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ORIGINAL ARTICLE

Retrospective study of biopsied oral and maxillofacial lesions in pediatric patients from Southern Taiwan

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Abstract *Background/purpose:* This study aimed to provide updated information on biopsied oral and maxillofacial (OMF) pediatric lesions in Southern Taiwan. The data were compared with our previous study (1985–1996) and also with previous studies from Northern Taiwan and other countries.
Materials and methods: Biopsied cases from a pediatric population (0–15 years old), between 1997 and 2011, were retrieved from the Oral Pathology Department of our institution. Age, sex, location, and histological diagnoses were recorded. The data were divided into three age levels: 0–5 years, 6–10 years, and 11–15 years. The OMF lesions were classified into four categories: tumor/tumor-like lesions, cystic/pseudocystic lesions, inflammation/reactive lesions and other miscellaneous lesions.
Results: Of a total of 36,264 biopsied cases, 1023 OMF biopsies were recorded for patients between 0 and 15 years. Most lesions were in the inflammation/reactive group ($n = 451$), followed by the groups of tumor/tumor-like lesions ($n = 240$), cystic/pseudocystic lesions ($n = 196$) and other miscellaneous lesions ($n = 136$). Most cases were located in the

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11—15 year age group. Mucocele was the most common lesion, followed by odontoma and dentigerous cyst, which comprised 41.5% of total numbers of pediatric biopsied lesions.
Conclusion: There was an increase of 52% of biopsied pediatric OMF lesions in the current series (1997—2011) as compared to our previous study (1985—1996). The present study showed a similar trend to our previous study, the study from Northern Taiwan and also other studies. However, some detailed information was different, perhaps due to the different criteria and different time range and population.

Introduction

Most surveys of the prevalence of oral and maxillofacial (OMF) lesions in pediatric populations have only dealt with specific lesions, particularly tumors.[1—5] Following a review of the English language literature, to our knowledge, 13 previous studies reported the frequency of biopsied OMF lesions in pediatric patients in different countries from different continents,[6—18] with the longest follow-up data (30 years) and the highest numbers of lesions reported by Jones and Franklin[13] in England (Table 1). However, the follow-up and comparative data from the same region has not been documented. A review of pediatric OMF pathology in Southern Taiwan (1985—1996) has been compiled based on an examination of available biopsy records.[9] To date, the data of pediatric OMF lesions would be expected to be altered. Therefore, the current study aimed to provide updated information on the biopsied OMF pediatric lesions in Southern Taiwan, which were compared with our previous study and also with previous studies from Northern Taiwan and other countries.[6—18]

Materials and methods

A total of 36,264 diagnosis cases in the OMF region, from 1997 to 2011, were analyzed from the Oral Pathology Department of our institution. Within these 36,264 biopsies, a pool of 1023 samples from pediatric patients (0—15 years old) was included. Similar to our previous study,[9] age, sex, location, and histological diagnoses were recorded. Additionally, the data was divided into three different age levels: 0—5 years, 6—10 years, and 11—15 years. The OMF lesions were classified into four different categories: tumor/tumor-like lesions, cystic/pseudocystic lesions, inflammation/reactive lesions and other miscellaneous lesions (Table 2). Specimens without diagnosis or special findings were excluded.

Table 1 Studies of biopsied oral and maxillofacial lesions in pediatric populations from different countries.

Authors (year)	Country	Cases	Age, y	Period
Skinner et al (1986)[6]	USA	1525	0—19	14
Keszler et al (1990)[7]	Argentina	1289	0—15	25
Das and Das (1993)[8]	USA	2370	0—20	11
Chen et al (1998)[9]	Taiwan	534	0—15	12
Lawoyin et al (2000)[10]	Nigeria	561	0—16	10
Sousa et al (2002)[11]	Brazil	2356	0—14	15
Gultelkin et al (2003)[12]	Turkey	472	0—15	8
Jones and Franklin (2006)[13]	UK	4406	0—16	30
Dhanuthai et al (2007)[14]	Thailand	1251	0—16	15
Lima et al (2008)[15]	Brazil	625	0—14	20
Wang et al (2009)[16]	Taiwan	797	0—14	29
Shah et al (2009)[17]	USA	5457	0—16	16
Zuniga et al (2012)[18]	Chile	542	0—16	15
Lei et al (present study)	Taiwan	1023	0—15	15

