-molar	
1. S 0.25	M14													I-----Uni ----(E,A&D)-----I						8
2. S 0.08	M16		I-----Uni----(E,A,RR&D)-----I																	8
3. I 0.04	M16		I-----Uni&scaloped---(E&D)-----I->orbit & post. concha																	8
4. P 0.02	F17		I-----Uni----(E,A&D)-----I																	8
5. D 0.25	M18		I-----Uni----(E,A&D)--- -----I->nose																	8
6. S ING	F18													I-----Uni ----(E,A&D)--- -----I						8
7. SB 2.50	M21												I-----Multi----(E,A,RR&D)--- -----I							8
8. S 0.50	F21							I--Uni-(E&D)-I -> nose												NONE
9. S 0.50	M24													I-----Uni----(E,A,RR&D)-----I						8
10. SD 0.50	F25													I-----Uni -----(E,A,RR&D)-----I						8
11. SD 2.00	F25											I-----Uni ----(E,A&D)-----I								8
12. I 0.06	M28													I-----Uni---(A,RR)-----I						None
13. P 0.04	F 40										I-----Multi (D)---- -I -> nose									None
Mandible		Vertical ramus	8	7	6	5	4	3	2	1	1	2	3	4	5	6	7	8	Vertical ramus	
1. S N/A	M17													I----Multi --(E,RLB(E&B),YYY,D)-- --SN&Con						8&7
2. I ING	F18	SN-----Multi--(E,RLB,YYN)----I																		NONE
3. SP 0.01	M19															I----Multi-(E,RLB,NNN)-----I				NONE
4. SP 1.00	F20																	I--Uni(NT,NNN,D)--MF		8
5. I 0.04	F20	SN-----Multi--(E,NT,YYN, D)-- -I																		8
6. SP N/A	F23	Uni(E,NT,YYN)																		8
7. S N/A	M24	SN-----Multi-(E,RLB,YYN)---- -I																		NONE
8. SP 0.04	F24	SN-Multi&scaloped(E,NT,YYY,RR&D)-I																		8
9. SPN. 0.02	F25	RMulti(E,NT,YYN)																		8Fd
10. SP 2.00	M28													I-Multi -(E,RLB(E&D),YYY,RR&D)-I						NONE
11. S 0.06	M28	MF ----Uni - (E,RLB(E&D),NYY)-----I																		NONE
12. D 3.00	M36	MF-Multi(NT,NNN)																		NONE
13. SP 0.50	F40													I--Uni--(E,RLB, NYY&D)--I						8
14. I ING	M41	MF-----Multi--(E,RLB(E),YYN,RR&D)-----I																		NONE
15. P ING	M47	MF-----Multi ---(RLB,YYY,RR&D)-----I																		NONE
16. P 0.08	M59													I-----Uni---(RLB,YYY,RR)-----I						NONE
17. SP 0.50	M59		I-----Multi---(E,NT,YYY,RR&D)-----I																	NONE

Table 1 Continued

Case number, presenting complaint, period of prior awareness (years); Sex (M/F) and age (years)			Right	Left																Associated unerupted teeth
																				I I
18. P 0.06	F60	MF---Multi-(E,RLB(E),NYY)---I																		edent
19. I 5.50	F61	I-----Multi--- (E,RLB(E&D),YYY,RR&D)-----I																		R4&L4
20. PB ING	M77	I-----Multi---(E,RLB,YYY,RR)-----I																		NONE
			Vertical ramus	8	7	6	5	4	3	2	1	1	2	3	4	5	6	7	8	Vertical ramus

Presenting complaints: S, swelling; P, pain; I, incidental finding; D, discharge; B, bad breath; A, affected antium; N, Numb. Anatomical parameters: A, angle; C, condyle; Cr, coronoid; MF, mandibular foramen; R, ramus (vertical); SN, sigmoid notch. Radiology data codes for each case: Uni, unilocular; Multi, multilocular. All multilocular lesions have a soap-bubble appearance. The presence of "E" means buccolingual expansion (when "E" is absent then no buccolingual expansion). Lower border of the mandible: RLB, reaches lower border (presences of "E" and/or "D" means "erosion" and "displacement", respectively; NT, "not to" (does not reach lower border); inferior dental canal triplet (N, no; Y, yes); first N or Y means narrowing of canal; second N or Y means displacement of canal; third N or Y means that at least part of the canal is absent or not seen; RR, root resorption, D, tooth displacement. Unerupted teeth: edent, edentulous; Fd, fused to bone ankylosis. ING, Information not given.

