Odontogenic Cysts and Tumors of Maxilla and Maxillary Sinus (A Clinicopathological Analysis)

Shahad A. Waheed[(1)], Taghreed F. Zaidan[(2)], Bashar H. Abdullah[(3)]

https://doi.org/10.26477/jbcd.v33i4.3018

ABSTRACT

Background: Knowledge about the prevalence and distribution of pathologies in a particular location is important when a differential diagnosis is being formulated. The aim of this study was to describe the prevalence and the clinicopathological features of odontogenic cysts and tumors affecting the maxilla and to discuss the unusual presentation of those lesions within the maxillary sinus.

Materials and Methods: A multicenter retrospective analysis was performed on pathology archives of patients who were diagnosed with maxillary odontogenic cysts and tumors from 2010 to 2020. Data were collected with respect to age, gender and location.

Result: A total of 384 cases was identified, 320 (83.3%) cases were diagnosed as odontogenic cysts and 64 (16.6%) as odontogenic tumors. The mean age was 30.5 years with a standard deviation of 16.2 years. Male patients were more commonly affected (n=220, 57.3%). Radicular cyst was the most common cyst (n=205, 64.1%), while the most common tumor was odontoma (n=14, 21.9%) and dentigerous cyst was the most common lesion to present within the maxillary sinus.

Conclusion: This study indicates that there are some geographic similarities and differences in regard to distribution of odontogenic cysts and tumors in the maxilla and it raises awareness of their presentation within the maxillary sinus especially if there is an association with an ectopic or adjacent impacted tooth.

Keywords: Odontogenic cysts, odontogenic tumors, maxillary sinus, maxilla. **(Received: 2/10/2021, Accepted: 31/10/2021)**

INTRODUCTION

The microscopic features of jaw bones are similar to any other bones in the body. Their uniqueness is derived from the fact that they enclose the odontogenic apparatus, a structure that gives rise to a variety of diseases: developmental, inflammatory and neoplastic.[(1)] Odontogenic cysts are the most common jaw lesions and can be either inflammatory or developmental in origin.[(2)] On the other hand, odontogenic tumors (OGTS) are rare, accounting for less than one percent of all oral tumors. These tumors might pose a diagnostic challenge for both clinicians and pathologists due to their rarity and overlapping histopathological features.[(3)] The anatomic location of the maxillary sinus within the body of maxilla makes it vulnerable to the same conditions that affect maxillary bone.

Odontogenic lesions of the maxillary sinus are scarce in literature limited mostly to case reports or series. [(4,5,6)]

Most studies investigated the prevalence and distribution of those lesions in both jaws, no study was found to focus entirely on the maxilla and data.

(1) Master Student, Department of Oral Diagnosis, College of Dentistry, University of Baghdad.
(2) Professor, Department of Oral Diagnosis, College of Dentistry, University of Baghdad.
(3) Professor, Department of Oral Diagnosis, College of Dentistry, University of Baghdad.
Corresponding author, Shahadwaheed@yahoo.com

MATERIALS AND METHODS

Data for this retrospective study were collected from the filing systems and pathology archives of nine histopathology laboratories in Baghdad city during the period from 2010 – 2020. The inclusion criteria were histopathology reports for patients diagnosed with an odontogenic cyst or odontogenic tumor affecting the maxilla or maxillary sinus**.** Duplicate reports and recurrences of previously diagnosed or treated lesions were excluded.

In relation to the anatomic site of the lesions, two main sites were identified (maxilla and maxillary sinus). Further maxillary lesions were divided into three segments based on the clinical and radiographic findings; anterior maxilla extends from right canine to left canine. Posterior maxilla extends from the first premolar to maxillary tuberosity, and the palate.

Histological diagnoses for the lesions were classified into categories according to the most recent World Health Organization (WHO) Classification of head and neck tumors 2017.[(7)] Cysts were classified according to origin into inflammatory and developmental while odontogenic tumors into epithelial tumors, mixed epithelial and mesenchymal tumors, and mesenchymal tumors. OGTS were also divided into two types; benign and malignant tumors. A histopathology examination was performed whenever there was a doubt about the diagnosis using available histologic slides stained with hematoxylin and eosin (H&E) by an oral pathologist (B.A).

The raw collected data were organized and subdivided into categories using Microsoft Excel 2010 sheets. No

formal statistical tests were done; only descriptive analysis was performed using Statistical Package for Social Sciences (SPSS) version 23.

RESULTS

A total of 7384 histopathology reports of oral and maxillofacial lesions was found, 1064 (14.4) of cases were identified in the maxilla, of those 384 (36.09%) cases in the odontogenic cysts and tumors group; 320 (30.07%) odontogenic cysts and 64 (6.01%) odontogenic tumors. The mean age was 30.5 years with a SD of 16.2 years. Male patients were affected more (n=220, 57.3%) than females (n=164, 42.7%).

Odontogenic cysts:

338 cases were identified in the cysts category; 18 (1.6%) cases were non-odontogenic cysts and 320 (29.5%) odontogenic cysts. Males were affected more (n=187, 58.4%) than females. The mean age was 30.48 years. Most cysts were inflammatory in origin (n=211, 65.9%), while developmental cysts represent (n=106, 33.12%) and 3 cases were not otherwise specified (0.9%). The most common diagnosis was radicular cyst (n=205) followed by dentigerous cyst and keratocyst. **Table 1** demonstrates the frequency and distribution of different odontogenic cysts according to gender and age.

