International Journal of Paediatric Dentistry 2006; **16:** 19–30

An analysis of oral and maxillofacial pathology found in children over a 30-year period

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Summary. *Aim.* The vast majority of oral diseases are confined to oral tissues, but numerous underlying systemic conditions may present with signs and symptoms within the oral cavity. Since the epidemiology of diseases is variable between regions, the authors carried out Europe's first paediatric-based survey of oral and maxillofacial pathology specimens submitted for diagnosis.
Design. All entries for specimens from children between the ages of 0 and 16 years during the 30-year period from 1973 to 2002 were retrieved and compiled into 12 diagnostic categories.
Results. During the study period, 4406 (8·2%) specimens came from children between the ages of 0 and 16 years, with a male to female ratio of 1·01. The diagnostic category with the largest number of specimens was tooth pathology (22·1%), followed by salivary gland disease (19·1%) and mucosal pathology (12·1%). In all, there were 114 benign tumours of nonodontogenic origin, 43 odontogenic tumours and 31 malignant tumours. The most frequently diagnosed lesions were mucous extravasation cysts, which accounted for over 16% of cases. Periapical pathology in the form of a radicular cyst, residual cyst or chronic periapical granuloma formed almost 13% of all cases.
Conclusions. This survey shows that, while nearly 10% of specimens submitted to the authors' laboratory are from children under 16 years of age, the majority of lesions are of a benign nature, requiring minimal intervention; less than 1% of cases comprise malignant lesions. Odontogenic tumours are relatively rare in this age group; however, certain lesions such as adenomatoid odontogenic tumour and ameloblastic fibroma occur predominantly in children and, therefore, remain an important diagnostic consideration.

Introduction

The Department of Oral Pathology in the School of Clinical Dentistry, Sheffield, UK, has recorded all acceded specimens on a computerized diagnostic index database for some time. Because the date of birth and demographic information are recorded, it is possible to retrieve data on patients in a specific age range. Currently, there is very little information in the literature as to the range and prevalence of oral disease in children. Most studies of oral disease in children have been epidemiological in nature, such as the presence of caries, periodontal disease, malocclusions and trauma to teeth. Other studies in the area of oral pathology in children have concentrated on identifying odontogenic tumours or reporting a series of cases such as odontogenic tumours [1–3], jaw tumours [4–6], salivary gland tumours [7] or salivary gland lesions [8].

To date, investigations from six studies have reported the histological range of oral conditions which present in paediatric patients. These studies were based in Latin America [9], Brazil [10], Africa [11], Turkey [12] and the USA [13,14]. The studies lacked uniformity with regard to the age range, period of study, sites examined and the classification of diseases into subgroups; this makes direct comparisons difficult and open to misinterpretation. From an analysis of a number of papers [9–14], the occurrence of maxillofacial pathology in a paediatric population is estimated at approximately 10% of all submitted specimens. Since the epidemiology of diseases is variable between regions, the authors of this study have, therefore, carried out the first European survey of oral and maxillofacial pathology specimens from children aged 0–16 years submitted for diagnosis to their laboratory over a 30-year period (1973–2002).

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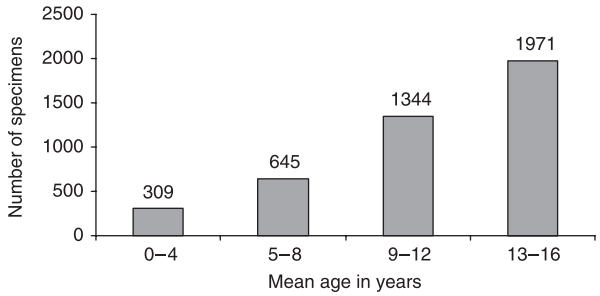

Fig. 1. Number (excluding cases where the diagnosis was normal tissue) and distribution of 4265 diagnosed specimens of oral pathology in children according to age.

Table 1. Oral pathology specimens in children from 1973 to 2003.

	Number			
Diagnostic group	Total	Male	Female	Percentage
Dental pathology	973	497	476	22·08
Salivary gland pathology*	840	393	448	19·07
Mucosal pathology*	533	263	268	12·10
Odontogenic cysts*	519	291	227	11·78
Gingival and periodontal pathology*	439	221	216	9·96
Miscellaneous pathology*	335	173	163	7·60
Odontogenic tumours*	243	117	125	5·52
Connective tissue pathology*	146	68	77	3·31
Bone pathology	143	59	84	3·25
Normal tissue	137	70	67	3·11
Non-odontogenic cysts*	67	36	30	1·52
Malignant tumours	31	13	9	0·70
Total	4406	2201	2190	100

*In some cases, the sex of the patient is unknown.

Materials and methods

Since 1989, data from all specimens received in the authors' department have been entered into a computer database, and subsequently, data from files covering the period from 1973 to 1988 were also computerized. The structure of the database has been modified several times and a Foxpro™ Windows database is now used. The initial demographic data are entered by technical staff when the specimen is received; the record is completed by secretarial staff when the final report has been issued. The diagnoses are entered using an alphanumeric code, comprising two letters, that designates the diagnostic category (e.g. OC = odontogenic cysts) and three numbers which refer to the specific condition within the diagnostic category (e.g. dentigerous cyst = OC202). There are 15 diagnostic categories which 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 the Systematized Nomenclature of Pathology (SNOP) or the Systematized Nomenclature of Medicine (SNOMED).

