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Range and demographics of odontogenic cysts diagnosed in a UK population over a 30-year period

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BACKGROUND: The aim of this study was to determine the range of all histologically diagnosed odontogenic cysts along with age range, sex distribution and site of presentation over a 30-year period.
METHODS: All entries for odontogenic cysts occurring during 1975–2004 inclusive were retrieved and analysed for demographic data.
RESULTS: A total of 55 446 specimens were received, of these 7121 (12.8%) specimens were diagnosed as odontogenic cysts. Radicular cyst was the most common diagnosis (52.3%), followed by dentigerous cyst (18.1) and odontogenic keratocysts (11.6%).
CONCLUSIONS: Our study provides demographic data on a large series of odontogenic cysts in a European population. This is one of the largest series reported to date. Cysts such as the paradental cyst have a predilection for certain ages, sexes and sites. Odontogenic keratocysts and glandular odontogenic cysts have a marked propensity to recur as well as behave aggressively. It is essential that such lesions are detected as early as possible to minimize any necessary surgery.
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Introduction

Odontogenic cysts are unique in that they only affect the oral and maxillofacial region. Both inflammatory and developmental odontogenic cysts may develop from epithelium associated with the tooth forming apparatus. Although some of these lesions such as radicular and dentigerous cysts are relatively common and easily diagnosed, other rare cystic lesions such as glandular odontogenic cyst may be less well recognized. Clinical misdiagnosis is possible as both clinical presentation and radiological signs are similar for many of these cysts. A typical example is the odontogenic keratocyst; this can present in a dentigerous relationship and therefore be misinterpreted as a dentigerous cyst. The correct diagnosis of odontogenic cysts is essential as some of these lesions are known to have an aggressive behaviour and a propensity to recur (1, 2). Therefore, all surgically removed tissue should be submitted for histological diagnosis to ensure appropriate treatment is provided.

Most previous reviews of odontogenic cysts have been concerned with specific diagnoses such as paradental cysts (3), odontogenic keratocysts (4), calcifying odontogenic cysts (5, 6), glandular odontogenic cysts (2) and lateral periodontal cysts (7). Demographic studies on odontogenic cysts have been based on populations from Mexico (8, 9), Canada (10) and Germany (11). Other studies on oral and maxillofacial disease have included limited data on the prevalence of odontogenic cysts in all populations (12–16) and in those limited to paediatric patients (17–20).

Owing to the general lack of data in the English language literature on the variety and presentation of odontogenic cysts, the aim of this study was to determine the range of all histologically diagnosed odontogenic cysts along with age range, sex distribution and site of presentation over a 30-year period.

Material and methods

The Department of Oral Pathology in the School of Clinical Dentistry, Sheffield, England, has recorded all acceded specimens into a computerized diagnostic index database for some time. As the date of birth, sex and demographic information as well as site of disease presentation and diagnosis is recorded, it is possible to retrieve data on all of these factors.

Since 1989, data from all specimens received in the department have been prospectively entered into a computer database. Subsequently, data from the files recorded during the period 1973–1988 were computerized; however, some data from this earlier period such as site of presentation, are missing from the database. The structure of the database has been modified several times and a Foxpro™ Windows database is now used.

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Table 1 Rank order distribution of odontogenic cysts by frequency 1975–2004

Diagnosis	*Number*	*%*	*No. of male*	*No. of female*	*M:F ratio*	*Age range (years)*	*Age (±SD) (years)*
Radicular cyst[a]	3724	52.3	1914	1801	1.06	4–100	37.3 (15.1)
Dentigerous cyst	1292	18.1	841	451	1.86	2–83	40.8 (18.1)
Odontogenic keratocyst	828	11.6	464	364	1.27	5–92	41.3 (21.2)
Residual cyst	573	8.0	340	233	1.46	12–89	50.7 (14.8)
Paradental cyst[a]	402	5.6	231	169	1.37	6–74	28.9 (10.9)
Unclassified odontogenic cyst	210	2.9	130	80	1.63	7–83	40.7 (18.9)
Lateral periodontal cyst	28	0.4	16	12	1.33	21–81	48.2 (14.3)
Calcifying odontogenic cyst	21	0.3	13	8	1.63	5–79	41.5 (24.6)
Gingival cyst	16	0.2	4	12	0.33	23–70	52.9 (12.2)
Eruption cyst	15	0.2	10	5	2	1–63	18.8 (17.9)
Glandular odontogenic cyst	11	0.2	8	2	4	31–81	48.5 (17.6)
Epstein pearl	1	0.0	1	0	∞	4	4.0 (0.0)
Total	7121	100	3972	3137	1.27		

