Scientific Article

A retrospective study of oral and maxillofacial biopsy lesions in a pediatric population from southern Taiwan

Yuk-Kwan Chen, DDS, MS Li-Min Lin, DDS, MS, PhD Hsien-Cheng Huang, DDS, MS Cheng-Chung Lin, DDS Yat-Han Yan, DDS, MS

Abstract

This is a retrospective study of biopsied oral lesions (N = 534) in a pediatric population (0–15 years old) in southern Taiwan. In this study, we included 6% of the 9672 biopsies performed from 1985 through 1996. The lesions were divided into three groups according to patients' ages, 0–5 years old; 6–10 years old; and 11–15 years old The majority of the samples were from the oldest age group (273/518). With the exclusion of normal tissues (N = 26), 518 samples were classified into four categories: inflammatory lesions, cystic lesions, tumor or tumor-like lesions, and other lesions. The largest number of lesions occurred in the inflammatory lesion group (46%). The 12 most frequently occurring lesions contributed about 78% of all the biopsies in the three age groups (0–15 years old). Frequencies of the lesions of mucous extravasation phenomenon, dentigerous cyst, fibrous dysplasia, and odontonia in our three pediatric age groups showed a significant proportion in the biopsies of the same lesions in the group of patients of all ages. These information may be valuable for both epidemiology and teaching. (Pediatr Dent 20:404–410, 1998)

The studies of biopsied oral and maxillofacial lesions in the pediatric population are relatively uncommon.[1–6] These studies were largely done on tumor or tumor-like lesions, but the status of other kinds of lesions were not mentioned. A few comprehensive surveys on a broad spectrum of oral biopsy lesions in pediatric age groups have been reported from North[7,8] and South America.[9] Regional and racial differences in oral lesions were observed between these surveys.[7–9] However, limited information of this nature is available in Asia, thus it is useful to analyze data on this subject. The age groups studied in previous reports were from 0 to 20 years, but the range of 0 to 15 years was most commonly used. Therefore, the age group we studied was 0–15 years. The objective is to review the oral biopsy specimens within a population of pediatric patients from southern Taiwan relative to age, sex, frequency, and distribution. Such information may be valuable for both epidemiology and teaching.

Methods and materials

Biopsy records of the 9672 microscopically diagnosed cases in the oral and maxillofacial region (1985–1996) were retrieved from the Oral Pathology Department of Kaohsiung Medical College Teaching Hospital. All tissues including teeth obtained from a patient were considered as one biopsy. A pool of 534 samples from pediatric patients (0–15 year old) were selected for detailed analysis. Age, sex, location of lesions, and histologic diagnosis were compiled for each case. The method of data collection and analysis used was described by Skinner et al.[10] with minor modifications. The collected data were divided into three groups according to age: 0–5, 6–10, and 11–15 years old. The lesions were classified into four categories: inflammatory lesions, cystic lesions, tumor or tumor-like lesions, and other. Specimens with no particular microscopic features were regarded as normal tissues and were excluded from further analysis.

Results

A total of 534 pediatric oral biopsies (6%) were diagnosed from the 9672 cases performed from 1985 through 1996. The age of the pediatric patients ranged from 3 months to 15 years old.

The 12 most frequently occurring lesions in the pediatric patients (0–15 years old) contributed about 78% of all the biopsies in the three groups (Table 1). It is worth noting that the percentages of lesions of mucous extravasation phenomenon (mucocele, ranula, mucous retention cyst, 29%), dentigerous cyst (28%), and fibrous dysplasia (26%) in the three pediatric age groups constituted a significant proportion of the same lesions in the group of patients of all ages. In addition, odontoma (49%) in the three pediatric age groups comprised nearly 50% of those in the group of patients of all ages.

With the exclusion of the 26 normal tissues, the remaining 518 samples consisting of 60 different diagnoses were categorized into four groups (Table 2). The largest number of lesions occurred in the inflam-

TABLE 1. NUMBER AND PERCENTAGES OF THE 12 MOST COMMON LESIONS IN PEDIATRIC PATIENTS

	Total	*% of total in pediatric-age group*	*% of the same lesions in all-age group*
Mucous extravastation phenomenon*	144	27.8%	28.6%
Dentigerous cyst	50	9.65%	28.4%
Odontoma	40	7.72%	49.3%
Inflammation	29	5.60%	5.62%
Radicular cyst	24	4.63%	3.33%
Hemangioma	23	4.44%	15.9%
Ameloblastoma	21	4.05%	17.5%
Periapical granuloma	19	3.67%	3.64%
Fibroma	16	3.09%	4.64%
Pyogenic granuloma	15	2.90%	12.8%
Ulceration	12	2.32%	6.0%
Fibrous dysplasia	10	1.93%	26.3%
Total	403	77.8%	

*Mucocele, ranula, mucous retention cyst.