All entries for specimens taken from children between the ages of 0–16 years during the 30-year period from 1973 to 2002, inclusive, were retrieved, and for the sake of brevity, the diagnoses were compiled into 12 diagnostic categories (see Tables 1–12). Each diagnosis included: the number of samples, male:female ratio, age range, mean age and standard deviation, except for occasional infrequent diagnoses which have been added in footnotes to the relevant tables.

Results

During the 30-year period, 53 666 specimens were received from hospitals in Sheffield and the former Trent Region National Health Service hospitals, occasional hospitals elsewhere, and general dental practitioners. Of these specimens, 4406 (8·2%) came from children between 0 and 16 years of age. There were 2201 specimens from males and 2190 from females (male:female ratio = 1·01). In 15 cases, the gender of the patient was not provided. The distribution of diagnosed specimens according to age at presentation shows an increase in incidence with age (Fig. 1). The figures exclude those where the diagnosis was normal tissue.

The diagnostic category with the largest number of specimens was dental pathology (Table 1). Of the 973 cases in this group (22·1% of all specimens) (Table 2), 332 (34·1%) were chronic periapical granulomas. Two hundred and seventy-seven (28·5%) dental follicles were submitted, of which nearly two-thirds were normal. The latest case of a tetracycline-stained tooth was submitted in 1984.

Salivary gland disease (Table 3) was the second largest category; however, this was because 735 of the 840 cases (87·5% of this category) were mucous extravasation cysts. In contrast, mucous retention cysts comprised just over 2% of salivary cases. There were six pleomorphic adenomas and six salivary carcinomas, and the latter group were placed under malignant conditions (see Table 12 below). One case of secondary Sjögren's syndrome was diagnosed in a 16-year-old female patient who presented with

Table 2. Dental pathology.

Diagnosis	Number			Male:female ratio	Age (years)		
	Total	Males	Females		Mean	SD	Range
Chronic periapical granuloma	332	176	156	1·13	13·39	2·44	3–16
Normal dental follicle	175	69	106	0·65	12·99	2·23	5–16
Inflamed dental follicle	102	59	43	1·37	12·29	2·82	2–16
External resorption	60	25	35	0·71	11·73	3·65	1–16
Caries primary tooth	30	16	14	1·14	4·47	2·39	1–11
Enamel hypoplasia	30	16	14	1·14	6·93	3·92	2–16
Tetracycline staining	28	18	10	1·80	9·46	3·48	1–16
Supernumery tooth	21	13	8	1·63	9·95	3·59	4–16
Dentinogenesis imperfecta	20	11	9	1·22	8·80	4·19	2–16
Pulp necrosis	20	11	9	1·22	6·20	3·93	1–16
Tooth anomaly	18	7	11	0·64	8·94	4·17	1–16
Amelogenesis imperfecta	12	4	8	0·50	10·42	3·68	3–16
Regional odontodysplasia	12	6	6	1·00	9·00	4·28	3–15
Vitamin-D-resistant rickets	11	9	2	4·50	6·73	3·41	1–13
Internal resorption	10	7	3	2·33	12·20	4·33	3–16
Hypophosphatasia	9	4	5	0·80	5·67	3·37	2–12
Carious permanent tooth	8	4	4	1·00	11·86	3·76	4–15
Others*	75	42	33				
Total	973	497	476				

*Other lesions included within this group: dilaceration ($n = 7$), abnormal dentine ($n = 6$), chronic pulpitis ($n = 6$), fusion ($n = 6$), acute pulpitis ($n = 5$), attrition ($n = 5$), periapical abscess ($n = 5$), pulp abscess ($n = 5$), tooth germ ($n = 5$), germination ($n = 4$), pulp fibrosis ($n = 4$), pulp polyp ($n = 3$), taurodontism ($n = 3$), tooth fragment ($n = 3$), ankylosis ($n = 1$), concrescences ($n = 1$), dentinal dysplasia ($n = 1$), enamel hypocalcification ($n = 1$), fluorosis ($n = 1$), macrodontia ($n = 1$), microdontia ($n = 1$) and pulp stone ($n = 1$).

Table 3. Salivary gland disease.

Diagnosis	Number			Male:female ratio	Age (years)		
	Total	Males	Females		Mean	SD	Range
Mucous extravasation cyst*	735	343	388	0·88	10·57	4·06	1–16
Chronic sialoadenitis	72	34	38	0·89	11·68	3·76	1–16
Mucous retention cyst	17	10	7	1·43	11·06	3·56	1–16
Pleomorphic adenoma	6	2	4	0·50	14·50	1·87	11–16
Benign lymphoepithelial lesion*	4	1	2	0·50	9·00	2·74	6–13
Sialolithiasis	2	0	2	0·00	15·00	1·00	14–16
Focal lymphocytic sialoadenitis	1	0	1	0·00	9·00	0·00	9
Hypoplastic salivary glands	1	0	1	0·00	16·00	0·00	16
Salivary duct hyperplasia	1	0	1	0·00	6·00	0·00	6
Sjogren's syndrome	1	0	1	0·00	16·00	0·00	16
Total	840	390	445				

*In some cases, the sex of the patient is unknown.

classical symptoms of xerophthalmia, xerostomia and polyarthritis rheumatica.