[a]In some cases the sex of the patient is unknown.

A member of staff enters initial demographic data when the specimen is received and a different member of staff completes the record when the final report has been issued. In addition, the pathologist responsible for signing out each report monitors further checking of input data. This policy is part of our risk procedures. The diagnoses are entered using an alphanumeric code comprising two letters, which designates the diagnostic category (e.g. OC = odontogenic cyst) and three numbers that refer to the specific condition within the diagnostic category (e.g. calcifying odontogenic cyst = OC201). There are 15 diagnostic categories that contain codes for 627 diagnoses and, as these codes are entered, a 'look-up' table containing the diagnoses is used to avoid the input of typographical errors. If necessary, the codes can be linked via further 'look-up' tables to other coding systems such as Systematised Nomenclature of Pathology or Systematised Nomenclature of Medicine.

All entries for odontogenic cysts from all ages during the 30-year period 1975–2004 inclusive were retrieved and analysed for each diagnosis including number of specimens, male:female ratio, mean age and standard deviation. These data were then analysed for the range of odontogenic cysts for our paediatric population (aged 16 years and under) and adult population (aged 17 years or older). Frequent diagnoses, such as radicular cysts, were subsequently investigated for their most common site of presentation in the mandible or maxilla. All data analysis and graph formation were performed using Microsoft Excel™.

Results

During the 30-year period, a total of 55 446 specimens were received from Hospitals in Sheffield and the South Yorkshire/East Midlands region, occasional hospitals elsewhere and General Dental Practitioners. Of these 7121 (12.8%) specimens were diagnosed as odontogenic cysts. Among these specimens, 553 (7.8%) odontogenic cysts were diagnosed in children 16 years and under and 6385 (89.6%) were diagnosed in adults 17 years or older. No information on age was provided for the remaining 183 cases (2.6%). Of the total cases, there were 3972 specimens from males and 3137 from female patients (M:F = 1.27:1). In 12 cases (0.2%), the gender of the patient was not provided with the clinical details. The site of presentation was known in 5323 (74.8%) of all submitted cases. Table 1 summarizes the rank order distribution of odontogenic cysts by frequency for all ages over the 30-year period. Tables 2 and 3 display the distribution of all odontogenic cysts in paediatric and adult populations respectively. Table 4 summarizes the distribution of odontogenic cysts according to developmental and inflammatory pathogenesis.

The largest diagnostic group was the radicular cyst (Fig. 1) which accounted for 52.3% of all odontogenic cysts (3724 cases) with a male:female ratio of 1.06:1. Of the 2801 cases, where the site of occurrence was indicated, the maxilla was the most commonly affected site with 1996 cases (71.3%), of which 1478 cases (52.8% of all radicular cysts) occurred in the anterior maxillary region (Fig. 2). Radicular cysts were more common in adults (3359 cases) than in paediatric patients (241 cases) accounting for 52.6% and 43.6% of the totals respectively. Residual cysts accounted for 573 cases (8.0%) with the most common site of presentation in the anterior maxilla closely followed by the mandibular molar region (Figs 3 and 4).