Results

A total number of 1023 OMF biopsies were diagnosed from 1997 through 2011 (Table 2). The greatest number of lesions was in the inflammation/reactive group ($n = 451$), followed by the groups of tumor/tumor-like lesions ($n = 240$), cystic/pseudocystic lesions ($n = 196$) and other miscellaneous lesions ($n = 136$). The age of the patients in this series ranged from 2 months to 15 years, with an average age of 10.6 years (Table 3). The ratio of male to female patients was 1.18:1. The majority of lesions were in the 11—15 year age group ($n = 569$) and the 6—10 year age group ($n = 363$); the minority of lesions ($n = 91$) were in the 0—5 year age group.

The 12 most common lesions are shown in Table 4, and contributed about 77% of total lesions. The most common lesion was mucocele, followed by odontoma and dentigerous cyst. The five most common lesions were about 54% of all the pediatric OMF lesions.

The number in the odontogenic cyst group is shown in Table 5. Most of the odontogenic cysts were intrabony lesions, with the exception of gingival cysts and eruption cysts. Most of the odontogenic cystic lesions were in the 11—15 year age group, and accounted for about 68% of all the odontogenic cyst lesions. Dentigerous cysts were distributed more in the mandible ($n = 50$) than in the maxilla ($n = 33$); radicular cysts were also encountered more in the mandible ($n = 40$) than in the maxilla ($n = 24$). Keratocystic odontogenic tumors were localized and occurred more frequently in the mandible ($n = 18$) than in the maxilla ($n = 10$).

There were only five cases in the non-odontogenic cyst group. Three were epidermoid cysts, with two cases in the tongue and one case in the lower lip; two cases were in the 0—5 year age group and the remaining case was in the 11—15 year age group. The two cases of simple bone cyst were localized in the mandible and were in the 11—15 year age group.

The data of the odontogenic (benign) tumors group are shown in Table 6. All the odontogenic/benign tumors were

Table 2 Number and percentages of the four categories of oral and maxillofacial lesions in pediatric patients of the current study.

Categories		Total number	% of total
Tumor/tumor-like lesions	Odontogenic (benign)	158	15.44
	Non-odontogenic (benign)	76	7.43
	Non-odontogenic (malignant)	6	0.58
Cystic/pseudocystic lesions	Odontogenic	191	18.67
	Non-odontogenic	5	0.49
Inflammation/reactive lesions		451	44.10
Other miscellaneous lesions		136	13.29
Total number		1023	100

intrabony lesions, and most of them were located in the 11–15 year age group. Almost 68% were odontoma ($n = 108$), which were mainly localized in the maxilla ($n = 78$). The lesions of odontoma consisted of a compound type (54%), a complex type (28%) and a compound and complex type (18%). In contrast to odontoma, all of the ameloblastomas were distributed in mandible, and all were distributed in the 6–10 year and 11–15 year age groups, respectively.

Data for the non-odontogenic (benign) tumor/tumor-like group are presented in Table 7. Approximately 16% lesions occurred in the 0–5 year age group. Nearly half of the lesions in the non-odontogenic (benign) tumor/tumor-like group were fibromas, which were slightly more predominant in the 6–10 year and 11–15 year age groups, respectively. The second most common lesion in this group was hemangioma, which comprised the capillary type (42%) and the cavernous type (58%).

Only five types of lesions with a total number of six were found in the non-odontogenic (malignant) tumors group (Table 8). Different lesions occurred in different age groups with three in the mandible, two in the maxilla, and one in the lymph node.

Various types of lesion were noted in the inflammation/reactive group (Table 9). Most lesions were located in the 6–10 year and 11–15 year age groups. Mucocele was the most common lesion, mainly occurred in the lower lip. Inflammation was comprised of acute inflammation (12%), subacute inflammation (32%) and chronic inflammation (56%). Granulation tissue was mainly in the 6–10 year and 11–15 year age groups, and was distributed equally in the maxilla and mandible. With regards to fungus infection, candidiasis was predominantly found in the young age group and mainly occurred in the buccal mucosa and lower lip; the two cases of actinomycosis occurred in the oldest age group and were located in the lower lip and in the mandible; the only case of phycomycosis occurred in the oldest age group and was located in the lower lip. There were six cases of salivary gland origin, of which four were sialadenitis. Significantly, three cases of tuberculosis, which were all found in the oldest age group, were included in the group of inflammation/reactive lesions.

