ORIGINAL RESEARCH

Prevalence of Cysts in a Teaching Institution – A Retrospective Study

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Abstract

Background: Orofacial cysts may be classified as odontogenic or non-odontogenic depending on their origin. Odontogenic cysts (OCs) are more common and can be developmental or inflammatory. These cysts present clinically in diverse patterns and may range from an innocuous, tiny lesion to a large, destructive lesion. Given their variable behavior and potential for recurrence, understanding their distribution and biological nature is essential. Therefore, the present study aimed to evaluate the prevalence and distribution of orofacial cysts reported in our institution.

Methods: Patient records diagnosed as orofacial cysts were collected from the institutional database over a period of 20 years. The cases were analyzed for anatomical locations, demographic and histopathological details.

Results: The present study revealed 398 jaw cysts, of which 95.97% were odontogenic and 4.02% non-odontogenic. Of odontogenic cysts, 99 were inflammatory and 191 were developmental. Radicular cysts (RC) had highest prevalence rate followed by odontogenic keratocyst (OKC) and dentigerous cysts (DC). Among non-odontogenic, dermoid cysts were the most prevalent. The third decade was seen to be the most predominant age of occurrence with a male predilection and a propensity for the mandibular posterior segment.

Conclusion: The prevalence rate of orofacial cysts emphasizes the need to differentiate them from other similar lesions to understand their nature, aid in early diagnosis and plan the treatment accordingly to ensure a better prognosis.

KEYWORDS: Orofacial cysts, odontogenic cysts, non-odontogenic cysts, jaw, odontogenic keratocyst, Goltz- Gorlin syndrome, recurrence.

Introduction

Cyst is a pathological cavity having fluid, semifluid, or gaseous contents.[1] Most of the Odontogenic cysts (OCs) originate from the epithelial component of the odontogenic apparatus or its remnants (odontogenic), while others arise from other epithelial sources in the jaws and surrounding soft tissues (non-odontogenic).[2,3] OCs occur more frequently than non-odontogenic.[4] Depending on the etiology, odontogenic and non odontogenic cysts (NOCs) are categorized into two types: developmental and inflammatory.[5] OCs has a diverse pattern of presentation clinically and may vary from being an innocuous tiny lesion, detected accidentally in radiographs to a large destructive lesion. Intraosseous lesions may weaken the bones, leading to functional changes, predispose to infection and pathologic fractures. It can also cause disruption of adjacent tooth eruption, tooth displacement, root resorption and canal displacement.[6]

Though cysts appear as a swelling on clinical examination, they are only liquid or gas filled cavities, not composed of a solid mass in contrast to neoplasms. Proper histopathological examination is required to distinguish these jaw lesions, as errors can result in inappropriate treatment. It is thus pivotal to have knowledge of the type, biological behaviour and distribution of the jaw cysts for ideal management of these lesions. Hence the present study is aimed to analyze the prevalence and distribution of histopathologically diagnosed orofacial cysts reported in our institution during a period of 20 years, using the data retrieved from patient records.

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Materials and Methods

The retrospective study was conducted in the Oral Pathology department, Meenakshi Ammal Dental College, India. Data was retrieved from the case records of the biopsied specimens received over a period of 20 years, from 2001 to 2020. All the histopathologically confirmed cases of orofacial cysts were included in the present study. Other cases were excluded. The collected data was segregated according to the type of jaw cyst, age of occurrence (by decade) and sex of the patient, site, side involvement (right/left/both), segment (anterior/posterior/both), type of cystic content, association with impacted tooth, nature of keratinization of lining epithelium, presence/absence of satellite cysts, cholesterol clefts, duration of recurrence and association with syndrome. Statistical analysis included multinominal logistic regression analysis and correlation within the different categories using "Stata/SE version 17.1" (Statacorp. College station, Texas).