Table 2 Differential and provisional diagnoses

Histology	No. of cases	Differential diagnosis																
		Cysts					Benign neoplasms							Malignant neoplasms			Keratocystic odontogenic tumour	
		OKC	Dent.	Rad.	TBC	ABC	A.bla	Myx	CEOT	GC	COT	AOT	FOLs	Ca	Sar	Malignant	Alone	Not cited
Total	32*	24	13	6	2	2	21	1	0	2	0	1	1	2	1	0	2	8
Maxillary	12*	9	7	2	0	0	6	0	0	2	0	1	1	1	1	0	0	3
Unilocular	11	8	7	2	0	0	6	0	0	1	0	0	1	1	1	0	0	3
Multilocular	1	1	0	0	0	0	0	0	0	1	0	1	0	0	0	0	0	0
Mandibular	20	15	6	4	2	2	15	1	0	0	0	0	0	1	0	0	2	5
Unilocular	5	3	3	1	0	1	2	0	0	0	0	0	0	0	0	0	0	3
Multilocular	15	12	3	3	2	1	13	1	0	0	0	0	0	1	0	0	2	2

*KCOT did not provide a differential diagnosis (Maxilla case 7).

ABC, aneurismal bone cyst; A. bla, ameloblastoma; AOT, adenoid odontogenic tumour; Ca, carcinoma; CEOT, calcified epithelial odontogenic tumour; COT, calcified odontogenic tumour; Dent, dentigerous cyst; GC, giant cell lesion; OKC, odontogenic keratocyst; Myx, myxoma; Rad, radicular cyst; Sar, sarcoma; FOL, Fibro-osseous lesion.

Table 3 Statistical analysis of ages (in years)

Feature					
Gender	Males – 18	31.78 SD 18.03	Females – 15	29.13 SD 14.51	$t = 0.47$; 31df; $P > 0.05$
Jaw	Maxilla – 13	21.77 SD 6.93	Mandible – 20	36.30 SD 18.20	$t = 3.27$; 31df; $0.01 > P > 0.001$
All maxillary cases	Males – 7	19.57 SD 5.03	Females – 6	24.33 SD 8.38	$t = 1.22$ 11df; $P > 0.05$
All mandibular cases	Males – 11	39.54 SD 19.15	Females – 9	32.33 SD 17.20	$t = 1.07$ 19df; $P > 0.05$
Period of prior awareness	Maxilla – 12	0.56 SD 0.82	Mandible – 14	0.95 SD 1.58	$t = 0.81$; 24df; $P > 0.05$
Swelling	With swelling – 20	25.60 SD 10.42	Without swelling – 13	38.23 SD 20.84	$t = 2.03$; 31df; $P > 0.05$
Pain	With pain – 12	39.00 SD 20.17	Without pain – 21	25.76 SD 11.66	$t = 2.08$; 31df; $0.05 > P > 0.01$
Found incidentally	Found incidentally – 6	33.66 SD 18.31	Not found incidentally – 27	30.38 SD 15.90	$t = 0.16$; 31df; $P > 0.05$
Cases completely confined to a posterior sextant in the mandible	Which were – 13	27.23 SD 11.97	Which were not – 7	53.14 SD 15.90	$t = 3.78$; 18df; $0.01 > P > 0.001$
Expansion	Expanded cases – 27	28.85 SD 16.59	Non-expanded cases – 6	38.33 SD 13.81	$t = 1.46$; 31df; $P > 0.05$
Unilocular or multilocular lesions	Multilocular – 17	36.12 SD 18.70	Unilocular – 16	24.50 SD 11.21	$t = 2.18$; 31df; $0.05 > P > 0.01$
Maxillary multilocular and unilocular lesions	Multilocular – 2	30.50 SD 34.44	Unilocular – 11	20.18 SD 4.65	$t = 1.07$; 11df; $P > 0.05$
Root resorption	With root resorption – 13	36.31 SD 20.63	Without root resorption – 19	25.10 SD 9.33	$t = 2.23$; 30df; $0.05 > P > 0.01$
Tooth displacement	With tooth displacement – 22	27.82 SD 13.92	Without tooth displacement – 10	33.70 SD 19.24	$t = 0.87$; 30df; $P > 0.05$
Unerupted teeth	With unerupted teeth – 20	23.55 SD 10.52	Without unerupted teeth – 12	39.83 SD 17.95	$t = 2.86$; 31df; $0.05 > P > 0.01$
Unerupted teeth between jaws	Maxillary cases – 10	19.40 SD 4.06	Mandibular cases – 10	27.70 SD 13.38	$t = 1.88$; 18df; $P > 0.05$
Cases which did not cite KCOT in their differential diagnosis	Females – 5	31.60 SD 16.46	Males – 3	44.33 SD 28.29	$t = 0.71$; 6df; $P > 0.05$
Jaws: enucleation and Carnoy's solution	Maxilla – 6	24.50 SD 8.22	Mandible – 20	36.30 SD 18.20	$t = 2.24$; 24df; $0.05 > P > 0.01$
Maxilla: enucleation only and enucleation and Carnoy's solution	Enucleation only – 4	19.00 SD 6.20	Enucleation and Carnoy's solution – 6	24.50 SD 8.22	$t = 1.20$; 8df; $P > 0.05$
Follow-up: jaws	Mandible – 20	8.62 SD 4.58	Maxilla – 11	8.09 SD 4.64	$t = 0.32$; 29df; $P > 0.05$