In the group of other miscellaneous lesions (Table 10), different types of lesion were found, which could be broadly divided into hard tissue and soft tissue pathology. The fewest number of lesions ($n = 8$) in the other miscellaneous lesions group occurred in the 0–5 year age group. In hard tissue pathology, most were dental pathology, whilst in soft tissue pathology, dental follicle ($n = 63$) was the most common lesion. Epithelial hyperplasia and hyperkeratosis predominated in the oldest age group.

Table 3 Age and sex distribution of all the oral and maxillofacial lesions in pediatric patients of the current study.

Age	Total number	Male:Female
0–5 y	91	1.2:1
6–10 y	363	1.3:1
11–15 y	569	1.1:1
Total number	1023	

Discussion

In the current study, we analyzed the prevalence of the biopsied OMF lesions occurring in a pediatric cohort in our institution. The OMF pathology service in our institution, not only serves biopsy lesions in the OMF area, but is also a major referral centre for biopsy in Southern Taiwan. Hence, the data presented in the current study can, in the most part, represent the prevalence of the pediatric OMF lesions in Southern Taiwan.

Table 4 Number and percentages of the 12 most common oral and maxillofacial lesions in pediatric patients in the current study.

12 most common lesions	Total	% of total
Mucocele	233	22.78
Odontoma	108	10.56
Dentigerous cyst	83	8.11
Inflammation	65	6.35
Radicular cyst	64	6.26
Dental follicle	63	6.16
Granulation tissue	46	4.50
Fibroma	36	3.52
Keratocystic odontogenic tumor	28	2.74
Pyogenic granuloma	21	2.05
Candidiasis	20	1.96
Ulcer	16	1.56
Total number	783	76.55

Table 5 Number and sex distribution of pediatric patients with odontogenic cyst in the current study.

Odontogenic cyst	0–5 years	6–10 years	11–15 years	Total number (male/female)
Dentigerous cyst	0	34	49	83 (57/26)
Radicular cyst	5	13	46	64 (41/23)
Keratocystic odontogenic tumor	0	4	24	28 (19/9)
Calcifying odontogenic cyst	0	1	4	5 (3/2)
Eruption cyst	0	1	4	5 (5/0)
Residual cyst	0	2	2	4 (2/2)
Globulomaxillary cyst	0	0	1	1 (1/0)
Gingival cyst	1	0	0	1 (1/0)
Total number	6	55	130	191 (129/62)

The total number of cases ($n = 1023$) (Table 1) was higher than that of our previous study ($n = 518$) published in 1998[9]; indicating an increase of about 50%. Furthermore, as indicated in Fig. 1, all of the four major categories of the biopsied pediatric OMF lesions in the current study have been highly, increased as compared with data from our previous study.[9] This large percentage of increase in the number of pediatric biopsies may suggest a higher awareness of pediatric oral health in our country. Most of the present pediatric cases were grouped in the inflammatory/reactive category (Table 2, 44.1%), which was similar to the study by Gültelkin et al (51%)[12] and Zuniga et al (75%)[18]; however, different in the percentage as compared to these two aforementioned studies.[12,18] Following the inflammatory/reactive lesions, cystic/pseudocystic lesions and tumor/tumor-like lesions were the second and third most common categories, respectively, and this finding was exactly the same as that in our previous study.[9] However, another study from Thailand[14] showed that cystic/pseudocystic lesions were the most common category.