Results

A total of 398 cases of jaw cysts were reported out of which 382 cysts (95.97%) were OCs and 16 (4.02%) were of NOCs. Among the OCs, 99 belonged to the inflammatory type, 191 to the developmental type and 92 were infected cysts. Among the 16 NOCs, 11 were developmental while only 5 were of inflammatory origin (Table 1). Radicular cyst (RC)(n=84,21.99%), showed the highest prevalence amongst the OCs followed by odontogenic keratocyst (OKC)(n=80, 20.9%)and dentigerous cyst (DC)(n=71,18.58%). Dermoid cyst, an extraosseous developmental type cyst showed the highest prevalence among NOCs (n=6, 37.5%). 30.40% cases were found to occur in the third decade of life, followed by the second and fourth decades (18.8%) with a male predilection (60.1%). Slight predilection for mandible (54.0%) over maxilla (46.0%) was noted. Left side (45.2%) was more commonly involved than the right (40.7%)

Table 1: Prevalence of cysts of the jaws reported in the institution from 2001-2020

Type of Jaw Cysts	N			Frequency (%) overall	Frequency (%) within category
ODONTOGENIC CYSTS		**382**		95.9798995	-
Developmental	**INTRAOSSEOUS**	**190**	**191**	-	
	Odontogenic Keratocyst	80		20.100506	20.9424084
	Orthokeratinized Odontogenic Cyst	21		5.27638191	5.497382199
	Dentigerous Cyst	71		17.8392	18.58639
	Lateral Periodontal Cyst	03		0.753768844	0.785340314
	Glandular Odontogenic Cyst	05		1.25628141	1.30890052
	Calcifying Epithelial Odontogenic Cyst	10		2.512562814	2.617801047
	EXTRAOSSEOUS	**01**		-	
	Eruption Cyst	01		0.251256281	0.261780105
Inflammatory	Apical Periodontal Cyst	84	**99**	21.1	21.99
	Residual Cyst	10		2.512562814	2.617801047
	Paradental Cyst	05		1.256281407	1.308900524
Infected Cyst	**Infected Cyst**	**92**	**92**	23.11557789	24.08376963
NON ODONTOGENIC CYSTS		**16**		4.020100503	-
Developmental	**INTRAOSSEOUS**	**04**	**11**	-	
	Nasopalatine Duct Cyst	03		0.753768844	18.75
	Median Palatal Cyst	01		0.251256281	6.25
	EXTRAOSSEOUS	**07**		-	
	Epidermoid Cyst	01		0.251256281	6.25
	Dermoid Cyst	06		1.507537688	37.5
Inflammatory	**TRAUMATIC**	**05**	**05**	-	
	Salivary Cyst	01		0.251256281	6.25
	Traumatic Bone Cyst	01		0.251256281	6.25
	Aneurysmal Bone Cyst	03		0.753768844	18.75

and the remaining cases involved both sides (14.1%). Posterior segment of the jaws (57.5%) was more commonly involved than the anterior (33.2%) with 9.3% involving both segments.

Among the jaw cysts, 68 (17.1%) cases (DCs, RCs, OKCs, OOCs, COC) were associated with an impacted tooth. Aspirate was present in 8.3% (n=33) of the jaw cysts with straw coloured fluid (n=12, 36.36%) being the most common, followed by cheesy white material (n=8, 24.24%) and pus (n=7, 21.21%). Recurrence was found in 5 cases (1.2%), of which 2 cases were OKCs associated with the Gorlin-Goltz syndrome. Histologically, most of the jaw cysts had a non-keratinized epithelial lining (68.1%) with keratinization (parakeratin-20.4%, orthokeratin-8%, mixed- 3.5%) seen in OKCs, OOCs, DCs, RCs, paradental and some infected cysts. Satellite cysts were found in 5 cases (1.2%) of which 2 were OKCs. Cholesterol clefts were observed in 11.8% of jaw cysts, predominantly in the RCs and DCs.