TABLE 2. NUMBER AND PERCENTAGE OF THE FOUR CATEGORIES OF LESIONS IN PEDIATRIC PATIENTS

	Total	*% of total no. of biopsy in pediatric population*
Tumor/tumor like lesions		
Odontogenic (benign)	74 (71/3)†	14.3%
Non-odontogenic (benign)	81 (18/63)†	15.6%
Non-odontogenic (malignant)	11 (9/2)†	2.1%
Cystic lesions	91	17.6%
Inflammatory lesions	238	45.9%
Other lesions	23	4.5%
Total	518*	100%

*Excluding the 26 normal tissues. †Values in parentheses indicated jawbone tumors/soft tissue tumors.

matory lesion group (46%). The majority lesions in the tumor/tumor-like category were benign cases (93%).

The purpose of dividing the lesions into three age groups was to note the number of patients and the frequency of the lesions relative to age (Table 3). The majority of the samples were in the oldest age group (273/518). Mucous extravasation phenomenon was the most common lesion in all of the three age groups. A total of 21 distinct types of lesions were identified in the 0–5-year-old group, in which mucous extravasation phenomenon was the most common lesion which was much more than the second-most common lesion type, which comprised of only five cases of inflammation. Thirty-one distinct types of lesions were noted in the 6–10-year-old group, and 42 in the 11–15-year-old group.

Most benign odontogenic tumors (Table 4) were jawbone lesions (N = 71); only three were soft-tissue lesions (three peripheral odontogenic tumors in gingiva). Two lesions predominated in the benign odontogenic tumor category, odontoma (N = 40), and ameloblastoma (N = 21). A slight maxilla predominance (52%) of the odontoma was noted. The dominant site of ameloblastoma was posterior mandibular body (N = 16) with four lesions located in the ramus. A single maxilla lesion was found in the anterior region.

In contrast to benign odontogenic tumors, most benign nonodontogenic lesions in this study (Table 5) were soft-tissue neoplasm (N = 63). Eighteen jawbone lesions were included. The most prevalent lesion was hemangioma (N = 23), which comprised of capillary (N = 8), cavernous (N = 11), and sclerosing (N = 4) types. Most fibromas occurred in the buccal mucosa (N = 14). Six fibrous dysplasias were located in the maxilla while four were in the mandible. Pleomorphic adenoma was the only benign salivary gland neoplasm which occurred in this series; six cases occurred in the palate

TABLE 3. AGE AND SEX DISTRIBUTION OF ALL THE PEDIATRIC PATIENTS

Age	*Biopsy (M/F)**	*Total*
0-5 yrs	60/25	85 (2.4:1)†
6-10 yrs	86/74	160 (1.2:1)
11-15 yrs	142/131	273 (1.1:1)
Total	288/230	518(1.3:1)

*M/F: male/female. †Value in brackets : male to female ratio.

TABLE 4. NUMBER AND SEX OF PEDIATRIC PATIENTS WITH ODONTOGENIC TUMORS (BENIGN)

	0-5 yrs	*6-10 yrs*	*11-15 yrs*	*Total (M/F)**
Bone tumors				
Odontoma	2	9	29	40(26/14)
Adenomatoid odontogenic tumor	0	1	0	1(0/1)
Ameloblastoma	0	0	21	21(10/11)
Ameloblastic odontoma	0	0	2	22/0)
Cementifying fibroma	0	0	4	5(5/0
Odontogenic myxoma	0	0	2	2(0/2)
Soft tissue tumor				
Peripheral odontogenic fibroma	0	0	3	3(1/2)
Total	2	11	6	74(42/32)

*M/F: male/female.

and one in the submandibular gland. Few other non-odontogenic tumors were found in this category. The only juvenile xanthanogranuloma,[11] and one of the three aneurysmal bone cysts have already been reported.[12]

Only five distinct types of malignant lesions were found in this study (Table 6). The predominant lesion type was Langerhans cell histiocytosis, and all lesions were located in the mandible.