Dentigerous cysts were the next most common diagnosis with 1292 (18.1%) cases and a male:female ratio of 1.86:1. The mean age at diagnosis was 41 years, with a gradual increase per decade until the fifth decade after which there is a steady decrease in number (Fig. 5). The site of presentation was known in 1001 cases and the mandible was the most commonly affected site with 817 (81.6%) cases. The lower molar region accounted for 73.2% of cases ($n = 733$) followed by the anterior maxilla with 106 (10.6%) cases (Fig. 6). Dentigerous cysts accounted for a higher proportion of odontogenic cysts in children (28.9%, $n = 160$) than in our adult population (17.4%, $n = 1114$).

Table 2 Rank order distribution of odontogenic cysts by frequency in paediatric populations 1975–2004

Diagnosis	*Number*	*%*	*No. of male*	*No. of female*	*M:F ratio*	*Age range (years)*	*Age (±SD) (years)*
Radicular cyst[a]	241	43.6	129	111	1.16	4–16	13.5 (2.3)
Dentigerous cyst	160	28.9	103	57	0.55	2–16	11.0 (3.1)
Odontogenic keratocyst	100	18.1	53	47	1.13	5–16	13.4 (2.3)
Unclassified odontogenic cyst	21	3.8	12	9	1.33	7–15	11.1 (2.3)
Paradental cyst[a]	15	2.7	8	6	1.33	6–16	11.9 (3.2)
Eruption cyst	8	1.4	5	3	1.67	1–12	7.8 (3.7)
Calcifying odontogenic cyst	4	0.7	1	3	0.33	5–13	11.8 (4.6)
Residual cyst	3	0.5	3	0	∞	12–15	13.7 (1.5)
Epstein pearl	1	0.2	1	0	∞	4.0	4.0 (0.0)
Total	553	100	315	236	1.33		

[a]In some cases the sex of the patient is unknown.

Table 3 Rank order distribution of odontogenic cysts by frequency in adult populations 1975–2004

Diagnosis	*Number*	*%*	*No. of male*	*No. of female*	*M:F ratio*	*Age range (years)*	*Age (±SD) (years)*
Radicular cyst[a]	3359	52.6	1735	1618	1.07	17–100	39.0 (14.2)
Dentigerous cyst	1114	17.4	722	392	1.84	17–89	45.1 (15.0)
Odontogenic keratocyst	717	11.2	402	315	1.28	17–92	45.2 (19.7)
Residual cyst	555	8.7	326	229	1.42	18–89	50.9 (14.6)
Paradental cyst[a]	376	5.9	215	160	1.34	17–74	29.6 (10.5)
Unclassified odontogenic cyst	189	3.0	118	71	1.66	17–83	44.1 (16.9)
Lateral periodontal cyst	27	0.4	15	12	1.25	21–81	48.2 (14.3)
Calcifying odontogenic cyst	16	0.3	11	5	2.20	2–79	51.6 (18.6)
Gingival cyst	16	0.3	4	12	0.33	23–70	52.9 (12.2)
Glandular odontogenic cyst	11	0.2	8	2	4.00	31–81	48.5 (17.6)
Eruption cyst	5	0.1	3	2	1.50	20–63	36.4 (17.6)
Total	6385	100	3558	2818	1.26		

[a]In some cases the sex of the patient is unknown.

Table 4 Distribution of odontogenic cysts according to classification 1975–2004

Diagnosis	*Number*	*% of group*	*% of all cysts*
Developmental cysts	2212	100	31.1
Dentigerous cyst	1292	58.4	18.1
Odontogenic keratocyst	828	37.4	11.6
Lateral periodontal cyst	28	1.3	0.4
Calcifying odontogenic cyst	21	0.9	0.3
Gingival cyst	16	0.7	0.2
Eruption cyst	15	0.7	0.2
Glandular odontogenic cyst	11	0.5	0.2
Epstein pearl	1	0.0	0.0
Inflammatory cysts	4699	100	66.0
Radicular cyst	3724	79.3	52.3
Residual cyst	573	12.2	8.0
Paradental cyst	402	8.6	5.6
Unclassified odontogenic cyst (Unknown type)	210	100	2.9
Total	7121	100	100

There were 828 odontogenic keratocysts (11.6% of all odontogenic cysts) with a mean age at presentation of 41 years and a male:female ratio of 1.27:1. Two peaks of presentation are evident: the first between the ages of 11–30 years and the second between the ages of 51–70 years (Fig. 7). Of the 795 (96%) cases with known site of presentation, the mandible was the most frequently affected site ($n = 545$, 71.1%). The exact site of presentation was known in 766 cases and the single most common location was the mandibular molar region with 236 (30.8%) cases (Fig. 8).