Most of the pediatric lesions of the current study presented in the oldest age group (Table 3, 56%), which was comparable to our previous study[9] as well as the study from Northern Taiwan,[16] and also similar to the study by Jones and Franklin.[13] There were slightly more male patients than female patients (Table 3, 1.18:1); this trend was consistent with other reported studies.[12–14]

The 12 most common biopsied pediatric OMF lesions in the present series are listed in Table 4, and comprised a total number of 783, which constituted almost 80% of the total number of cases ($n = 1023$). Most of these 12 lesions were similar to our previous study[9] as well as the study from Northern Taiwan,[16] and despite a little difference in the order, with the first four lesions (i.e., mucocele, odontoma, dentigerous cyst and inflammation), were exactly the same. These first four most frequent lesions comprised approximately two-thirds of the 12 most common pediatric lesions. By contrast, dental follicles, keratocystic odontogenic tumors and candidiasis became the new common lesions documented in the present investigation. Similar to our previous study,[9] as well as the study from Northern Taiwan,[16] and also to the other previous studies,[13,14,18] mucocele was the most common lesion. Mucocele lesions in the present study consisted of more than one-fifth of all of the pediatric OMF lesions.

The odontogenic cyst category in our study comprised nearly 20% (191/1023) of all pediatric cases (Table 5), and males were more predominant than females. Dentigerous cysts and radicular cysts were the first two common lesions, which was similar to our previous study[9] and the study from Northern Taiwan.[16] The dentigerous cyst was the most common lesion in this group and most of these lesions were distributed in the 11–15 year age group, which was similar to the data of Dhanuthai et al[14] and Butt et al.[19] However,

Table 6 Number and sex distribution of pediatric patients with benign odontogenic tumors in the current study.

Benign odontogenic tumors	0–5 years	6–10 years	11–15 years	Total number (male/female)
Odontoma	2	41	65	108 (54/54)
Ameloblastoma	0	7	7	14 (8/6)
Odontogenic fibroma	0	4	6	10 (4/6)
Ameloblastic odontoma	0	2	5	7 (5/2)
Odontogenic myxoma	0	0	5	5 (2/3)
Ameloblastic fibroma	0	3	1	4 (3/1)
Cemento-ossifying fibroma	0	0	4	4 (0/4)
Adenomatoid odontogenic tumor	0	0	2	2 (1/1)
Ameloblastic fibro-dentinoma	0	1	0	1 (1/0)
Ameloblastic fibro-odontoma	0	0	1	1 (1/0)
Squamous odontogenic tumor	0	1	0	1 (0/1)
Cementoblastoma	0	0	1	1 (1/0)
Total number	2	59	97	158 (80/78)

Table 7 Number and sex distribution of pediatric patients with benign non-odontogenic tumors/tumor-like lesions.

Benign non-odontogenic tumor/tumor-like lesions	0–5 years	6–10 years	11–15 years	Total number (male/female)
Fibroma	10	13	13	36 (13/23)
Hemangioma	1	5	9	15 (11/4)
Papilloma	0	0	6	6 (5/1)
Lymphangioma	0	2	1	3 (1/2)
Nevus	1	0	2	3 (2/1)
Schwannoma	0	1	2	3 (0/3)
Verruca vulgaris	0	1	1	2 (1/1)
Fibrous dysplasia	0	0	2	2 (1/1)
Osteoma	0	0	1	1 (1/0)
Central giant cell granuloma	0	1	0	1 (1/0)
Lymphangiohemangioma	0	0	1	1 (0/1)
Neurofibroma	0	0	1	1 (0/1)
Fibromatosis	0	0	1	1 (0/1)
Verrucous hyperplasia	0	0	1	1 (1/0)
Total number	12	23	41	76 (37/39)

it was in contrast to the studies of Jones and Franklin[13] and Skiavounou et al,[20] in which radicular cysts were the most common lesions in these two studies.[13,20] The occurrence of keratocystic odontogenic tumors is relatively uncommon in pediatric patients and the peak of occurrence of this lesion is reported to be in the third decade of life.[21,22] Significantly, there were 28 cases of keratocystic odontogenic tumor in the present study, most of which were located in the 11–15 year age group. Moreover, the keratocystic odontogenic tumor lesions were more likely found in the mandible (19/28), which was similar to other previous reports.[21–23]