Fibrous hyperplasia in the form of a nodule (fibroepithelial polyp) comprised 191 (35·8%) of the 533 cases of mucosal pathology (Table 4). Squamous papillomas were the next largest group, comprising 184 cases (34·5%). There were 21 (3·9%) cases of Crohn's disease, but only two cases of sarcoidosis. Lichen planus occurred in six cases (1·1%) with an age range of between 6 and 16 years. A single case of oral submucous fibrosis occurred in a 14-year-old Asian girl who habitually used betel quid.

Odontogenic cysts were the fourth largest group (Table 5). Two hundred and thirty-eight (45·9%) of the 519 cases were inflammatory in origin. There were 157 (30·3%) dentigerous cysts and 71 (13·7%) odontogenic keratocysts, plus two additional cases which were associated with Gorlin's syndrome (see Table 7 below). The remaining cysts accounted for just over 10% of the odontogenic cysts.

Fibrous epulides comprised 43·7% of the gingival and periodontal disease group (192 of 439 cases), and pyogenic granulomas 30·8% (135 cases). In addition, there were 45 (10·3%) peripheral giant cell

Table 4. Mucosal pathology.

Diagnosis	Number			Male:female ratio	Age (years)		
	Total	Males	Females		Mean	SD	Range
Fibrous hyperplasia*	191	91	99	0·92	11·14	3·92	1–16
Squamous papilloma	184	85	99	0·86	10·22	3·85	1–16
Nonspecific mucosal inflammation	30	20	10	2·00	11·83	4·42	1–16
Hyperkeratosis	29	13	16	0·81	11·41	4·09	2–16
Crohn's disease	21	15	6	2·50	11·90	4·00	3–16
Ephelis	6	3	3	1·00	13·67	3·20	7–16
Intradermal nevus	6	2	4	0·50	11·33	5·02	1–16
Lichen planus/lichenoid reaction	6	4	2	2·00	14·00	3·61	6–16
Orofacial granulomatosis	6	5	1	5·00	9·33	3·50	3–13
Morsicatio buccorum/linguorum	6	4	2	2·00	14·17	2·61	10–16
White sponge nevus	5	1	4	0·25	11·40	2·06	10–15
Compound nevus	3	1	2	0·50	10·67	4·19	5–15
Eosinophilic granuloma (soft tissue)	3	1	2	0·50	10·33	3·30	8–15
Epithelial hyperplasia	3	3	0	0·00	14·00	1·41	12–15
Focal melanosis	3	1	2	0·50	12·0	1·63	10–14
Giant cell fibroma	3	2	1	2·00	11·33	4·50	5–15
Antral polyp	2	0	2	0·00	13·50	0·50	13–14
Epithelial dysplasia	2	1	1	0·00	8·00	2·00	6–10
Erythema migrans	2	1	1	1·00	10·00	3·00	7–13
Erythema multiforme	2	2	0	0·00	11·00	1·00	10–12
Fibrous hyperplasia (denture)	2	2	0	0·00	14·50	0·50	14–15
Frenulum	2	1	1	1·00	13·00	0·00	13
Melanin pigmentation	2	0	2	0·00	6·00	1·00	5–7
Sarcoidosis*	2	1	0	0·00	11·50	0·50	11–12
Sebaceous cyst	2	1	1	1·00	12·00	2·00	10–14
Aphthous ulcer	1	0	1	0·00	8·00	0·00	8
Blue nevus	1	0	1	0·00	13·00	0·00	13
Calcifying epithelioma of Malherbe	1	0	1	0·00	8·00	0·00	8
Discoid lupus erythematous	1	0	1	0·00	15·00	0·00	15
Fibrous histiocytoma	1	1	0	0·00	14·00	0·00	14
Fibrous hyperplasia (tuberosity)	1	1	0	0·00	3·00	0·00	3
Hairy leukoplakia	1	1	0	0·00	4·00	0·00	4
Labial fissure	1	0	1	0·00	15·00	0·00	15
Leukodema	1	0	1	0·00	16·00	0·00	16
Oral submucous fibrosis	1	0	1	0·00	14·00	0·00	14
Total	533	263	268				

*In some cases, the sex of the patient is unknown.

Table 5. Odontogenic cysts.

Diagnosis	Number			Male:female ratio	Age (years)		
	Total	Males	Females		Mean	SD	Range
Radicular cyst*	238	127	110	1·15	13·60	2·22	4–16
Dentigerous cyst	157	97	60	1·61	10·92	3·11	3–16
Odontogenic keratocyst	71	36	35	1·03	13·42	2·50	5–16
Odontogenic cyst unclassified	18	9	9	1·00	11·06	2·04	7–15
Paradental cyst	14	8	6	1·33	11·86	3·18	6–16
Eruption cyst	11	7	4	1·75	6·82	4·15	1–12
Calcifying odontogenic cyst	5	2	3	0·67	11·60	3·56	5–15
Residual cyst	3	3	0	0·00	13·67	1·25	12–15
Epstein pearl	1	1	0	0·00	4·00	0·00	4
Keratinizing odontogenic cyst	1	1	0	0·00	7·00	0·00	7
Total	519	291	227				

*In some cases, the sex of the patient is unknown.

Table 6. Gingival and periodontal pathology.