Discussion

Jaw cysts are often asymptomatic and most of the times detected accidentally by the clinicians while examining the patients for other dental complaints. However, some cysts, like developmental OCs, have a tendency to destroy adjacent tissues, which leads to expansion or resorption of the jaw and at times, pathological fractures. In addition to this, there are chances of infection and even neoplastic changes in the cystic epithelium. Moreover, these cysts could present with overlapping clinical, radiographic, histopathological features with other cysts and tumours that could occur in the same site. Hence knowledge regarding the frequency of jaw cysts, predilection with respect to patient's age and gender, site, content, association with syndrome, etc. helps to determine their demographic data, which is essential for the diagnosis and management.[7]

In the present study, 95.97% were OCs which was in accordance with studies done in Brazil (92.9%) and Turkey (98.5%).[8,9] RCs (21.99%) were most common, followed by OKCs and DCs respectively (20.68%, 18.58%). About 24% of the OCs were reported as infected cysts, characterized by the absence of epithelium, without any characteristic feature of a specific OC (RCs/DCs/OKCs). As the cysts are mostly asymptomatic, they are often neglected and the patients report only when they turn symptomatic due to infection making precise diagnosis challenging.

RCs were predominantly reported in our study which can

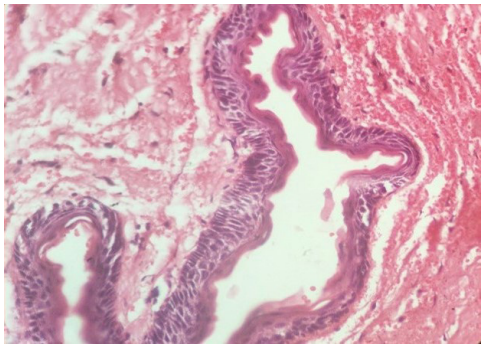

Fig 1: Microscopic image showing corrugated, parakeratinized stratified squamous cystic lining epithelium of odontogenic keratocyst with palisaded basal cells exhibiting tombstone like appearance. (10X magnification)

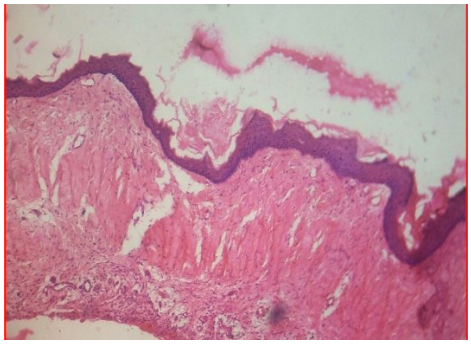

Fig 2: Microscopic image showing stratified squamous cystic lining epithelium of Orthokeratinized odontogenic keratocyst with prominent stratum granulosum exhibiting onion peel appearance of orthokeratin within the lumen. (10X magnification)

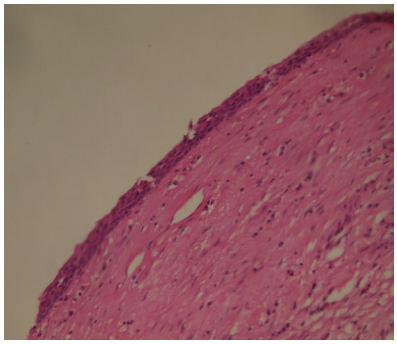

Fig 3: Microscopic image showing cystic lining epithelium of dentigerous cyst resembling reduced enamel epithelium. (4X magnification)

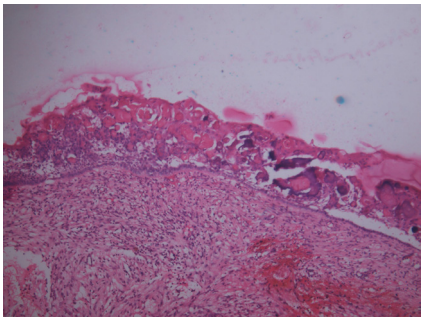

Fig 4: Microscopic image showing cystic lining epithelium of calcifying odontogenic cyst showing numerous ghost cells and calcifications. (10 X magnification)