The first two most frequent lesions which occurred in the odontogenic (benign) tumor group were odontoma and ameloblastoma, respectively (Table 6), which was the same as the results from the study from Northern Taiwan[16] and the study by Dhanuthai et al.[14] In the current study, the frequencies of the different types of odontoma were 54%, 28% and 18% for compound, complex and mixed types, respectively, which was comparable to the report by Soluk Tekkesin et al,[24] with reported frequencies of 62%, 36% and 2%, respectively. Additionally, our data showed that more odontoma lesions were located in the 11–15 year age group, which was similar to the study by Soluk Tekkesin et al.[24] By contrast, two previous studies[25,26] showed that ameloblastoma was more common than odontoma, which was in contrast to our findings. Furthermore, all cases of pediatric ameloblastoma were located in the mandible, which was compatible with the findings of Servato et al (66%).[25] Ameloblastoma occurred more frequently in the oldest age group in the current study, which was consistent with the data of the study from Northern Taiwan.[16]

It is worth noting that the most common lesion was fibroma (47%), followed by hemangioma (20%) in the non-odontogenic (benign) tumor group/tumor-like group (Table 7) in our series; however, the reversed order of fibroma and hemangioma was noted in our previous study[9] as well as in the studies from Northern Taiwan[16] and another study from Thailand.[14] It is noteworthy that fibroma consisted of only about 27% in the group of non-odontogenic tumor/tumor-like lesions in the study on Israeli pediatric patients[4] as compared to the data from our series (47%). A probable reason for this disparity could be due to the geographic variation.

The fewest number of lesions were noted in non-odontogenic (malignant) tumors group as compared to the other groups (Table 8), which was comparable to our previous study[9] and the study from Northern Taiwan.[16] A similar trend of a relatively low number of non-odontogenic malignant tumors was also observed in other previous studies.[13,14,18] Taken together, this indicates that the incidence of non-odontogenic malignant OMF tumors is relatively uncommon in a pediatric population.

The highest number of the lesions in the present series was in the inflammation/reactive group (Table 9; 451/1023, 44%). Mucocele was the most common lesion, comprising about 52% of the lesions of the inflammation/reactive group; this was comparable to our previous study,[9] and

Table 8 Number and sex distribution of pediatric patients with non-odontogenic (malignant) tumors lesions in the current study.

Non-odontogenic (malignant) tumors	0–5 years	6–10 years	11–15 years	Total number (male/female)
Leukemia	0	2	0	2 (0/2)
Neuroblastoma	1	0	0	1 (0/1)
Follicular lymphoma	0	0	1	1 (1/0)
Ewing sarcoma	0	0	1	1 (1/0)
Rhabdomyosarcoma	0	0	1	1 (0/1)
Total number	1	2	3	6 (2/4)

Table 9 Number and sex distribution of pediatric patients with inflammation/reactive lesions in the current study.

Inflammation/reactive lesions	0—5 years	6—10 years	11—15 years	Total number (male/female)
Mucocele	35	107	91	233 (111/122)
Inflammation	7	19	39	65 (42/23)
Granulation tissue	2	13	31	46 (22/24)
Pyogenic granuloma	2	9	10	21 (10/11)
Candidiasis	11	3	6	20 (7/13)
Ulcer	0	6	10	16 (12/4)
Periapical granuloma	1	2	9	12 (3/9)
Gingival hyperplasia	0	3	2	5 (3/2)
Osteomyelitis	1	0	4	5 (2/3)
Necrotic tissue	0	3	1	4 (2/2)
Sialadenitis	1	1	2	4 (2/2)
Sequestrum	0	1	2	3 (3/0)
Tuberculosis	0	3	0	3 (2/1)
Lymphadenitis	0	1	1	2 (1/1)
Actinomycosis	0	0	2	2 (1/1)
Mucositis	0	1	1	2 (2/0)
Phycomycosis	0	0	1	1 (0/1)
Sinusitis	0	0	1	1 (1/0)
Myositis	0	1	0	1 (1/0)
Pulpitis	0	1	0	1 (1/0)
Periapical scar	0	0	1	1 (1/0)
Fistula	0	0	1	1 (1/0)
Sialolithiasis	0	0	1	1 (0/1)
Sialodochitis	0	0	1	1 (1/0)
Total number	60	174	217	451 (231/220)

Table 10 Number and sex distribution of pediatric patients with other miscellaneous lesions in the current study.

