

Clinical and Histopathological Profiles of Pediatric and Adolescent Oral and Maxillofacial Biopsies in a Persian Population

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Abstract

Introduction

The frequency of pediatric and adolescent oral and maxillofacial lesions is various in different societies. The present study was aimed at investigating the frequency of oral and maxillofacial pediatric and adolescent biopsies in Zahedan, Southeastern Iran, and compare the results with other epidemiologic studies.

Methods and Materials

This retrospective study reviewed oral and maxillofacial lesions in patients aged 0-18 years old referring to the treatment centers of Zahedan University of Medical Sciences, during 12-years period. Patients' demographic information including age, gender and location of the lesion were collected and statistically analyzed with SPSS software, version 19.

Results

In general, among 1112 oral and maxillofacial lesions, 154 (13.9%) cases were related to children and adolescents younger than 18 years old. The average age of patients was 11.4 ± 4.9 , 53.2% and 46.8% of them were boys and girls, respectively. The most frequent sites of lesions were the gingiva and lip. The most prevalent lesions included inflammatory/reactive, cystic and neoplastic lesions, respectively. Benign and malignant tumors comprised 12.3% and 4.5% of cases. Moreover, pyogenic granuloma and Peripheral Giant Cell Granuloma (PGCG) were the most frequent lesions.

Conclusion

The most prevalent oral and maxillofacial lesions in patients under 18 years old were inflammatory/reactive lesions in gingiva and in the 13-18 age range. Determining the characteristics of these lesions in the children and adolescents population provides a firm groundwork for proper diagnosis and treatment.

Key words: Adolescents, Children, Oral biopsy, Oral lesions.

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Introduction

Identification of oral and maxillofacial pathologies plays a major role in improved early prevention and detection, rapid treatment and provision of better health services (1). In spite of World Health Organization (WHO) suggestions regarding the epidemiologic assessment of oral lesions, the majority of studies on oral conditions in children include the investigation of caries, periodontal disease, malocclusion and dental trauma(1, 2). Also the majority of pediatric oral and maxillofacial pathological studies are limited to certain lesions, such as jaw cysts, odontogenic tumors, malignant tumors and salivary gland tumors (3-6).

In Asia, few studies have investigated all pediatric and adolescent oral and maxillofacial lesions (7-10). The oral lesions of this age range have only been studied in the northern part of Iran(11). Since the epidemiology of oral and maxillofacial lesions is different in various geographic regions, the present study investigated the frequency of oral and maxillofacial biopsies in the children and adolescents referring to the treatment centers of Zahedan University of Medical Sciences, Southeastern Iran, and compared the results with other epidemiologic studies.

Methods and Materials

This retrospective cross-sectional study investigated documents of patients with oral and maxillofacial lesions referring to the treatment centers of Zahedan University of Medical Sciences, including Khatam-Al-Anbia Hospital and Faculty of Dentistry between 2001 and 2012. This project has been approved as a dentistry dissertation in the Ethics Committee of Zahedan University of Medical Sciences, with ID code: 545 in July 2011. Samples of oral and maxillofacial

biopsies related to child and adolescent patients aged 0-18 years old were separated and reviewed by two pathologists then categorized into four general categories: inflammatory/reactive, neoplastic, cystic and others lesions. Afterwards, patients' demographic information including age, gender and location of lesion were extracted from the documents and records. The patients were also categorized into 0-5, 6-12 and 13-18 age ranges. Statistical analysis was performed using SPSS software version 19.0 (SPSS Inc, Chicago, IL) and chi-square test (P-value<0.05 was considered statistical significant).

Results

In this study a total of 1,112 oral and maxillofacial samples were investigated; 154 cases of them (13.9%) related to children and adolescents. In addition, 79 cases (51.3%) were boys with the average age of 10.9 ± 5.1 and 75 cases (48.7%) were girls with the average age of 12 ± 4.7 . The occurrence of lesions in boys and girls were mostly in the age range of 6-12(43%) and 13-18(60%), respectively, which was statistically significant (P=0.047).