Diagnosis	Number			Male:female ratio	Age (years)		
	Total	Males	Females		Mean	SD	Range
Fibrous epulis*	192	88	103	0·85	11·35	3·76	1–16
Pyogenic granuloma	135	72	63	1·14	11·29	3·51	1–16
Peripheral giant cell granuloma	45	28	17	1·65	10·13	2·87	3–16
Chronic gingivitis*	39	23	15	1·53	11·69	2·80	6–16
Periodontitis	7	3	4	0·75	10·86	3·91	3–15
Dilantin (Epanutin) hyperplasia	5	2	3	0·67	10·40	3·01	6–14
Fibtromatosis gingivae	5	1	4	0·25	8·00	5·22	2–16
Gingival abscess	5	1	4	0·25	8·60	3·56	4–15
Pericoronitis	5	2	3	0·67	10·80	3·82	5–15
Acute periodontitis	1	1	0	0·00	5·00	0·00	5
Total	439	221	216				

*In some cases, the sex of the patient is unknown.

Table 7. Miscellaneous pathology.

Diagnosis	Number			Male:female ratio	Age (years)		
	Total	Males	Females		Mean	SD	Range
Nondiagnostic*	105	45	59	0·76	11·51	3·86	1–16
Nonspecific ulceration	40	17	23	0·74	11·03	3·59	1–16
Scar tissue	40	27	13	2·08	10·88	4·03	2–16
Granulation tissue	36	19	17	1·12	11·58	3·39	3–16
Granulomatous inflammation*	16	9	6	1·50	9·56	5·00	1–16
Lymphoid hyperplasia (reactive)	15	12	3	4·00	11·40	4·08	5–16
Cyst fluid	12	6	6	1·00	11·67	3·79	2–16
Foreign body reaction	12	5	7	0·71	12·33	4·19	2–16
Sinus	12	5	7	0·71	10·92	3·43	5–16
Tuberculosis	4	3	1	3·00	7·00	3·94	2–13
Foreign body granuloma	3	2	1	2·00	12·00	3·74	7–16
Chronic hyperplastic candidosis	2	2	0	0·00	15·50	0·50	15–16
Condyloma acuminatum	2	1	1	1·00	9·50	2·50	7–12
Cyst – undetermined origin	2	0	2	0·00	7·00	2·00	5–9
Gorlin's syndrome	2	0	2	0·00	10·50	1·50	9–12
Molluscum contagiosum	2	0	2	0·00	10·50	5·50	5–16
Sarcoidosis*	2	1	0	0·00	11·50	0·50	11–12
Atypical mycobacterium	1	0	1	0·00	4·00	0·00	4
Behçet's syndrome	1	1	0	0·00	14·00	0·00	14
Others†	28	17	11				
Total	335	171	162				

*In some cases, the sex of the patient is unknown.
†Other lesions included within this group: foreign body ($n = 6$), amalgam tattoo ($n = 5$), abscess ($n = 3$), artefact ($n = 3$), candida smear ($n = 2$), haematoma ($n = 2$), necrotic tissue ($n = 2$), cellulitis ($n = 1$), choristoma ($n = 1$), developmental mucosal pits ($n = 1$), Ehlers–Danlos syndrome ($n = 1$) and healing tissue ($n = 1$).

granulomas. Thus, the various epulides made up 84·7% of this group (Table 6).

To facilitate organization of the data, the miscellaneous group (Table 7) comprised a broad mixture of diagnostic categories which could not be placed into any other diagnostic group. Of the 335 cases, 105 (31·3%) were nondiagnostic. The cases in this category (2·4% of all the specimens taken from the children) were usually either too small, or inadequate in some way or another for accurate reporting to be carried out.

The various types of odontomes comprised just less than 80% of cases ($n = 193$) in the odontogenic tumours and hamartoma group (Table 8). The majority of ameloblastomas (eight cases out of nine) occurred in females between 11 and 16 years of age, while ameloblastic fibroma (six cases out of eight) and all five cases of ameloblastic fibro-odontoma occurred in males between 7 and 16 years of age. Adenomatoid odontogenic tumours accounted for 10 cases (4·1%), with a male:female ratio of 0·43.

Table 8. Odontogenic tumours and hamartomas.

Diagnosis	Number			Male:female ratio	Age (years)		
	Total	Males	Females		Mean	SD	Range
Odontome:							
complex	73	43	30	1·43	11·51	2·74	4–16
compound*	60	22	37	0·59	11·33	3·29	1–16
dens in dente	25	8	17	0·47	11·56	2·62	6–16
other	22	12	10	1·20	12·40	2·17	9–16
developing	13	6	7	0·86	10·46	3·61	2–16
Adenomatoid odontogenic tumour	10	3	7	0·43	12·90	2·07	9–16
Ameloblastoma	9	1	8	0·13	13·44	1·95	11–16
Ameloblastic fibroma	8	6	2	3·00	11·75	2·90	7–16
Odontogenic hamartoma	6	3	3	1·00	10·50	2·69	8–16
Ameloblastic fibro-odontome	5	5	0	0·00	10·00	2·97	7–15
Odontogenic myxoma	5	2	3	0·67	12·00	2·19	8–14
Odontogenic fibroma	3	3	0	0·00	12·30	3·30	8–16
Cementoblastoma	2	2	0	0·00	16·00	0·00	16
Dentinoma	1	0	1	0·00	10·00	0·00	10
Odontogenic gingival epithelial hamartoma	1	1	0	0·00	10·00	0·00	10
Total	243	117	125				

*In some cases, the sex of the patient is unknown.