A total of 402 paradental cysts were identified with a mean age at diagnosis of 29 years and a male:female ratio of 1.37:1 (Fig. 9). Of these cases, 340 (84.6%) were located in the mandibular molar area, with one each at the mandibular premolar and maxillary third molar region. No site of presentation was provided for the remaining 60 cases (14.9%). In the respective age groups (Tables 2 and 3), paradental cysts were twice as common in adults than in children (5.9% and 2.7% respectively).

Because of a lack of clinical information provided, 210 (2.9%) cysts were unclassified owing to histopathological diagnostic difficulties in differentiating between an inflamed developmental and a true inflammatory cyst. The remaining 92 odontogenic cysts (1.3%) comprised 28 (0.4%) LPCs, 21 (0.3%) calcifying odontogenic cysts, 16 (0.2%) gingival cysts, 15 (0.2%) eruption cysts, 11 (0.2%) glandular odontogenic cysts and one Epstein pearl. The latter lesion is more commonly

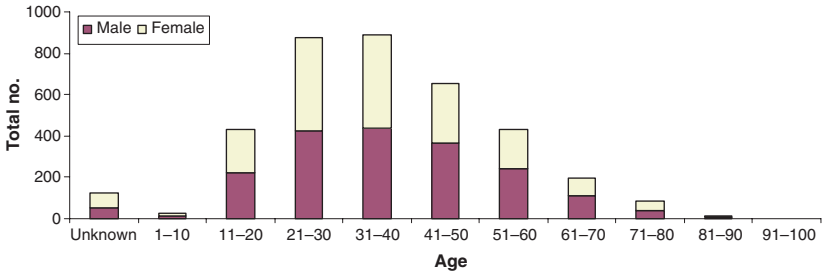

Figure 1 Age distribution of radicular cysts 1975–2004 ($n = 3724$).

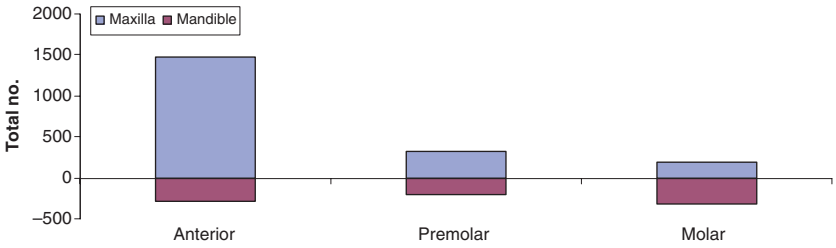

Figure 2 Site distribution of radicular cysts 1975–2004 ($n = 2801$).

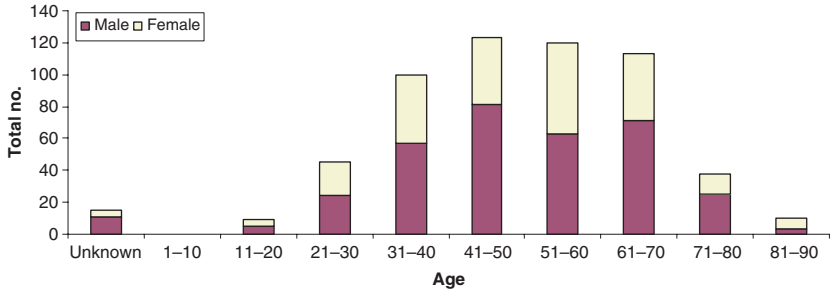

Figure 3 Age distribution of residual cysts 1975–2005 ($n = 573$).