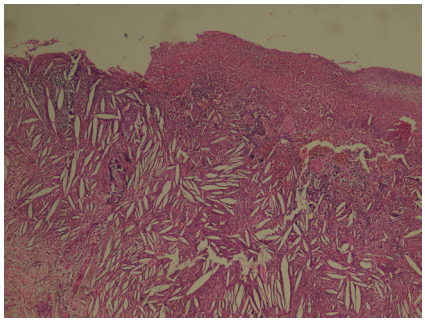

Fig 5: Microscopic image showing cholesterol clefts present in Periapical cyst (10 X magnification)

be attributed to their origin and association with non-vital tooth due to dental caries and trauma. Various studies have reported that the frequency of RCs among OCs range from 40-60%, which was comparatively higher probably due to the fact that some of them were reported as infected cysts in the present study.[1,4,10] 10 cases of residual cyst, a subtype of RC was recorded. It occurs at the extraction site of the offending tooth in cases, where a periapical cyst or granuloma is left untreated or incompletely removed. Paradental cysts (1.30%) in our study was much lower than a Brazilian study (8.7%).[8] Associated with vital, partially erupted mandibular third molars, the cystic lining could either be reduced enamel epithelium or sulcular/ junctional epithelium.[5] Usually, DCs are known to be more prevalent than OKCs.[9,11,12,13] However in our study, a marginally higher prevalence of OKCs over DCs was evident, which was in accordance with other studies done in the South Indian and Iranian population.[10,14] This could be ascribed to the fact that not all specimens of DCs get sent for histopathological examination during the removal of impacted tooth if the diagnosis is clinically obvious. Also, the geographical differences and sample size variations could result in the differences observed.[15] Out of the 80 cases of OKCs (Figure 1), 4 cases were reported as 'orthokeratinised OKC' as areas of orthokeratinisation were evident within the lining epithelium. 9 cases were reported as 'infected OKC' due to the presence of inflammatory cells infiltrating the connective tissue with thickening and loss of keratinization in the adjacent epithelium and rete ridge formation. However, features of typical OKC were present in few areas.

Orthokeratinised odontogenic cyst (OOC), previously a variant of OKC, has been recognized as a separate entity in the 2017 WHO classification owing to its characteristic histological features (Figure 2). In the present study, 5.49% of cysts were reported as OOCs which was much higher than that observed in studies done in Brazil (0.5%) and South India (0.36%).[8,16]

The third most commonly occurring cyst in this study was the DC (Figure 3), which is formed as a result of fluid accumulation between the reduced enamel epithelium and the crown of an impacted tooth. They usually comprise about 20% of all OCs.[5] In a study by Olusanya et al involving a PubMed literature search, DC was the second most frequent OC in 52.8% of the articles,however OKC was the second most frequent in our study.[15] The infected DCs reported here exhibited few areas of reduced enamel epithelium and in other areas;the lining stratified squamous epithelium was very thin with absence of rete pegs.[17] One case of eruption cyst was also recorded.

The glandular odontogenic cyst (GOC) is a rare lesion with a frequency rate of only 0.012% to 1.3% of all the jaw cysts, which was in concurrence with the present study (1.25%).[18] GOC should be differentiated from lateral periodontal cyst (LPC), botryoidodontogenic cyst (polycystic variant of LPC) and low-grade mucoepidermoid carcinoma due to overlapping histological features.

The calcifying odontogenic cysts (COCs) are characterized histologically by the presence of focal accumulation of ghost cells in the lining epithelium, which often calcify (Figure 4). They constitute about 0.3–0.8% of all OCs(5), however in the present study the prevalence was comparatively higher (2.61%).

The prevalence of NOCs was about 4.02%, which was relatively higher than in Turkey (1.3%) and much lower when compared to studies from Nairobi (42.8%) and Brazil (7.1%).[8,9,19] In the present study, dermoid cysts (1.51%) were the most common followed by nasopalatine duct cysts (NDC) (0.75%) and aneurysmal bone cysts (ABC) (0.75%). Literature shows that NDC were the most common NOC as seen in Nairobi (26.3%) and Turkey (1.5% and 3.2%).[9,19,20] However, in a Brazilian study, epidermoid cysts (3.3%) were more prevalent. The NDC arises from the epithelial remnants of the nasoplatine duct, which connects the nasal cavity with the anterior maxilla in a developing fetus. The proliferation of epithelial remnants resulting in cyst formation is either spontaneous or following trauma, infection or mucous retention. Both dermoid and epidermoid cysts are slow growing developmental cysts arising from ectodermal tissue and about 7% of these cysts occur in the head and neck region with 1.6% in the oral cavity.[19] The occurrence of ABCs is quite rare in the jaws as it has a predilection for bones with a high venous and marrow content.[19] With only 3 cases in the current study, it shows a mandibular predominance and debatable etiology, arising either due to trauma, local change in hemodynamics or degeneration of a pre-existing lesion in the area. Yet another pseudocyst, mucous extravasation cyst, was not included in this study as it is primarily a salivary gland associated soft tissue cyst.