Table 9. Connective tissue pathology.

Diagnosis	Number			Male:female ratio	Age (years)		
	Total	Males	Females		Mean	SD	Range
Haemangioma – capillary	34	17	17	1·00	7·91	5·04	1–16
Neurofibroma	20	9	11	0·82	12·30	3·45	2–16
Hamartoma – unclassified	18	8	10	0·80	8·83	4·98	1–16
Lymphangioma	16	6	10	0·60	10·19	3·64	2–16
Haemangioma – cavernous	14	5	9	0·56	9·14	3·66	3–16
Vascular anomaly	12	8	4	2·00	11·00	4·22	1–16
Traumatic neuroma	6	3	3	1·00	12·83	3·67	5–16
Congenital epulis*	5	0	4	0·00	1·00	0·00	1
Neurilemmoma	5	4	1	4·00	11·60	2·06	8–14
Granular cell tumour	4	4	0	0·00	12·25	2·17	9–15
Others†	16	4	8				
Total	146	68	77				

*In some cases, the sex of the patient is unknown.
†Other lesions included within this group: fibromatosis ($n = 3$), lipoma ($n = 2$), angiofibroma ($n = 1$), benign teratoma ($n = 1$), desmoplastic fibroma ($n = 1$), haemangioma – juvenile ($n = 1$), nasopharyngeal angiofibroma ($n = 1$), phlebolith ($n = 1$) and xanthogranuloma ($n = 1$).

In the connective tissue disease group (Table 9), over 40% of cases were haemangiomas of one sort or another. Neuronal tumours in the form of neurofibromas (20 cases), traumatic neuroma (six cases) and neurilemmoma (five cases) accounted for over 20% of cases. Of the neurofibromas, none were known at the time to be associated with neurofibromatosis. Amongst the bone lesions (Table 10), the largest group was that of exostoses (16·8%) closely followed by central giant cell lesions (16·1%), one of which was associated with hyperparathyroidism in a 14-year-old male patient. There were 64 (44·8%) benign tumours in the bone category excluding exostoses. Fibrous dysplasia and condylar hyperplasia were the third and fourth largest categories, forming 11·2% and 9·1% of the bone lesions, respectively.

The ratio of odontogenic to nonodontogenic cysts was 7·8:1. A total of 67 nonodontogenic cysts occurred, and the largest group comprised 19 cases of solitary bone cyst (28·4%). This was closely followed by 17 nasopalatine and 15 epidermoid cysts (Table 11). There were 31 (0·7%) cases of malignancy (Table 12). Those of salivary origin (three mucoepidermoid carcinoma, two acinic cell carcinoma and one epithelial myoepithelial carcinoma) formed the largest group. Three cases of multiple mucosal neuromas

Table 10. Bone pathology: (TMJ) temporomandibular joint.

Diagnosis	Number			Male:female ratio	Age (years)		
	Total	Males	Females		Mean	SD	Range
Exostosis	24	15	9	1·67	13·25	2·33	7–16
Central giant cell granuloma	22	8	14	0·57	10·45	2·93	5–15
Fibrous dysplasia	16	7	9	0·78	10·88	3·20	4–16
Condylar hyperplasia	13	3	10	0·30	14·31	1·59	11–16
Periostitis	9	3	6	0·50	9·00	3·59	4–16
Osteoarthrosis	6	3	3	1·00	11·00	4·58	4–16
Sequestrum	6	3	3	1·00	7·50	2·93	3–13
Bone sclerosis	5	1	4	0·25	13·40	1·36	12–16
Cherubism	5	3	2	1·50	7·80	3·54	4–13
Chronic osteomyelitis	5	0	5	0·00	8·80	3·66	2–12
Osteoblastoma	5	3	2	1·50	13·20	0·40	13–14
Aneurysmal bone cyst	4	2	2	1·00	10·25	2·77	8–15
Condylar hypoplasia	4	1	3	0·33	12·50	1·50	11–15
Ankylosis (TMJ)	3	1	2	0·50	11·00	1·63	9–13
Ossifying/cementifying fibroma	3	2	1	2·00	14·00	2·16	11–16
Cleidocranial dysostosis	2	0	2	0·00	10·00	1·00	9–11
Eosinophilic granuloma (bone)	2	0	2	0·00	6·50	2·50	4–9
Osteoma	2	1	1	1·00	11·00	3·00	8–14
Acute osteomyelitis	1	0	1	0·00	12·00	0·00	12
Chondroma	1	1	0	0·00	2·00	0·00	2
Hyperparathyroidism	1	1	0	0·00	14·00	0·00	14
Infantile cortical hyperostosis	1	0	1	0·00	2·00	0·00	2
Osteitis	1	0	1	0·00	11·00	0·00	11
Osteochondroma	1	0	1	0·00	12·00	0·00	12
Sclerosing osteomyelitis	1	1	0	0·00	11·00	0·00	11
Total	143	59	84				

Table 11. Non odontogenic cysts

Diagnosis	Number			Male:female ratio	Age (years)		
	Total	Males	Females		Mean	SD	Range
Solitary bone cyst*	19	8	10	0·80	13·63	1·49	10–16
Nasopalatine cyst	17	14	3	4·67	11·29	3·92	4–15
Epidermoid cyst	15	7	8	0·88	9·73	5·73	1–16
Dermoid cyst	8	3	5	0·60	13·13	2·80	7–16
Oral lymphoepithelial cyst	3	1	2	0·50	10·00	5·72	2–15
Epithelial inclusion cyst	2	1	1	1·00	6·50	1·50	5–8
Globulomaxillary cyst	1	1	0	0·00	13·00	0·00	13
Nasolabial cyst	1	0	1	0·00	12·00	0·00	12
Thyroglossal cyst	1	1	0	0·00	13·00	0·00	13
Total	67	36	30				

*In some cases, the sex of the patient is unknown.