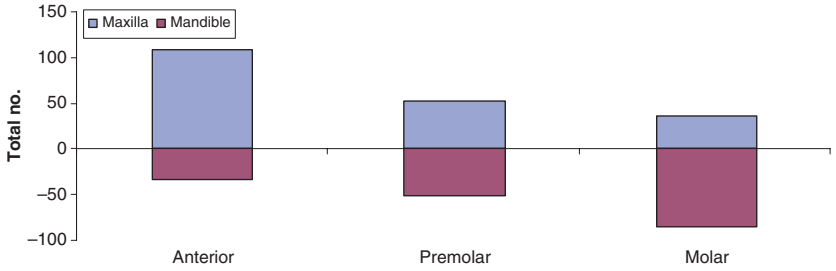

Figure 4 Site distribution of residual cysts 1975–2004 ($n = 366$).

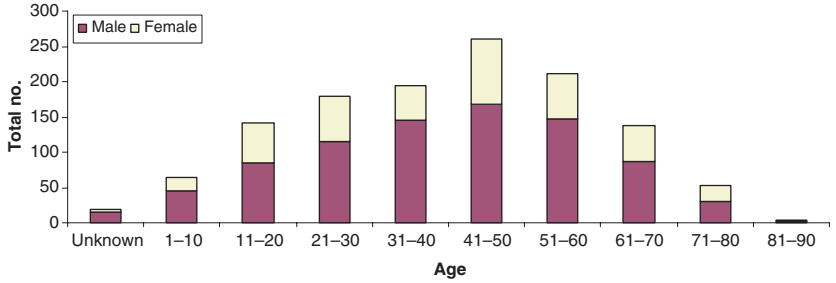

Figure 5 Age distribution of dentigerous cysts 1975–2004 ($n = 1292$).

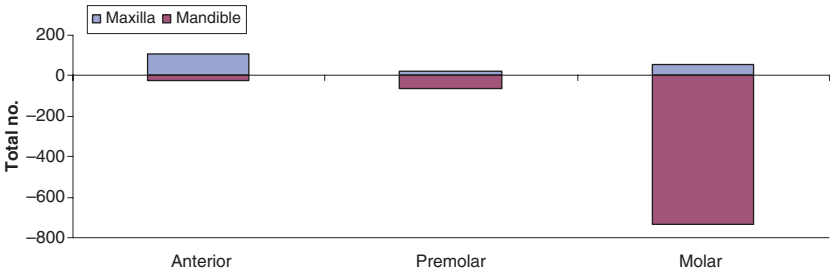

Figure 6 Site distribution of dentigerous cysts 1975–2004 ($n = 1001$).

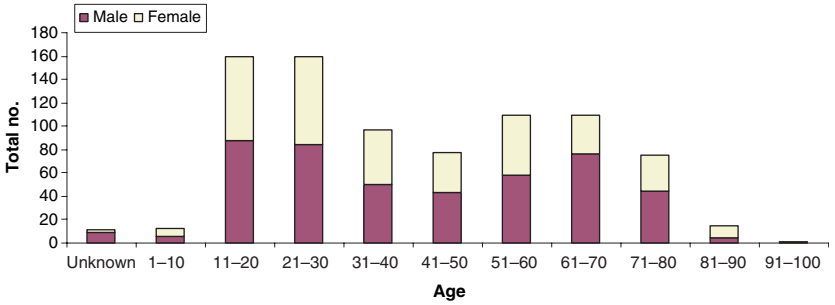

Figure 7 Age distribution of odontogenic keratocysts 1975–2004 ($n = 828$).