Eight lesions occurred in the posterior maxilla, two in the anterior maxilla, and three crossed slightly from the posterior sextant into the anterior. All mandibular lesions affected at least one posterior sextant. The 13 patients with mandibular lesions confined completely to a posterior sextant were significantly younger than the 7 whose lesions were not. In all patients, mandibular lesions up to and including 25 years of age were completely confined to the posterior sextants (Table 1). Among the 12 older patients only 4 lesions were confined to the posterior sextants; this feature was significant ($\chi^2 = 9.07$; 1df; $0.01 > P > 0.001$). Five cases completely involved not only the anterior sextant, but also the posterior sextants, four bilaterally. Those five cases occurred among the seven oldest patients (from 41 years of age). There was no significant difference between the jaws with regards to the distribution of KCOTs between sextants ($\chi^2 = 0.49$; 1df; $P > 0.05$). Females predominated in those cases

first presenting at younger than 25 years-old, whereas males predominated in those cases first presenting at older than 25 years-old, this difference was not significant ($\chi^2 = 1.85$; 1df; $P > 0.05$).

KCOTs appeared radiographically as well-defined unilocular or multilocular radiolucencies. Those affecting the maxilla were predominantly unilocular (11:2 unilocular-multilocular), whereas those affecting the mandible were predominantly multilocular (Figure 1) (5:15, unilocular-multilocular); this difference was significant ($\chi^2 = 12.23$; 1df; $P < 0.001$).

Although unilocular KCOTs had shorter mesiodistal dimensions (4.22 (SD 1.64) dental units) than the multilocular KCOTs (5.70 (SD 3.48) dental units), this difference was not significant ($t = 1.81$; 31df; $P > 0.05$). The mandibular unilocular KCOTs dimensions were even smaller (3.20 (SD 2.39) units) than the multilocular KCOTs (6.17 (SD 3.70) units); this only tended to significance ($t = 2.08$; 18df; $P > 0.05$).

The 17 patients with KCOTs presenting as multilocular radiolucencies were also significantly older than those with lesions presenting as unilocular radiolucencies. All multilocular lesions displayed a soap-bubble appearance; none displayed a honeycomb or tennis racket appearance.

The KCOT displayed a corticated periphery in 29 cases, except for 2 maxillary and 2 mandibular cases. Erosion of these cortices was observed in three maxillary and seven mandibular cases; this was not significant ($\chi^2 = 0.19$; 1df; $P > 0.05$).

27 lesions exhibited buccolingual expansion (Figure 2): 11 and 16 in the maxilla and mandible, respectively, there was no significant difference between the jaws ($\chi^2 = 0.14$; 1df; $P > 0.05$). In the case of the mandible, the younger patients displayed buccolingual expansion more frequently than the older patients, but the difference was not significant ($\chi^2 = 1.76$; 1df; $P > 0.05$).