(MEN IIb) are listed here because they were associated with medullary carcinoma of the thyroid.

In all, there were 114 benign tumours of nonodontogenic origin, 43 odontogenic tumours and 31 malignant tumours. Therefore, neoplasms accounted for only 4·2% of all cases submitted. The most frequently diagnosed lesion was the mucous extravasation cyst (Table 13). This accounted for over 16% of cases in this age range. Periapical pathology in the form of a radicular cyst, residual cyst or chronic periapical granuloma formed almost 13% of all cases. Table 13 shows the more frequent diagnoses over the 30 years as a percentage of the total submitted.

Discussion

Over 30 years, less than 10% of all cases submitted for histopathological report were from children aged 0–16 years. Overall, the male:female ratio was 1·01, and therefore, there was no greater propensity for

Table 12. Malignant tumours.

Diagnosis	Number			Male:female ratio	Age (years)		
	Total	Males	Females		Mean	SD	Range
Histiocytosis X	3	2	1	2·00	7·67	5·25	3–15
Mucoepidermoid carcinoma	3	2	1	2·00	14·00	1·63	12–16
Multiple endocrine neoplasia syndromes	3	2	1	2·00	9·00	5·35	2–15
Neurosarcoma	3	2	1	2·00	8·67	2·36	7–12
Rhabdomyosarcoma	3	3	0	0·00	6·67	0·94	6–8
Squamous cell carcinoma	3	2	1	2·00	12·33	5·51	6–16
Acinic cell carcinoma	2	0	2	0·00	14·00	1·00	13–15
Burkitt's lymphoma	2	1	1	1·00	4·50	0·50	4–5
Retinoblastoma	2	1	1	1·00	3·00	2·00	1–5
Teratoma	2	0	2	0·00	9·50	6·50	3–16
Epithelial myoepithelial carcinoma	1	1	0	0·00	13·00	0·00	13
Ewing's tumour	1	1	0	0·00	3·00	0·00	3
Leukaemia	1	0	1	0·00	13·00	0·00	13
Osteosarcoma	1	1	0	0·00	16·00	0·00	16
Unclassified malignant tumour	1	0	1	0·00	7·00	0·00	7
Total	31	18	12				

Table 13. Frequent histological diagnoses from 1973 to 2002.

Diagnosis	Number	Percentage
Mucous extravasation cyst	735	16·68
Chronic periapical granuloma	332	7·54
Radicular cyst	238	5·40
Fibrous epulis	192	4·36
Fibrous hyperplasia	191	4·33
Normal dental follicle	175	3·97
Dentigerous cyst	157	3·56
Pyogenic granuloma	135	3·06
Nondiagnostic	105	2·38
Inflamed dental follicle	102	2·32
Squamous papilloma	94	2·13
Verruca vulgaris	90	2·04
Odontome – complex	73	1·66
Chronic sialoadenitis	72	1·63
Odontogenic keratocyst	71	1·61
Odontome – compound	60	1·36
External resorption	60	1·36
Peripheral giant cell granuloma	45	1·02
Nonspecific ulceration	40	0·91
Scar tissue	40	0·91
Total	3007	68·23

oral and maxillofacial disease in either sex. Less than 5% of cases required major surgery or complicated long-term clinical management.

The percentage of samples from paediatric patients was similar to that of previous studies; this ranged between 5·5% and 12·8% of all histopathological specimens [9–14]. The variation in number is most probably explained by differences in study design, including the period of the study (ranging from 8 to 25 years) and the age range studied. In three studies, a similar age range to the present authors' [9–11] was used; Keszler's [9] investigation was most like the present authors' owing to the use of a similar age range and period. In Keszler's study, 1289 biopsies from 0–15-year-old children were recovered from a total of 18 966 biopsies (6·8%) over a 25-year period (1960–1985). However, whereas the diagnoses are grouped into 12 categories in this analysis, Keszler *et al*. [9] used only five categories. Therefore, it is difficult to make direct comparisons in some cases. Allowing for the extra year (i.e. the present authors of this study included children up to 16 years of age) and the increased number of specimens, the proportion of specimens from children (8·2%) as a percentage of all specimens received is very similar. Also, the male:female ratio (633:623) of 1·02 is virtually identical. The age distribution was also similar in that the majority of specimens came from the 13–16-year-old group in this study, as compared with that of Keszler *et al*. [9]. Unlike Keszler's study, in which cysts (including odontogenic, nonodontogenic and soft tissue cysts) were the most frequently diagnosed lesion (25·4%), followed by tumour-like lesions (25%) (e.g. fibrous hyperplasia, peripheral and central giant cell granuloma, and fibrous dysplasia), the present authors' most frequent diagnosis was mucous extravasation cyst, followed by periapical pathology. In the report by Keszler *et al*. [9], mucoceles comprised only 6% of all lesions compared with almost 16% in this one. In both studies, radicular cyst was the most frequent odontogenic cyst and solitary bone cyst the most frequent nonodontogenic cyst. Table 14 compares the number and percentage of some of the diagnoses in this study with that of Keszler *et al*. [9].