Most cystic lesions (Table 7) were odontogenic (N = 89); there were only two nonodontogenic cystic lesions (epidermoid cyst and nasopalatine duct cyst). The most common site of dentigerous cyst was the mandible (N = 29). Radicular cysts were nearly equally disributed between the maxilla (N = 11) and the mandible (N = 10). All of the eight keratocysts were situated in the mandible (one in the ramus, one in the symphysis, six in the posterior mandibular body).

Mucous extravasation phenomenon was the most common lesion with a predilection for the lower lip (N = 106) and female patients (N = 75). (Table 8). The frequency of other anomalies was low (Table 9).

Discussion

The Oral Pathology Department of our hospital not only provides services for almost all of the biopsied oral and maxillofacial lesions (Medical Pathology contributed only an insignificant number of cases) but also the most important referral center for these lesions in southern Taiwan. Therefore, the various types of lesions in this report are representative of the occurrence of these lesions in that region.

It is worthwhile to note that the numbers of mucous extravasation phenomenon, dentigerous cyst, fibrous dysplasia, and especially odontoma, were found in significant frequency in the biopsies of the same lesions obtained from a survey of patients of all ages. This implies that the above-mentioned four pediatric lesions constitute an essential frequency in the biopsies submitted in the same period (1985–1996).

The percentage (6%) of the pediatric oral biopsy specimens in the present study is lower than in the reports of Skinner et al. (13%)[7] and Sumitra and Das (12%).[8] This may be because only patients up to 15 years old were included in the present series, while they included patients up to 19 and 20 years old, respectively. On the other hand, it is compatible to the percentage (7%) of patients in the report by Keszler et al.[9] who comprised samples from birth to 15 year old.

The majority of the lesions were in the inflammatory lesion group, which concurs with two previous reports[7, 8] of the same nature. However, it is different from the report of Keszler et al.,[9] who found cystic lesions to be the most prevalent sample group.

The percentage (93%) of occurrence of benign cases in the tumor or tumor-like lesions category in our study were much higher than the results of Godwin (60%) and Keszler et al.[9] (84%). On the other hand, our results are similar to a report from Japan (93%).[6] The view that pediatric jawbone tumors are usually non-odontogenic[2, 4, 9, 13] does not apply in the present study because 74% of the jawbone tumors in this report were odontogenic. This is similar to the findings of Sato et al.,[6] who also noted an odontogenic predominance in the jawbone tumors (88%).

Odontoma and ameloblastoma comprised most of the samples in the category of benign odontogenic tumors; other lesions were infrequent. A similar pattern of occurrence was also observed in a study by Sato et al.[6]

TABLE 5. NUMBER AND SEX OF PEDIATRIC PATIENTS WITH NON-ODONTOGENIC TUMORS (BENIGN) OR TUMOR LIKE LESIONS

	0-5 yrs	*6-10 yrs*	*11-15 yrs*	*Total (M/F)**
Bone Tumors				
Osteoma	0	1	3	4 (2/2)
Osteoblastoma	0	0	1	1 (0/1)
Fibrous Dysplasia	0	2	8	10 (8/2)
Aneurysmal bone cyst	1	0	2	3 (3/0)
Soft tissue tumors				
Papilloma	0	2	2	4 (3/1)
Fibroma	4	3	9	16 (12/4)
Hemangioma	4	4	15	23 (9/14)
Lymphangioma	1	1	0	2 (0/2)
Cystic hygroma	1	0	0	1 (1/0)
Schwannoma/neurilemmoma	0	0	2	2 (1/1)
Juvenile xanthogranuloma	0	0	1	1 (1/0)
Congenital epulis	2	0	0	2 (0/2)
Pilomaxtrioma	0	1	0	1 (1/0)
Myxofibroma	0	0	1	1 (1/0)
Nevus	0	0	3	3 (3/0)
Pleomorphic adenoma	0	1	6	7 (1/6)
Total	13	15	53	81(46/35)

*M/F: male/female.

TABLE 6. NUMBER AND SEX OF PEDIATRIC PATIENTS WITH NON-ODONTOGENIC TUMORS (MALIGNANT)

	0-5 yrs	*6-10 yrs*	*11-15 yrs*	*Total (M/F)**
Bone tumors				
Mucoepidermoid carcinoma	0	0	1	1 (1/0)
Langerhans cell histocytosis	2	3	2	7 (6/1)
Rhabdomyosarcoma	0	0	1	1 (1/0)
Soft tissue tumors				
Lymphoma	1	0	0	1 (1/0)
Leukemia	1	0	0	1 (1/0)
Total	4	3	4	11 (10/1)

*M/F: male/female.