13 KCOTs reached the lower border of the mandible (denoted by RLB in Table 1, Figure 3). 4 KCOTs displaced it downwards and/or eroded (thinned) it (denoted by D and E in parenthesis after RLB in Table 1) and a further 2 eroded it without displacement. The inferior dental canal (IDC) was downwardly displaced in 17 cases (narrowing, displacement and absence of the IDC are denoted in that order by the triplets of Y (Yes) or N (No) in Table 1). Although this phenomenon was more frequently observed in patients older than 25 years of age, the difference was not significant ($\chi^2 = 0.58$; 1df; $P > 0.05$). Parts of the IDC associated with the KCOT were absent significantly more often in patients with mandibular lesions who were aged over 25 than in younger patients ($\chi^2 = 9.90$; 1df; $0.05 > P > 0.001$). A reduction in diameter of the IDC was just as frequent in both age groups ($\chi^2 = 0.47$; 1df; $P > 0.05$). Although displacement of the canal was more frequently seen in the older patients, this difference was not significant ($\chi^2 = 1.82$; 1df; $P > 0.05$).

All 11 maxillary cases subjacent to the maxillary antrum displaced the antral floor upwards. One reached the orbit (Figure 4). The two solely affecting the anterior sextant reached the floor of the nose.

There was no significant difference in frequency between the jaws among those cases displaying root resorption ($\chi^2 = 0.91$; 1df; $P > 0.05$). Overall, the 13 patients displaying root resorption were significantly older than the 19 who did not. Tooth displacement was significantly more frequently observed in the maxilla ($\chi^2 = 5.78$; 1df; $0.01 > P > 0.001$). Only one KCOT occurred in a edentulous sextant (Mandibular case 18).

The unerupted tooth most frequently associated with the KCOT was the third molar, and this was the only unerupted tooth associated with a KCOT in the maxilla. The 20 KCOTs associated with unerupted teeth occurred in significantly younger patients than the 12 that were not.

The provisional diagnoses produced by the clinicians on the basis of the clinical and radiological findings alone were available in 32 cases (Table 3); maxillary case 7 had none. Eight KCOTs diagnoses did not cite

OKC (Table 2). The diagnosis of dentigerous cyst was most frequently given when the lesion was associated with an unerupted tooth. The multilocularity of the lesion did not appear to deter the inclusion of dentigerous cyst in the differential diagnosis.

26 cases, 6 maxillary cases and all mandibular cases, were primarily treated by enucleation and Carnoy's solution. One such case recurred after 1 year (mandibular case 5). Although patients with maxillary lesions treated by enucleation and Carnoy's solution were, on average, older than those who were not, they were still significantly younger than those with mandibular lesions cases (Table 3). One maxillary case (case 12) was first decompressed prior to enucleation and Carnoy's solution. Two were resected (maxillary cases 4 and 6). Four cases (maxillary cases 1, 3, 5 and 11) were treated by enucleation alone, very early in the study. Two of the enucleated-only lesions recurred (maxillary cases 1 and 3) after 2 years. Follow-up information was available in nearly all cases, with the exception of maxillary case 8 and mandibular case 9. Three were lost to follow-up, 2 within a year of surgery (mandibular cases 9 and 15) and one after 6 years of follow-up (mandibular case 16). A further 6 patients were discharged or allowed to leave the programme after a mean of 5.33 years (SD 2.42 years) of follow-up. Three of the discharged cases were maxillary (cases 8, 10 and 13) and three were mandibular (cases 6, 8 and 11). All nine lost and discharged patients had been treated by enucleation and Carnoy's solution. They had largely all been lost or discharged during the first half of the study period. The overall mean period of follow-up was 8.32 (SD 4.56 years; range 1–14 years). 20 cases were still being followed up at the time of the conclusion of this study. 5 of them had been followed up for 14 years.