In the order of frequency, lesions include inflammatory/reactive lesions (48.1%), cystic lesions (22.7%), neoplastic lesions (19.5%) and others lesions (9.7%). The most prevalent lesion types were pyogenic granuloma (13.6%), peripheral giant cell granuloma (9.1%), dentigerous cyst (7.8%), radicular cyst (5.2%) and mucocele (5.2%). Hemanangioma was the most prevalent benign tumor (26.1%) and mucoepidermoid carcinoma (28.6%) and squamous cell carcinoma (28.6%) were the most prevalent malignant tumors. The Male: Female (M:F) ratios of inflammatory/reactive, cystic and neoplastic lesions were 1.2:1, 1.1:1 and 0.5:1, respectively. Chi-square test didn't

show significant statistical relationship between genders in the four lesion groups ($P=0.1$). (Table.1) represents the age and gender distribution of oral and maxillofacial lesions in patients younger than 18 years old. The overall incidence of lesions in the 13 -18 age range was significantly higher than the 5-0 and 6-12 age ranges ($P=0.01$). Most inflammatory/reactive oral and maxillofacial lesions in children and adolescents under 18 years old were on the gingiva (41.9%), while 77.1% of cystic lesions were intraosseous (Table. 2). The majority of benign neoplastic lesions were on the gingiva and palate (26.1%), while

malignant neoplastic lesions were scattered in different oral and maxillofacial sites to an almost equal extent.

Other lesions included two precancerous leukoplakia in the tongue of 6 and 14 years old boys, two tumor-like fibrous dysplasia in the mandible of 6 months old girl and 13 years old boy, and the remaining cases included lesions with no definitive histopathological diagnosis. The age and gender distribution of children and adolescents are presented in (Tables. 3-5) separately according to each category.

Table 1: The age and gender distribution of oral and maxillofacial lesions in children and adolescents in Southeast Iran

Pathological diagnosis	Gender			Age(yr.)		
	Male	Female	Total	Mean	SD	Range
Reactive/inflammatory lesions	41(55.4)	33(44.6)	74(100)	11.46	4.66	1-18
Cystic lesions	18(51.4)	17(48.6)	35(100)	11.46	4.38	0.2-18
Neoplastic lesions	10(33.3)	20(66.7)	30(100)	11.10	5.95	1-18
Other lesions	10(66.7)	5(33.3)	15(100)	12.30	5.30	0.5-18
Total	79(51.3)	75(48.7)	154(100)	11.47	4.90	0.2-18

Table 2: The location distribution of oral and maxillofacial lesions in children and adolescents in Southeast Iran

Pathological diagnosis	Locations								Total
	lip	palate	tongue	buccal mucosa	gingiva	maxilla	mandible	others	
Reactive/inflammatory lesions	17 (23)	2 (2.7)	4 (5.4)	6 (8.1)	31 (41.9)	2 (2.7)	2 (2.7)	10 (13.5)	74 (100)
Cystic lesions	0	0	0	0	0	12 (34.2)	15 (42.9)	8 (22.9)	35 (100)
Neoplastic lesions	2 (6.7)	5 (16.7)	2 (6.7)	2 (6.7)	3 (10)	3 (10)	2 (6.7)	11 (36.7)	30 (100)
Other lesions	1 (6.7)	1 (6.7)	2 (13.3)	1 (6.7)	3 (20)	0	2 (13.3)	5 (33.3)	15 (100)
Total	20 (13.1)	8 (5.2)	8 (5.2)	9 (5.9)	37 (24.2)	17 (11.1)	19 (12.4)	35 (22.9)	154 (100)

Table 3: The age and gender distribution of inflammatory/reactive lesions in children and adolescents in Southeast Iran