The distribution of jaw cysts by age showed that about 30% of the overall cysts occur in the third decade, followed by the second and fourth decades. The same pattern was noted for most of the OCs except COC (2nd and 6th decades) and GOC (2nd decade). About 47% of DC occurred in the first decade; however it peaked only in the third decade probably due delayed presentation of symptoms. Literature shows that, developmental OCs occur predominantly in early years of life as the odontogenic rests are more active and the inflammatory cysts occur later in adult life.[10] Most of the NOCs in this study also occurred in the 1st and 2nd decades of life.

About 60% of the jaw cysts occurred in males, which was in accordance with various reported studies.[4,21] Two possible reasons for that could be that men are more susceptible to trauma and have poor oral hygiene than women, both favorable for cyst formation.[9]

54% of jaw cysts occurred in the mandible, which was similar to studies in Busan (56%) and Brazil (56.6%).[8,21] The left side (45.2%) was more commonly involved than the right side (40.7%). A previous study conducted on South Indian population showed a prevalence of 43.3% OCs on the right side and 40.3% on the left side.[10] Another study conducted on Turkish population found no significant difference between right and left sides. It is therefore noteworthy that side should be considered as random occurrence rather than an attribute to a specific cause.

Residual cysts (60%), radicular cysts (73.7%), and dermoid cyst (83.3%) were noted with a greater frequency in the maxilla. About 70% of COCs were observed in the mandible. Though anterior mandible is the most common site for GOC, our results were contradictory.[5]

Aspiration of a cyst is the norm for initial diagnosis of all cysts. In this study, 8.3% cases yielded cystic content on aspiration including DCs, OKCs, OOCs, RCs, residual cyst, infected cysts and retention cyst. The nature of the contents range from clear to colored fluid, and liquid to creamy or cheesy substance. Cheesy white keratin was aspirated from 12%of the OKCs,a rate which was much lower than a study by Mohajerani et al. This could be attributed to the fact that the cyst could have been infected leading to loss of epithelium and lack of keratin production, or the cystic content might have emptied due to the presence of a sinus.[14]

46.2% DCs, 42.9% OOCs, 16.7% OKCs and 10%COCs were associated with an impacted tooth, third molar being the most common. Although all DCs are expected to be associated with impacted tooth, we were limited by the information provided by the clinicians and hence did not have a clear record for all cases pertaining to the association of impacted tooth. The frequency of association of impacted tooth with OKCs (6%) and OOCs (1.7%) was much lower in a study by Mohammad et al as compared to the present study.[22]

13.2% infected cysts, 15.4% DCs, 9.3% RCs, 6.1% OKCs, 19.0% OOCs, 40% residual cysts, 60% GOCs exhibited cholesterol clefts in this study. Although cholesterol crystals are often associated with OCs, they are no longer recognized as a characteristic feature. (Figure 5) Studies suggest that the origin might be from disintegrating erythrocytes, degenerating epithelial cells, foamy macrophages or associated with inflammatory reaction.[23]