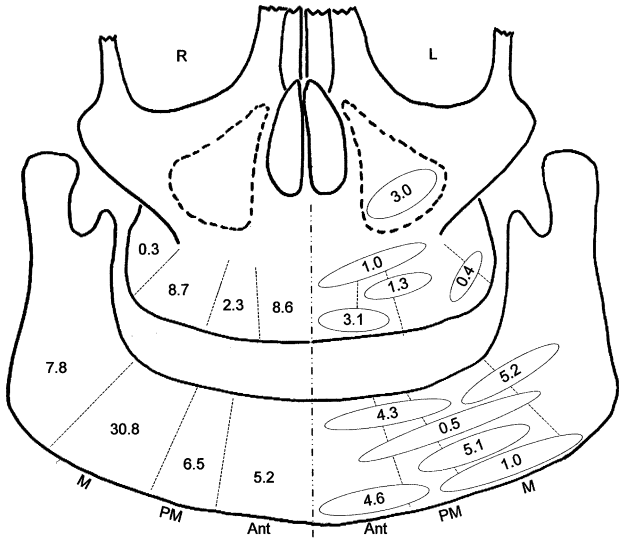

Figure 8 Percentage site distribution of odontogenic keratocysts, where site is known ($n = 766$). The right side shows cysts located at an individual site, the left side shows cysts that extend into adjacent areas. Cysts perforating the maxillary antrum are shown on the left side. M, molar region, PM, premolar region, Ant, incisor–canine region].

known as a Bohn's nodule. Of all the odontogenic cysts, only the gingival cysts of adults occurred more frequently in female than in male patients (M:F 0.33:1). However, in paediatric patients both dentigerous and calcifying odontogenic cysts were less common in males than in female patients (0.55:1 and 0.33:1 respectively).

Discussion

Although there is much information in the literature about odontogenic cysts, these data are largely based on clinicopathological and aetiological factors. In general, epidemiological and incidence data are lacking and most studies tend to be small in nature (8, 9, 11). Among cystic lesions of the jaws, there is a number that share similar clinical and radiographical features; therefore, knowledge on the incidence of odontogenic cysts as well as their commoner sites of presentation and age distribution may help practitioners to determine a likely clinical diagnosis.

Although odontogenic cysts are relatively common diagnoses within departments whose material is limited to head and neck pathology (14, 15, 18), such lesions are

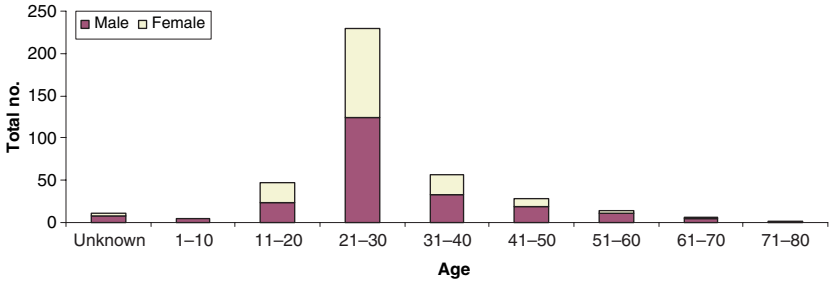

Figure 9 Age distribution of paradental cysts 1975–2004 ($n = 402$).

probably less frequently encountered in general histopathology departments. Correct diagnosis of odontogenic cysts and their variants is essential owing to the propensity of some lesions to recur and have an aggressive nature.

Large series such as that reported here can present a biased picture as the types of specimens received may be determined by the interests of the referring practitioners. Over the 30-year period in our study, an average of 12.5% of our specimens was received from general dental practitioners and specialist practitioners in primary care (the current figure is 17%) such as those practices limited to endodontics and oral surgery (14). Most of our specimens (87.5%) were received from the local dental hospital together with those from oral and maxillofacial surgery units in local hospitals. It is therefore reasonable to assume that our data more closely reflect the relative incidence of odontogenic cysts in a European population.

A total of 55 446 specimens was received over the 30-year period (1975–2004); among these, 7121 odontogenic cysts were diagnosed; this accounted for 12.8% of all submitted specimens. From the current literature, it appears that odontogenic cysts account for between 0.8% and 45.9% of all submitted specimens. This range is large and probably reflects the result of differences in referral practice. In addition, some of the studies are biased towards specific age groups such as children; this may account for the low incidence of particular cysts in some reports (17, 20, 21). Our results closely parallel those obtained from studies in Canada (10), North America (12, 16) and Mexico (8, 9) but are substantially less commonly encountered compared with those from Finland (13).