Pathological diagnosis	Gender		Total	Age(yr.)		
	Male	Female		0-5	6-12	13-18
Mucocele	6	2	8	1	4	3
Chronic sialadenitis	3	1	4	0	1	3
Sjögren syndrome	1	0	1	0	0	1
PG	12	9	21	0	10	11
PGCG	8	6	14	2	9	3
POF	0	3	3	0	1	2
Inflammatory fibrous hyperplasia	0	1	1	0	0	1
Fibroepithelial hyperplasia	3	1	4	2	2	0
Oral focal mucinosis	0	1	1	0	0	1
CGCG	1	2	3	0	2	1
Lichen planus	0	1	1	0	0	1
Periapical granuloma	0	2	2	0	0	2
Unspecified ulcer	3	2	5	1	1	3
Granulomatous inflammation	2	1	3	1	0	2
Granulation tissue	2	1	3	2	1	0
Total	41	33	74	9	31	34

PG: Pyogenic Granuloma, PGCG: Peripheral Giant Cell Granuloma, POF: Peripheral Ossifying Fibroma, CGCG: Central Giant Cell Granuloma.

Table 4: The age and gender distribution of cystic lesions in children and adolescents in Southeast Iran

Pathological diagnosis	Gender		Total	Age(yr.)		
	Male	Female		0-5	6-12	13-18
Odontogenic cysts						
Dentigerous cyst	6	6	12	0	10	2
Radicular cyst	5	3	8	0	3	5
Buccal bifurcation cyst	1	1	2	0	2	0
Orthokeratinized odontogenic cyst	0	1	1	0	1	0
Infected odontogenic cyst	1	1	2	0	0	2
Total	13	12	25	0	16	9
Nonodontogenic cysts						
Aneurysmal bone cyst	1	1	2	0	1	1
Simple bone cyst	0	1	1	0	0	1
Globulomaxillary cyst	0	1	1	0	0	1
Epidermoid cyst	2	2	4	1	1	2
Thyroglossal duct cyst	1	0	1	1	0	0
Bronchial cyst	1	0	1	0	0	1
Total	5	5	10	2	2	6

Table 5 :The age and gender distribution of neoplastic lesions in children and adolescents in Southeast Iran

Pathological diagnosis	Gender		Total	Age		
	Male	Female		0-5	6-12	13-18
Benign tumors						
PA	0	3	3	0	0	3
Papilloma	2	2	4	3	0	1
Fibroma	0	4	4	0	0	4
Angiofibroma	1	0	1	1	0	0
Neurofibroma	1	0	1	0	0	1
Hemangioma	2	4	6	3	1	2
Ossifying fibroma	1	1	2	0	0	2
Complex odontoma	0	1	1	0	0	1
Unicystic ameloblastoma	1	0	1	0	0	1
Total	8	15	23	7	1	15
Malignant tumors						
MEC	0	2	2	1	0	1
SCC	1	1	2	1	0	1
Fibrosarcoma	1	0	1	1	0	0
Osteosarcoma	0	1	1	0	0	1
Langerhans cell histiocytosis	0	1	1	0	0	1
Total	2	5	7	3	0	4

PA: Pleomorphic Adenoma, MEC: Mucoepidermoid Carcinoma, SCC: Squamous Cell Carcinoma

Discussion

In the present study, 13.9% of oral and maxillofacial samples related to children and adolescents under 18 years old, which is more than studies in Brazil and India and less than Thailand, Nigeria, Chile, Uganda and northern Iran surveys (8, 11-18). General differences between various studies can be due to the inclusion criteria, including age range or locations (oral or oral and maxillofacial). In addition, population's genetic background, geographical area, study period and the type of medical center in which the study is conducted can play a role in these differences (8).

The majority of lesions (51.3%) were in the 13-18 age range and it may be generally said that the occurrence of oral and maxillofacial lesions increase in children and adolescents with age increasing. This result is consistent with those of other studies (11, 17, 19, 20). Nevertheless, some studies have reported increased occurrence of oral lesions in the mixed dentition period (8, 21) and suggested that maintaining oral health problems and trauma can be the primary factors in the development of oral mucosal lesions in this dentition period (21).

Similar to the present study, studies in UK, Thailand and Uganda have shown the higher prevalence of oral mucosal lesions in boys (2, 8, 18). However, the results of other studies are different, in a way that some studies have reported equal or even more prevalence in girls (15-17). In the present study, the gingiva was the most common site of oral and maxillofacial lesions (22.7%) and the lip was the second most common site. This result is consistent with those of the study by Gültelkin et al. (21). It should be noted that in the majority of studies, the localization of lesions are mentioned according to their type.