Table 14. Comparison of data from Keszler *et al*. [9] and Jones *et al*. [19].

Diagnosis	Kezler *et al*.		Jones *et al*.	
	Number	Percentage	Number	Percentage
Radicular cyst	148	11·5	238	5·4
Dentigerous cyst	68	5·3	157	3·6
Odontogenic keratocyst	12	< 1·0	71	1·6
Solitary (traumatic) bone cyst	14	1·1	19	< 1·0
Fibrous hyperplasia (gingival fibrous epulis)	55	4·3	192	4·4
Fibrous hyperplasia (mucosal)	35	2·7	191	4·3
Mucocele/mucous extravasation cyst	76	6·0	735	16·7
Pleomorphic adenoma	1	< 1·0	6	< 1·0
Fibrous dysplasia	33	2·6	16	< 1·0
Central giant cell granuloma	21	1·6	22	< 1·0
Peripheral giant cell granuloma	63	5·0	45	1·0
Eosinophilic granuloma (bone)	6	< 1·0	2	< 1·0
Cherubism	5	< 1·0	5	< 1·0
Osteomyelitis	28	2·2	17	< 1·0
Odontomata	34	2·6	193	4·4
Ameloblastoma	8	< 1·0	9	< 1·0
Odontogenic myxoma	8	< 1·0	5	< 1·0
Adenomatoid odontogenic tumour	5	< 1·0	10	< 1·0
Ossifying/cementifying fibroma	2	< 1·0	3	< 1·0
Odontogenic fibroma	2	< 1·0	3	< 1·0
Calcifying odontogenic cyst	2	< 1·0	5	< 1·0
Malignant neoplasms	21	1·8	31	< 1·0

Dental pathology

It is not surprising that dental pathology was the largest diagnostic category (22%). Epidemiologically, this diagnosis is likely to be grossly underestimated because of the extent of caries [15]; carious teeth are not usually sent for histological diagnosis. With clinical diagnosis of dentinogenesis and amelogenesis imperfecta masking the true extent of these conditions, it is likely that the numbers (20 cases and 12 cases seen over a 30-year period, respectively) have again be underestimated. Out of the previously reported data, only two studies included a subclassification for dental pathology. Since Maia's [10] study lacked information on the total number of specimens per group, direct comparisons with the data in this work cannot be drawn. From data collected by Keszler's group [9], it appears that dental pathology accounted for 6·5% of specimens; this is substantially lower than the authors found in this study.

Salivary pathology

Mucous extravasation cysts comprised the largest single diagnostic group, accounting for over 16% of specimens. In the majority of other studies, mucous extravasation and retention cysts were not subclassified, and collectively, these comprised between 5·0% and 21·8% of cases. A total of six pleomorphic adenomas were found in the 11–16-year-old age range. Although salivary tumours are generally rare within paediatric populations, the ratio of benign to malignant salivary tumours was 1:1. Therefore, any persistent swelling of unknown aetiology within the salivary glands requires urgent investigation.

Cystic lesions

The ratio of odontogenic to nonodontogenic cysts was 7·8:1, and of these, radicular cyst was the most common diagnosis, accounting for 45·9% of odontogenic cysts and 40·6% of all cysts. It has been reported in previous studies that dentigerous cysts are more common than radicular cysts in paediatric populations [10,12]. One study of a group of African children showed that radicular cysts were more common [11]. The reason for this difference may relate to the prevalence of caries and oral health regimes between different countries. Over 14% of odontogenic cysts were odontogenic keratocysts, with a male:female ratio of 0·97, including two cases associated with Gorlin Gortz syndrome. Shear [16] showed a similar occurrence pattern of odontogenic keratocyst in a study of 2616 cysts from all ages, while an occurrence rate between 4 and 9% has been

reported in other paediatric studies. From large-scale studies of all ages, it appears that the occurrence of odontogenic keratocyst is more frequent in males than females [17,18].

Odontogenic tumours

Within the odontogenic tumours and hamartomas group, the mean age at presentation for ameloblastic fibro-odontome and dentinoma (ameloblastic fibro-dentinoma) was similar to that described by Hansen and Ficarra [2] in San Francisco, CA, USA; however, for ameloblastic fibroma, the mean age in the present authors' data was slightly older at 12 years (SD = 2·9 years, range = 7–16 years), compared with 9 years (range = 3–19 years). The oldest patient with this diagnosis for all the authors' records (adults included) was 15 years of age. In this study, eight of nine ameloblastomas were found in females aged 11–16 years. This age range is similar to that described by Taiwo *et al*. [6], who reported that nine of their 10 cases were in that age group; however, in their study, the majority of cases were male. The present authors' is the only work that demonstrates such a large difference in male:female ratios in paediatric populations. The 10 cases of adenomatoid odontogenic tumour (age range = 9–16 years) were also more common in females, with a male:female ratio of 0·43. In this study, there were 43 odontogenic tumours, representing less than 1% of cases. This confirms the views of some researchers [5,6] that odontogenic tumours are rare in children.