Discussion

Haring and Van Dis¹⁰ suggested that KCOTs begin as unilocular lesions and gradually become multilocular. This report, in part, supports this suggestion in two ways: the fact that the multilocular KCOTs were found in significantly older patients raises the possibility that unilocular cases eventually became multilocular; the KCOTs affecting the maxilla were in significantly younger patients and were more frequently unilocular than those first presenting in the mandible. Nevertheless, inspection of the mandibular cases individually indicated that Haring and Van Dis's¹⁰ conclusions, when applied to this Hong Kong case series, were not entirely clear-cut. Although, on average, mandibular unilocular KCOTs were smaller, and patients with unilocular KCOTs were 3 years younger, than mandibular multilocular KCOTs, only 2 of the 10 youngest patients presented with unilocular radiolucencies in the youngest 10 cases in contrast to 3 of the 10 oldest patients. Therefore, the above-mentioned significant age difference between multilocular and unilocular KCOTs was largely because of the predilection of the maxilla for

unilocular lesions and younger patients on first presentation. The unilocularity of KCOTs affecting the maxilla may occur because the air-filled space offers relatively little resistance to expansion.

Although unilocular KCOTs affecting the mandible tended to be shorter in their mesiodistal extent than multilocular KCOTs, there were no significant differences between them with regard to their buccolingual expansion or their confinement to the posterior sextants. All that can be reasonably said of multilocular KCOTs is that they are more extensive mesiodistally, and their surgery may compromise the integrity and/or vitality of more teeth than unilocular KCOTs.

The significant association between KCOTs presenting at a younger age, and unerupted third molars suggested that, in general, KCOTs affecting the young developed swellings sooner, whereas those affecting the older patient may remain undetected for a long time until pain occurs. The cases not associated with unerupted third molars presenting in older patients arose after the adult dentition, mainly third molars, had erupted.

The role of the primary care dentist is crucial to detection and appropriate referral of oral and maxillofacial lesions. As only six of the KCOTs were detected as incidental findings, the likelihood that substantially more may remain undetected in the community at large is a possibility because of the still widespread lack of professional dental care among the Hong Kong Chinese.¹¹

Philipsen¹ recently stated that root resorption was a rarity in KCOT. This study and that by Haring and Van Dis¹⁰ reported root resorption in 41% and 8% of KCOTs, respectively. These percentages suggest that, although root resorption associated with the KCOT may not be common, it may not be rare in certain communities.

The association with an unerupted tooth appeared to lead to the inclusion of dentigerous cyst within the differential diagnosis. This inclusion was not deterred by the presence of multilocularity. This was likely to reflect the paucity of reported series of cases focusing on the radiology of the dentigerous cyst.¹²

The fact that KCOTs were associated with third molars of both jaws almost equally in the Hong Kong Chinese disagrees with Gonzalez-Alva and co-authors¹³ Japanese report, in which KCOTs were predominantly associated with the mandibular third molars. A reason for this difference was that Gonzalez-Alva and co-authors reported a significantly greater proportion of KCOTs affecting the mandible: 80% compared with 61% in the present report; $\chi^2 = 35.59$; 1df; $P < 0.001$.

The number of KCOTs per year was 2.1 and was lower than the 5.6 ameloblastomas^{6,7} found within the bony jaws per year in the same community. In a European community¹⁴ KCOTs were found almost six times as frequently as ameloblastomas. A mainland Chinese report stated that they occurred almost equally: ameloblastomas and KCOTs account for 40% and 36%, respectively, of all odontogenic tumours in that study.¹⁵ Therefore, the difference in frequency in KCOT can vary markedly between communities.

From the differential diagnoses of the 32 KCOTs in Table 3, it is clear that the ameloblastoma was cited almost as frequently as the keratocyst itself. Although both lesions affecting this community display expansion, the pattern of expansion associated with KCOT is more likely to be fusiform than the balloon-like expansion of the ameloblastoma; compare Figure 2 with MacDonald-Jankowski and co-authors⁷ Figure 4. This pattern of growth arises from the propensity of the KCOT to extend mesiodistally through the body of the mandible, with minimal expansion.¹⁶ This pattern of growth may be less frequently observed in the vertical ramus and very infrequently in the posterior maxilla. A further expansile characteristic that KCOT does not share with the ameloblastoma is the latter's downward expansion of a lateral cortex past the lower border of the mandible; the KCOT only downwardly displaces

Figure 1 This is part of a panoramic radiograph of mandibular case 1. The keratocystic odontogenic tumour occupied the posterior body of the mandible. It presents as a well-defined multilocular radiolucency extending back from the mesial root of the first molar, almost to the sigmoid notch and the base of the condylar process. Its periphery is also largely corticated. It has both displaced the lower border of the mandible downward and eroded it. It is associated with the crown of an unerupted third molar. The cortical margins of the inferior dental canal can be seen faintly below the third molar, but are absent elsewhere. The inferior dental canal can no longer be observed anteriorly and becomes narrowed posteriorly.