In the present study, inflammatory/reactive lesions (48.1%) were the most common, which is similar to other studies (9,11,16). Siadati et al. stated higher prevalence of inflammatory/reactive lesions due to symptomatic nature of them compared to other lesions (11). In studies of Thailand, Nigeria and Uganda, these lesions are the second most prevalent category (8, 14, 18). The majority of inflammatory/reactive lesions in this study are in the permanent dentition period similar to those in the study by Kamulegeya and Lakor (18), while Dhanuthai et al., Gultelkin et al. and Wang et al. reported higher prevalence of oral lesions in the mixed dentition period (8,9,21). Present study showed the majority of inflammatory/reactive lesions occurred in boys, which is inconsistent with similar studies (8, 9, 11, 16). Pyogenic granuloma was the most prevalent inflammatory/reactive lesions (28.4%) and oral and maxillofacial lesions (13.6%) in patients younger than 18 years old, while the majority of studies reported mucocele as the most prevalent inflammatory/reactive lesion in children and adolescents(9, 11, 16, 20). Poor oral hygiene, abnormal tooth development and some drugs are suggested

as the etiologic causes of pyogenic granuloma. Also, these lesions probably develop in the second decade of female's life due to the vascular effects of female hormones (22).

Cystic lesions were the most prevalent category in the studies in Thailand, Saudi Arabia and India (8, 10, 17); while in the present study, they were the second most prevalent category (22.7%). The majority of cystic lesions were observed in the 13-18 age range. Wang et al., Al-Yamani et al. and Kamulegeya and Lakor showed the higher prevalence of cystic lesions in the permanent dentition period (9, 10, 18), while Dhanuthai et al. and Gultekin et al. showed this higher prevalence in the mixed dentition period (8, 21). Sexual predominance of cystic lesions is reported in different studies (11, 17, 23) while the present study showed the approximately equal prevalence in both genders (M:F=1.1:1), which is only consistent with the results of Bonder(3).

In this study, the majority of cystic lesions in children and adolescents were in mandible that is similar to other studies (16, 21, 23). The prevalence of odontogenic cysts were 2.5 times non-odontogenic cysts. The majority of studies have reported the prevalence of odontogenic cysts to be higher than non-odontogenic cysts (2, 8, 11, 23). However, Kamulegeya and Lakor have shown equal prevalence of odontogenic and non-odontogenic cysts(18). Similar to other studies, dentigerous cyst was the most prevalent jaw cyst in children and adolescents (3, 17, 23). However, studies in the UK, Turkey and northern Iran have reported radicular cyst as the most common cystic lesion in children (2, 11, 21). Dentigerous cyst can be associated with

more important conditions such as odontogenic keratocyst and cystic ameloblastoma. This association with more serious conditions represents the significance of emphasizing the jaw cysts histopathology, even when they are clinically a dentigerous cyst (24).

Oral and maxillofacial tumors in children are indicative of heterogeneous pathological conditions from real neoplasms to hamartoma (25). The etiology of most oral and maxillofacial neoplasms remains unknown. Genetic susceptibility as well as environmental factors such as viral infection, nutritional deficiencies, trauma and consumption of alcohol and tobacco have suggested as their risk factors (5). Neoplastic lesions comprised 19.5% of all oral and maxillofacial lesions in patients younger than 18 years old, which is less than the study in northern Iran but higher than other studies (11, 26-28).

Among oral and maxillofacial tumors in patients younger than 18 years old, 73.1% and 26.9% were benign and malignant, respectively. In the majority of other studies as well, the prevalence of benign tumors were more than malignant ones except for the African studies (18, 26, 29-34) (Table.6).

About two thirds of neoplastic lesions were observed within the 13-18 age range. This age range comprises the most common period in the majority of studies (28, 32-34). Neoplastic lesions in female children and adolescents were twice boys in Zahedan, while in other parts of Iran; this prevalence was higher in boys (11, 28).