It is not surprising that radicular cyst was the most common diagnosis, accounting for 52.3% of all odontogenic cysts (6.7% of all specimens) with an approximately equal sex distribution. There was a sharp increase in their incidence until the third decade and then a gradual decline from the fourth decade onwards. These results were identical to those of Shear (22), Kreidler et al. (11) and Bhaskar (12) who found similar age distributions. A lower incidence of radicular cysts was found by Ledesma-Montes et al. (8), Mosquedo-Taylor et al. (9) and Thomson (16), while a greater proportion was reported by Daley et al. (10) and Happonen et al. (13). The site of presentation, in our study, was known for 2801 cases (75.2%) with the most common presentation in the anterior maxilla (52.8%) followed by an approximately equal distribution between the remaining sites. In addition, the anterior maxilla was by far the most common site of presentation for residual cysts, closely followed by the mandibular molar region.

Dentigerous cysts accounted for 2.3% of all diagnoses, with a male:female ratio of 1.9:1. This ratio is similar to that found by Daley et al. (10), Kreidler et al. (11) and Happonen et al. (13), but substantially lower than those reported by Ledesma-Montes et al. (8), Mosqueda-Taylor et al. (9) and Thompson (16). Dentigerous cysts are diagnosed over a wide age range, with a peak incidence in the fifth decade. The lower third molar region was by far the most common site of presentation accounting for 73.2% of all known sites of occurrence. The next most frequently affected site was the anterior maxilla. This is perhaps not a surprising finding given the fact that lower third molars and upper canines are the teeth most commonly affected by impaction. Rare cases were also found in the upper premolar and lower anterior regions. Eruption cysts were relatively rare diagnoses with only 15 cases; 62% ($n = 8/13$) of these occurred in children under 16 years of age (Table 2). The oldest patient diagnosed with this cyst was a 63-year-old female with a late erupting mandibular molar. These cysts are likely to be more common than reported, as they tend to rupture following eruption of the relevant tooth.

Odontogenic keratocysts previously included both para and orthokeratinized variants. According to the new WHO classification (23), these cysts have now been reclassified as keratocystic odontogenic tumours and jaw cysts with keratinization respectively. For the sake of comparison we have maintained the previous classification system (24). Odontogenic keratocyst was the third most common diagnosis. In previous studies, the reported incidence of odontogenic keratocysts has varied, with a range from 1.8% to 21.5% (8–12, 16, 19–22, 25). Our distribution was most similar to that reported by Kreidler et al. (11) and Brannon (25); the latter author also found a similar male:female ratio to ours at 1.3:1. Two peaks of incidence were found, one between the second and third decade and the other between the sixth and seventh decades. A similar distribution was found in a US population (26) and two UK populations (27–29). The greatest number of diagnoses was found in the second and third decades; this is consistent with findings in other studies (25, 27–30). One hundred odontogenic keratocysts (12.1%) were diagnosed in patients 16 years and younger; these accounted for 18.1% of all paediatric odontogenic cysts (Table 2). This incidence is substantially greater than those found in reports on other paediatric populations (19–21). Gorlin Gortz syndrome was suspected in 25 patients but this was confirmed clinically in only nine patients.

Paradental cyst was a term first proposed by Craig in 1976, following a study on 49 cases (31). A paradental cyst is an inflammatory odontogenic cyst usually occurring on the lateral root surface of a partly erupted tooth, and arising secondary to inflammation associated with pericoronitis leading to proliferation and cystic degeneration of follicular-reduced enamel epithelium. In the present series, 402 (5.6%) cases were diagnosed with 52.7% of cysts occurring in the third decade. This age of presentation is not a surprising finding given the frequency of association of this cyst with partly erupted lower third molar teeth with a history of pericoronitis. Philipsen et al. (3) recently published a critical review of all reported cases of paradental cysts ($n = 342$) in the English language literature. Philpsen et al. demonstrated that only 61.4% of paradental cysts occurred in the mandibular third molar region, with a male:female ratio of 2.4:1. This is in contrast to our data of 99.4% and 1.4:1 respectively for all mandibular sites. The paucity of cases involving first and second permanent molars in our series may reflect a more conservative custom and