The most common odontogenic tumor was odontoma, which is consistent with previous reports.[7, 8, 14–16] A number of studies[7, 8, 14, 16, 17] indicated the second decade of life was the most common period of occurrence. Our finding is similar to these previous studies.[7, 8, 14–17] However, the present series showed a much higher rate (8%) of odontoma than two studies from the US.[7, 8] On the other hand, the number of odontoma of the present series was compatible to a study from Asia.[6] Two reports[7, 15] found an equal distribution among males and females while Budnick[16] reported a slight male predominance. The present study demonstrated a definite male dominance. Our finding revealed a 52% rate of occurrence in maxilla, which correlates with the 51% reported by Owens et al.,[17] 55% by Skinner et al.,[7] 55% by Sato et al.[6], and 65% by Budnick.[16]

No single case[1,7,18] or only small number of ameloblastomas were found in previous studies.[3, 8] In the present series, the percentage of ameloblastoma (13%) was much higher than other related studies.[19, 20] On the other hand, the frequency of ameloblastoma in the present study was similar to that of Sato et al.[6] All of the ameloblastoma in this report were in the oldest age group, as found in other studies.[8, 19, 20] In our study, a slight female predominance was found in patients with ameloblastoma, which correlates with the report of Kahn.[20] However, this is in contrast to a study on Nigerian children by Godwin,[5] who reported a male predominance and is also in contrast to the study of Keszler and Dominguez,[19] who found no differences in the occurrence of ameloblastoma between the sexes in pediatric patients. The common site of occurrence was the mandible in the present study, which concurs with other studies.[6, 19, 20]

A higher relative frequency of adenomatoid odontogenic tumors were supposedly seen among black Africans.[21] In the report of Godwin et al.,[22] 28 out of 57 patients with adenomatoid odontogenic tumors

TABLE 7. NUMBER AND SEX OF PEDIATRIC PATIENTS WITH CYSTIC LESIONS

	0-5 yrs	*6-10 yrs*	*11-15 yrs*	*Total (M/F)**
Radicular cyst	1	4	19	24 (14/10)
Dentigerous cyst	1	18	31	50 (34/16)
Eruption cyst†	0	1	0	1 (1/0)
Calcifying odontogenic cyst	0	0	2	2 (0/2)
Residual cyst	0	0	3	3 (0/3)
Periodontal cyst	0	0	1	1 (1/0)
Keratocyst	0	0	8	8 (6/2)
Nasopalatine duct cyst	0	1	0	1 (0/1)
Epidermoid cyst†	1	0	0	1 (0/1)
Total	3	24	64	91 (56/35)

*M/F: male/female. †Soft tissue cysts.

TABLE 8. NUMBER AND SEX OF PEDIATRIC PATIENTS WITH INFLAMMATORY LESIONS

	0-5 yrs	*6-10 yrs*	*11-15 yrs*	*Total (M/F)**
Inflammation	5	11	13	29 (15/14)
Tuberculosis	0	3	0	3 (1/2)
Actinomycosis	1	0	0	1 (0/1)
Condensing osteitis, osteosclerosis	0	1	0	1 (1/0)
Osteomyelitis	1	2	4	7 (5/2)
Pulpitis	1	0	1	2 (2/0)
Polyp	0	1	0	1 (0/1)
Periapical granuloma	0	2	17	19 (9/10)
Mucositis	0	1	0	1 (0/1)
Garre's osteomyelitis	0	2	0	2 (1/1)
Pyogenic granuloma	2	6	7	15 (6/9)
Fibrous hyperplasia	0	1	6	7 (7/0)
Gingivitis	0	2	1	3 (1/2)
Mucous extravasation phenomeon†	21	66	57	144 (69/75)
Sialadentitis	0	0	2	2 (2/0)
Gingival hyperplasia	0	0	1	1 (1/0)
Total	31	98	109	238 (120/118)

*M/F: male/female. †Mucocele, ranula, mucous retention cyst.

were younger than 15 years old. However, only one case of adenomatoid odontogenic tumor was noted in the present study, as in a report from Japan.[6] This may be due to the racial differences.