Hemangioma, benign proliferation of endothelial cells, is the most common neoplasm in children which is similar to our studies (21, 25, 31). However, in some studies, odontogenic tumors such as ameloblastoma and odontoma were the most prevalent benign tumor in children (8, 26, 33). Nevertheless, since many cases of hemangioma are clinically diagnosed, this lesion is not always biopsied, and its real prevalence is higher than existing reports.

Generally, the prevalence of oral and maxillofacial malignant tumors is reported 1.2% to 83.6% (12, 18, 32, 34). Albright et al. showed that after a 23 years period, the prevalence of head and neck malignancies increased in children younger than 15 years old (35). Squamous cell carcinoma (SCC) and mucoepidermoid carcinoma were the most common malignancies in this study. Chow et al. showed the higher prevalence of SCC in patients younger than 20 years compared to the past. Awareness of this increased prevalence in children, early biopsies in disposed individuals and consultation with adult pathologist can decrease delayed diagnosis (36). Furthermore, no case of lymphoma was observed in this study despite lymphoma was reported as the most common malignancy in children in different studies (5, 18, 26, 34).

It should also be noted that oral and maxillofacial tumors in children are significantly different comparison with adults regarding treatment, clinical behavior and histopathologic feature(25). These differences increase the necessity of early diagnosis and timely treatments in children and adolescents.

Table 6: Comparison between the oral and maxillofacial tumors of the present study and other studies

Author et al.	year	country	Period (years)	Age range	M:F ratio	Localization	Benign	Malignant	Odontogenic
Tanaka(29)	1999	Japan	20	≤15	0.88:1	Oral & Maxillofacial	97.1%	2.9%	*
Adebayo(30)	2001	Nigeria	20	≤15	1.4:1	Oral & perioral	70.2%	29.7%	25.9%
Kalyanyama(34)	2002	Tanzania	15	≤16	1.1:1	Oral	57%**	43%	5.1%
Al-Khateeb(25)	2003	Jordan	10	<19	1.2:1	Oral & Maxillofacial	90%	10%	7%
Tröb(31)	2003	Germany	30	≤16	0.7:1	Oral	87%**	13%	***
Gültekin(21)	2003	Turkey	8	≤15	0.8:1	Oral	89	11	*
Tanrikulu(32)	2004	Turkey	18	≤15	1.1:1	Maxillofacial	93.3%	6.7%	23.3%
Aregbesola(26)	2005	Nigeria	10	≤19	1.4:1	Orofacial	49%	51%	49%
Elarbi(27)	2009	Libya	17	≤18	0.7:1	Orofacial	96.2%	3.7%	16.4%
Jaafari-Ashkavandi(28)	2011	Iran	5	≤18	1.4:1	Orofacial	75.3%	24.6%	24.6%
Kamulegeya(18)	2011	Uganda	5	≤16	1.3:1	Oral & Maxillofacial	16.4	83.6	5.1
Iatrou(33)	2013	Greek	11	≤15	1.1:1	Orofacial	90.05	9.95	19.0
Present study	2012	Iran	11	≤18	0.5:1	Oral & Maxillofacial	76.7%	23.3%	6.7%

* Not exactly defined. **Benign tumors Plus Tumor like lesions. ***Odontogenic tumors were excluded.

Conclusion

The frequency of oral and maxillofacial lesions is different in various geographical regions. This study investigated the frequency of these lesions in Zahedan, Iran. The most prevalent lesions included inflammatory/reactive, cystic and neoplastic lesions, respectively. Benign tumors were more prevalent than malignant tumors. Determining the characteristics of these lesions in the child and adolescent population provides a firm groundwork for proper diagnosis and treatment.

Authors' Contribution

Saravani S: concept and design, data analysis, manuscript preparation.

Kadeh H: concept and design and interpretation and critical revision of the manuscript.

Amirabadi F: concept and design and interpretation and critical revision of the manuscript.

Keramati N: Acquisition of data; Data Analysis and Interpretation.

All authors approved the final version of the paper.

Conflict of Interest: None

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