Other miscellaneous lesions	0—5 years	6—10 years	11—15 years	Total number (male/female)
Dental follicle	0	27	36	63 (29/34)
Epithelial hyperplasia	0	3	8	11 (9/2)
Hyperkeratosis	2	2	5	9 (4/5)
Scar	0	2	7	9 (5/4)
Supernumerary tooth (exclude mesiodens)	0	4	3	7 (6/1)
Bone fragment	0	1	3	4 (2/2)
Mesioden	0	2	2	4 (1/3)
Fibrosis	0	1	2	3 (1/2)
Tooth fragment	1	1	1	3 (2/1)
Oral mucosa	1	2	0	3 (2/1)
Oral submucous fibrosis	0	0	2	2 (2/0)
Root resorption	0	0	2	2 (0/2)
Exostosis	0	1	1	2 (1/1)
Foreign body	0	0	2	2 (2/0)
Dentinogenesis imperfecta	2	0	0	2 (0/2)
Hematoma	0	2	0	2 (1/1)
Melanosis	1	1	0	2 (1/1)
Cemento-osseous dysplasia	0	0	1	1 (0/1)
Osteosclerosis	0	0	1	1 (1/0)
Blood clot	0	0	1	1 (1/0)
Natal tooth	1	0	0	1 (0/1)
Tooth germ	0	0	1	1 (0/1)
Adipose tissue	0	1	0	1 (1/0)
Total number	8	50	78	136 (71/65)

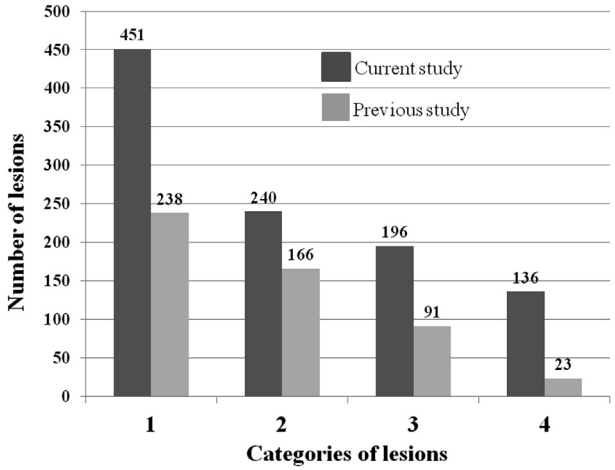

Figure 1 All of the four major categories (1, inflammation/reactive lesions; 2, tumor/tumor-like lesions; 3, cystic/pseudocystic lesions; 4, other miscellaneous lesions) of the biopsied pediatric oral and maxillofacial lesions in the current study (*) were highly increased as compared with the data of our previous study (**).[9]

other previous studies,[13,14,18] including the study from Northern Taiwan.[16] Inflammation and granulation tissue were, respectively, the second and the third common lesions, which was also the same as in our previous study[9] and the study from Northern Taiwan.[16]

For lesions categorized in the group of other miscellaneous lesions, dental follicle was the most common lesion (46%), which was the same as the study from northern Taiwan.[16] Significantly, 11 cases of epithelial hyperplasia (the second most common lesion) and nine cases of hyperkeratosis (the third most common lesion) were diagnosed in our series, which were predominantly located in the 11—15 year age group. These two oral disease entities, to our knowledge, have not been reported in previous retrospective studies of biopsied OMF lesions in pediatric patients (Table 1).[6—18]

In conclusion, we present epidemiological data (1997—2011) from a pediatric population in Southern Taiwan, in which there was an increase of about 50% of the number of biopsied OMF lesions as compared to our previous data (1985—1996). The highest number of all lesions in the current study was located in the 11—15 year age group and belonged to inflammatory/reactive diseases. Mucocele was the most common of all lesions; odontoma was the most common odontogenic tumor/tumor-like lesion, whereas fibroma was the most frequent non-odontogenic tumor/tumor-like lesion. Additionally, some detailed information was different, which perhaps due to the different time range and geographic variation as compared with previous studies. Hence, the current data would aid both the epidemiologic investigation and educational teaching in pediatric OMF lesions.

Conflicts of interest

The authors declare that there are no conflicts of interest that could influence their work.

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