The most frequent benign nonodontogenic tumor in this study was hemangioma. It is different from the reports from the U.S.,[7,8] which indicated papilloma as the most prevalent lesion. However, the present series is similar to a study from Japan,[6] which also reported hemangioma as the most common benign tumor. The low percentage of papilloma in the present study may be due to geographic variations. The percentage of hemangioma (4%) in this study was much greater than the 1% rate of Sumitra et al.,[8] and also higher than the report by Godwin,[5] which included only three hemangiomas. Both the occurrence of fibroma (10%) and fibrous dysplasia (6%) are higher than in our study than in the report by Godwin (6 and 4% respectiveiy).[5] However, the frequency of pleomorphic adenoma (4%) is compatible with that of Godwin (5%).[5] Palatal pleomorphic adenoma in children is relatively uncommon; however, a few cases were recently presented.[23,24] In the present study, there were six palatal pleomorphic adenomas which were all in the 11–15-year-old group, as in the report by Godwin.[5]

Langerhans cell histiocytosis was the most common malignant nonodontogenic lesion, which is consistent with the findings of Kaban.[25] This is in contrast to a report from Africa, in which the most common malignant nonodontogenic neoplasm was Burkitt's lymphoma.[5] In addition, a report from Asia[6] noted the dominant malignant nonodontogenic lesions was sarcoma. These differences may be attributed to regional or racial variations. No squamous cell carcinoma was found in this series but there was a case of mucoepidermoid carcinoma.

Ninety-eight percent of the cystic lesions were located in the jaw; which is much higher than reported by Keszler et al.[9] (75%). The occurrence of dentigerous cyst was higher (10%) than that reported by Skinner et al.[7] (9%) and Sumitra et al.[8] (5%). The most

TABLE 9. NUMBER AND SEX OF PEDIATRIC PATIENTS WITH OTHER LESIONS

	0-5 yrs	*6-10 yrs*	*11-15 yrs*	*Total (M/F)**
A-V fistula	0	0	1	1 (0/1)
Ulceration	1	7	4	12 (10/2)
Calcification/sclerosis	0	0	1	1(1/0)
Internal/external tooth resorption	0	1	0	1(1/0)
Simple bone cyst	0	0	3	3(0/3)
Tooth abrasion	0	0	3	3(0/3)
Hematoma	1	1	0	2(2/0)
Total	2	9	12	23(14/9)

*M/F: male/female.

frequent site was the mandibular posterior region, which correlated with the findings of both Skinner et al.[7] and Sumitra et al.[8]

The occurrence of radicular cysts (4%) in this study is compatible to that of Skinner et al.,[7] (5%) but is much lower than those of Sumitra et al.[8] (8%) and Bhaskar[1] (11%). Radicular cysts occurred more often in the maxilla than in the mandible in some studies[1,7]; however an equal distribution between maxilla and mandible was noted in this study. The present study showed a much higher occurrence of odontogenic keratocyst than the report by Sumitra et al.,[8] who found only one such lesion. Our findings were higher than the 6% of Skinner et al.[8] and 5% of Keszler et al.[9]

We found that mucous extravasation phenomenon was the most common lesion in the inflammatory lesions category, as did Skinner et al.[7] and Sumitra et al.[8] Most studies[7–9] reported that the lower lip was the most predominant site; our study concurs with this finding. The majority of the cases from other studies occurred in females,[7–9] which was similar to our study. The percentage of chronic or acute inflammation (6%) in our study was lower than in the reports by Skinner et al.[7] (9%) and Sumitra et al.[8] (7%). The majority of granuloma in the current study were found in the anterior maxilla, which is consistent with two previous studies.[7,8] We found the most common site of this lesion was the gingiva, which concurs with Skinner et al.[7] and Sumitra et al.[8]

In conclusion, little data of this nature has been reported from Asia. Several salient points regarding regional and racial differences are highlighted by the data in this study. The differences include the majority of lesions were occurred in the inflammatory lesions category. The dominant jawbone pediatric tumor is odontogenic, with a high rate of odontomas and ameloblastomas but a low rate of adenomatoid odontogenic tumors. The most frequent benign non-odontogenic tumor was hemangioma and the most common malignant counterpart was Langerhans cell histiocytosis. In addition, a high occurrence of odontogenic keratocyst was found. Thus, this information may be valuable for both epidemiology and teaching.

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