Keszler *et al*., in a separate study from Buenos Aires [3], presented a series of 10 cases of odontogenic myxoma in children aged 16 years or under (six males and four females). These cases comprised 12·5% of all cases with this diagnosis in their records. The age range in these children was 5–16 years, with a mean age of 11·6 years. These data reflects the present authors' observations (Table 8). The five cases in this survey represent 14% of the 35 cases in the authors' records; this compares with a figure of 8·5% for the literature, as derived by Keszler *et al*. [9]. In this study, the authors recovered just three cases of ossifying fibroma (ages of subjects = 11, 14 and 15 years); in comparison, Taiwo and co-workers [6] found 20 such cases over a shorter time period. The majority of their cases ($n = 14$) were in the same age range as the present group (11–16 years) and the difference in numbers may be because of the increased incidence of this disease in Africa.

Malignant tumours

In the study by Keszler *et al*. [9], the number of malignant neoplasms ($n = 21$) was small and was similar to the present authors' study ($n = 31$). Taiwo *et al*. [6] reported 37 lymphomas, of which 33 cases were Burkitt's lymphoma, a not unexpected finding in Nigeria. The other malignant neoplasms in their study amounted to 16. It has been reported in two studies [4,6] that, when squamous cell carcinoma occurs in children, it is usually poorly differentiated; this was also true for the cases in this study. Both these authors found that mesenchymal malignant tumours occurred more frequently in their studies and this is also reflected in the present data. Although Taiwo *et al*. [6] reported that the majority of their malignant tumours occurred in the 12–16-year-old age group, only 14 of the 31 cases in this study were in the same age range. Like Taiwo *et al*. [6], all three cases of rhabdomyosarcoma in this study occurred in the first decade. Salivary tumours accounted for nearly 20% ($n = 6$) of all malignancies, with an equal number of benign salivary tumours ($n = 6$). In two previous studies [7,8], mucoepidermoid carcinoma was the most common malignant salivary gland neoplasm in children, and this was consistent with the present results.

The authors are conscious that there is no data on ethnicity within the demographic information on their database. They realize that such data might

What this paper adds

- This paper provides a comprehensive analysis of the range of histopathological diagnoses in a European paediatric population.
- Paediatric biopsy specimens account for approximately 8% of all those submitted for histological diagnosis.
- The majority of lesions are of a benign nature requiring minimal intervention; however, it is important to recognise that malignant lesions can occur in children.

Why this paper is important for paediatric dentists

- There is a wide range of conditions that may present within the oral cavity; knowledge of their prevalence allows a clinician to have a better understanding of likely diagnoses.
- The largest number of specimens was derived from tooth pathology followed by salivary gland disease and mucosal pathology. There were 157 benign tumours and 31 malignant tumours. The most frequently diagnosed lesions were mucous extravasation cysts.
- Any persistent swelling of unknown aetiology within the salivary glands requires an urgent investigation to eliminate the possibility of benign or malignant neoplasm.

provide a valuable insight into relative frequency of diseases or conditions which may present in different ethnic communities, or be useful when comparing data from different geographical areas or socioeconomic regions in different parts of the UK or other countries. Unfortunately, our pathology request forms do not yet seek information on ethnicity. Clearly, it would be useful if such data were requested in an internationally agreed format that was comparable with that collected in all studies such as this one. Whatever system may be in place, it is in the hands of clinicians submitting specimens as to whether such data is actually provided.

Conclusion

This survey covers a wide spectrum of disease that should be of interest to the general dental practitioner and paediatric dentists. The results do not represent the actual prevalence of oral disease in the general population, but simply the relative frequency of histologically diagnosed lesions over a 30-year period. Diagnosis is primarily based on clinical information and the supply of adequate histological material. Frequently, the information provided is sparse and occasionally neglected, and the pathological specimen insufficient for accurate diagnosis. Approximately 2·4% of all specimens ($n = 105$) were categorized as nondiagnostic, compared with 10·1% in a study by Keszler *et al*. [9].

The vast majority of oral diseases are confined to oral tissues, but numerous underlying systemic conditions may present with signs and symptoms within the oral cavity. This survey has shown that, while nearly 10% of specimens submitted to the authors' laboratory are from children under 16 years, the majority of lesions are of a benign nature requiring minimal intervention. There were 31 malignant lesions, of which six were salivary gland in origin. These malignant salivary gland tumours accounted for 50% of all salivary tumours. Although malignant lesions comprised less than 1% of cases, it is nevertheless important to recognize that such lesions can occur in children. Odontogenic tumours are relatively rare in this age group; however, certain lesions such as adenomatoid odontogenic tumour and ameloblastic fibroma occur predominantly in children, and therefore, remain an important diagnostic consideration. Overall, mucous extravasation cysts and periapical pathology are the most commonly diagnosed lesions.

Acknowledgements

The senior author (C.D.F.), who devised the database, wishes to acknowledge the diagnostic input of his two colleagues, Professor C. J. Smith (now retired) and Dr Geoffrey Craig, without whom the database would not be available to use. We also wish to thanks the various technicians and secretaries who, over the years, have also input data and carried out the day-to-day work of the surgical diagnostic service.

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