Keratinisation may be present focally in about one third of OCs making it a diagnostic pitfall due to the overlapping of histological features. It is relatively common in cysts with low inflammation probably due to an adaptive change to environmental factors within the cyst. The exact factors responsible for keratinization is vaguely understood though mechanical stress, radiation, vitamin A deficiency, lack of proper nutrition and oxygenation have been mentioned to affect keratinization.[24] In this study, 68.1% of cases showed non-keratinized lining epithelium excepting OKCs and OOCs which showed obvious keratinization and few DCs, RCs, residual, paradental, eruption, infected OCs, nasopalatine and dermoid cysts with focal areas of keratinization. Literature shows that recurrent cases of OKCs consisted of parakeratinized lining epithelium and satellite cysts. Even in our study, 80.3% of cases exhibited parakeratinization with two cases showing satellite cysts. The aggressive nature could be attributed to the presence of MMPs and ILα-1.[25]

A mere 5 cases showed recurrence (3OKCs,2 RCs). The RCs showed recurrence after 4 and 6 years due to inadequate removal of the primary lesion. However, recurrences of OKCs were seen after an interval of 6 and 10 years. Earlier studies have reported that the recurrence rate of OKCs range from 5%-62% and the disparity could be due to the characteristics of the lesion and the treatment done. The high recurrence is due to the presence of thin friable cystic lining epithelium, satellite cysts remaining after removal of the primary cyst and active budding of basal layer of lining epithelium.[26,27] Factors such as multilocularity, frequent deep in foldings of the cystic epithelium, large size and cortical bone perforation were also found to be associated with high recurrence rate.[28] These factors were not analyzed in the present study.

Nevoid Basal Cell Carcinoma Syndrome (NBCCS), a rare autosomal dominant disorder caused due to variants in PTCH1, PTCH2 or SUFU genes, is most commonly associated with multiple OKCs.[29] In our study 2 cases of NBCCS were recorded in female patients at a younger age (second decade) who presented with multiple OKCs involving both the jaws and a hereditary pattern was identified. Thorough screening of patients with multiple OKCs is thus mandatory in order to rule out syndromic cases because multiple OKCs are usually the first manifestation in NBCCS while other symptoms like basal cell carcinoma can develop later. Karhade et al in their study observed that one third of children diagnosed with OKCs have an undiagnosed NBCCS and about 44% of these patients present with a single OKC.[30] Further, the risk of developing recurrence increases by 3.4 times in patients with NBCCS irrespective of the treatment method.[26]

Interestingly, the present study reveals an almost equal prevalence of developmental and inflammatory cysts. This near-equal prevalence could be attributed to the institutional setup of the study, with improved access to healthcare, which facilitated the incidental finding of developmental cysts through routine radiographic examinations. However, people from certain sections of society with limited oral health awareness do not report to the dentists unless there are evident clinical symptoms, leading to persistent dental caries and pulpal pathology, resulting in inflammatory cysts. Similar patterns have been reported in other South Indian studies, where variations in cyst prevalence were linked to diagnostic accessibility, socioeconomic diversity, and regional oral disease burden.[31,32]

There are a few limitations in the present study. The results of the present study cannot be generalized to the entire regional population as it is localized to only one dental institution. Details regarding the radiographic, imaging features and treatment done were not included in the current study as the primary focus was on the demographic, clinical, radiographic, and histopathological features. The follow up of patients was not possible in certain cases. Hence the recurrence rates and malignant transformation rates could not be determined with certainty.

Conclusion

This institutional study has compiled information regarding the demographic and biological behavior of jaw cysts for a period of 20 years. The most prevalent type of OC encountered in this study was RC followed by OKCs and DCs. Among NOCs, dermoid cysts were most prevalent. OKCs had a marginally higher prevalence than DCs which was contrary to many previous reports. The prevalence rate emphasizes the need to differentiate it from other similar OCs and tumors to avoid treatment procedures that may be too little or too extensive. Most of the jaw cysts were detected in the third decade of life. Two cases of multiple OKCs associated with NBCCS and satellite cysts were reported in this study highlighting the need for regular follow up to detect recurrences, especially in

case of OKCs. Thus, a thorough clinical, familial and genetic assessment of patients is pivotal to rule out syndromic from non-syndromic cases. Special attention should be given to children presenting with solitary OKCs for early detection of association with NBCCS. Knowledge of the biological behavior and clinicopathological features of jaw cysts is thus extremely essential in early diagnosis, management and better prognosis of these cystic lesions.

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