Figure 2 This is part of an occlusal radiograph of Figure 1. It displays fusiform-like buccolingual expansion with marked erosion of both buccal and lingual cortices. The roots of the second and third molar teeth are displaced buccally, so that the occlusal surfaces face lingually.

the lower border itself (compare Figures 1 and 2 with MacDonald-Jankowski and co-authors⁷ Figure 1).

The ameloblastoma displayed root resorption in 59% of cases, in contrast to 41% of KCOTs in the same community; this difference was not statistically significant ($\chi^2 = 2.72$; 1df; $P > 0.05$).

Although the number of KCOT patients lost to follow-up was small and similar to the number of ameloblastoma patients in the same community, a further six patients had been discharged or permitted to leave the follow-up programme. Nevertheless, the recurrence of

Figure 3 This is part of a panoramic radiograph of mandibular case 17. This keratocystic odontogenic tumour is a well-defined multi-loculated radiolucency that has extended throughout the body of the mandible. It has reached the lower border of the mandible.

Figure 4 This is part of a panoramic radiograph of maxillary case 3. The second premolar has been displaced distally and the unerupted third molar has been displaced up to the floor of the orbit.

only three cases at PPHD (10% recurrence rate) compares favourably with the 20% recurrence rate found throughout Hong Kong. As Lam and Chan's report³ did not appear to take account of the orthokeratotic and mixed cases, which may be less likely to recur, their recurrence rate may actually be higher. The marked difference between the outcomes in the present report and that of Lam and Chan³ may reflect the long-established expertise and experience of a well-staffed specialist oral and maxillofacial facility at PPHD already intimate with oral and maxillofacial pathology, in comparison with that of another head and neck service specializing in ear, nose and throat rather than in oral and maxillofacial pathology and its management. So far, it appears that those cases treated by enucleation and Carnoy's solution, which quickly became the treatment of choice at PPHD, are less likely to recur. Nevertheless, Lam and Chan's report³ that one case recurred 4 times between 10 and 15 years after initial surgery, and that another recurred twice 20 and 22 years post-operatively, suggests that at least long-term if not life-long follow-up of every KCOT should be undertaken until results of long term follow-up of conservative treatments have been produced.

Conclusions

- (1) Almost half of the cases had first presented to primary care dentists in the community who referred them onward to PPHD for diagnosis and treatment.

- (2) Those KCOTs that presented early were significantly associated with unerupted teeth.
- (3) The majority of KCOTs presented with symptoms, most commonly with swelling. Those patients presenting with pain were significantly older.
- (4) KCOTs present with not only a well-defined, but frequently a corticated, margin.
- (5) Other than multilocular KCOTs' significant predilection for older patients and unilocular KCOTs' predilection for the maxilla, and a tendency for multilocular KCOTs to be longer in their mesiodistal extent, there is no difference between them in extent of buccolingual expansion or confinement to the posterior sextant. The longer mesiodistal extent of multilocular KCOTs may result in the integrity and/or vitality of more teeth being compromised by surgery than in the cases of unilocular KCOTs.
- (6) Although it presents several features observed in the ameloblastoma, there are a number of differences. The KCOT's buccolingual expansion tends to minimally expand the lesion, assuming a fusiform shape, rather than the balloon expansion more typical of the ameloblastoma.
- (7) Although root resorption occurs in a minority of KCOTs, it is not as rare in this Hong Kong community as recently stated.¹ KCOTs associated with root resorption first presented in significantly older patients.
- (8) The majority of KCOTs were associated with unerupted teeth. Such cases first presented in significantly younger patients.
- (9) If appropriately treated, recurrences can be minimized. Three cases recurred out of 31 cases, 2 after 2 years and 1 after 1 year. Two had been treated by enucleation alone. The mean period of follow-up for this case series was 8 years.

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