practice on the part of UK oral surgeons given the evidence suggesting that the paradental cyst may be self-limiting in these locations (3). Certainly the associated molar teeth usually erupt successfully and are far less likely to be extracted than are third molar teeth. Considering the number of diagnoses in our study, it is perhaps surprising that so few case series have been published. It is therefore reasonable to assume that these lesions are far more common than previously suggested.

Only 28 LPCs were diagnosed with a peak age of presentation in the fifth decade. This was similar to that found by Carter et al. (7) and Kreidler et al. (11). No LPCs were diagnosed in patients less than 20 years of age, although previous studies indicate that these do occur (19, 32). Polycystic variants of a histologically similar lesion to the LPC were first reported by Weathers and Waldron (33) in 1973. This was termed the botryoid odontogenic cyst owing to its similarity, in macroscopic appearance, to a bunch of grapes. These lesions tend to have an aggressive behaviour with a propensity to recur many years after treatment (7, 34). No such lesions were diagnosed in our series, although previous studies indicate that they can account for between 8% and 16% of LPCs (7, 35).

Calcifying odontogenic cyst was a term first coined by Gorlin in 1962 and is analogous to the calcifying epithelioma of Malherbe; it can present in a solid or more common cystic variant. The solid lesions tend to behave in a more aggressive manner (5). Some authors tend to consider COCs as two entities both a cyst and a tumour (5, 36–38), while others regard them as true odontogenic tumours with a tendency towards cyst formation (39, 40). This has resulted in widespread disagreement on the classification of COCs; that currently advocated by the World Health Organisation describes these as a benign cystic neoplasm of odontogenic origin (41). For the sake of comparison we have included these lesions under the category of odontogenic cysts. The frequency of occurrence is estimated at less than 1% (11, 42) and this was true for our study (0.3%). There was a predilection for males (1.63:1) with an approximately equal distribution for each decade and an age range from 5 to 79 years. The sex distribution was similar to that found by Hong et al. in their study of 92 cases (5).

Gingival cysts of adults arise from the epithelial remnants within gingival connective tissue. These cysts commonly present in the fifth and sixth decades within the mandible (60% of our known sites of presentation) and were more frequent in females than in males (0.33:1). These findings are similar to those reported by Bell et al. (43). The gingival cyst of infants (Epstein Pearl) is rarely submitted for histopathological diagnosis as most undergo spontaneous involution within 3 months of birth (44).

Glandular odontogenic cyst (45) has acquired numerous synonyms including sialo-odontogenic cyst, a term first proposed by Padyachee and van Wyk in 1987 (46) and polymorphous odontogenic cyst (47). These lesions predominantly occur in the fifth decade, with a predilection for the anterior mandible (2, 47) and appear to affect male patients (2, 48, 49). Our results were similar. Three out of 11 sialo-odontogenic cysts recurred; one of these was associated with a squamous odontogenic tumour.

Our results demonstrate that there is a wide range of odontogenic cysts, with some cysts having a predilection for certain ages, sexes and sites. It is important to realize that some of these cysts have a marked propensity to recur as well as behave in a locally aggressive manner. Lesions such as the odontogenic keratocyst can grow to a large size, resulting in facial deformity, destruction of surrounding structures and difficult surgical management. It is essential that these lesions are detected as early as possible to minimize any necessary surgery. Following treatment, such patients need to be reviewed on a regular basis to monitor possible recurrence. Finally, it is important for those pathologists not used to dealing with oral and maxillofacial conditions to be aware of the histology of normal odontogenic tissues so as to avoid misinterpretation of dental follicles and papillae as odontogenic cysts and tumours (50).

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