Table 1: The frequency and distribution of odontogenic cysts according to gender and age.

Diagnosis	Frequency		Gender			Age (years)			
	N	%	M	F	M:F	Mean	SD	Min	Max
Radicular cyst	205	64.1	113	92	1.2	32.3	14.65	8	80
Dentigerous cyst	49	15.3	38	11	3.45	23	17.54	6	61
Keratocyst	45	14.1	26	19	1.36	29.08	15.48	10	85
Calcifying odontogenic cyst	8	2.5	4	4	1	39.37	23.07	9	64
Residual cyst	6	1.9	2	4	0.5	43.66	15.27	27	65
Odontogenic cyst not otherwise specified	3	0.9	1	2	0.5	24.66	5.507	19	30
Eruption cyst	1	0.3	1	0		3	.	3	3
Gingival cyst of newborn	1	0.3	0	1		1 week	.	1 week	1 week
Lateral periodontal cyst	1	0.3	1	0		19	.	19	19
Orthokeratinized cyst	1	0.3	1	0		15	.	15	15
Total	320	100	187	133	1.4	30.48	15.97	1 week	85

Odontogenic tumors:

Males were affected slightly more (n= 33, 51.6%) than females. The mean age was 31 years. Most tumors were of epithelial origin (n=25, 39.1%) while mesenchymal origin represent (n=23, 35.9%) and mixed origin was (n=16, 25%). Almost, all were benign tumors (n=62, 96.9%) except for two malignant cases (n=2, 3.1%). The most common diagnosis was Odontoma (n=14) followed by both ameloblastoma and odontogenic myxoma (n=12, 18.8%), as shown in **(Table 2)**.

Site:

The precise site was reported in 196 cases with most lesions affecting the anterior maxilla (n=122, 62.2%), while posterior maxilla was (n=38, 19.38%), palate (n=16, 8.16%). Few cases were reported in maxillary sinus (n=20, 10.2%) (n=8 dentigerous cysts, n=6 radicular cysts, n=2 keratocyst, n=1 Orthokeratinized cyst, n=1 Adenomatoid odontogenic tumor, and n=2 myxoma).

Table 2: The frequency and distribution of odontogenic tumors according to gender and age

Diagnosis	Frequency		Gender			Age (years)			
	N	%	M	F	M:F	Mean	SD	Min	Max
Odontoma	14	21.9	5	9	0.55	19.4	11.58	8	45
Ameloblastoma	12	18.8	10	2	5	51.83	17.19	30	85
Myxoma	12	18.8	5	7	0.7	28.25	11.09	15	45
Odontogenic fibroma	10	15.6	5	5	1	34.5	13.8	14	54
Pindborg tumor	5	7.8	2	3	0.66	24.2	11.4	13	43
Adenomatoid odontogenic tumor	4	6.3	2	2	1	19.25	5.67	13	25
Unicystic ameloblastoma	2	3.1	0	2		21.5	4.9	18	25
Ameloblastic fibroma	1	1.6	0	1		12	.	12	12
Cementoblastoma	1	1.6	1	0		25	.	25	25
Ghost cell carcinoma	1	1.6	1	0		48	.	48	48
Malignant ameloblastoma	1	1.6	1	0		74	.	74	74
Primordial odontogenic tumor	1	1.6	1	0		14	.	14	14
Total	64	100	33	31	1.06	31.1	17.7	8	85

DISCUSSION

Many previous studies analyzed those lesions in both jaws; however, no study focus entirely on the maxilla; because of that to facilitate comparison, some of these studies were reviewed and information regarding maxillary odontogenic lesions was extracted and displayed in **(Table 3)** and **(Table 4)**

In regard to odontogenic cysts, most of the cysts were of inflammatory origin and radicular cyst was the most common diagnosis which is in accordance with all previous studies in (Table 3).[8, 9,10,11,12]

The second most common odontogenic cyst was dentigerous cyst (15.3%) which is similar to other studies.[8, 9,10,12]. But it disagrees with Del Corso et al.[11] These cysts are developed in association with unerupted teeth mostly maxillary canine and third molars with predilection for patients in the second decade of life.[9]

Odontogenic keratocyst was the third most common cyst in the maxilla which is similar with Izgi et al., and Kambalimath et al studies.[8,10] This study followed the 2017 WHO classification that reclassified keratocyst from benign tumor to a cyst; in contrast to some previous published studies that followed the old classification.[9,14] This study showed a close prevalence between dentigerous cyst and keratocyst (15.3%, 14.1%) respectively. This could be explained by the fact that most dentigerous cysts are small, symptomless, discovered accidently on radiographs and clinicians may not request a histopathology examination relying only on clinical diagnosis. Keratocyst usually grow to larger sizes, produce more obvious clinical symptoms and require further surgical intervention. Regardless the size or appearance, any cystic lesions of the jaw should raise the suspicion of more aggressive odontogenic lesion such as keratocyst, ameloblastoma, pindborg tumor especially due to the fact that many of them can be associated with impacted teeth and resemble the radiographic appearance of dentigerous cyst.[13]

Table 3: The characteristics of maxillary odontogenic cysts in comparison studies

Study (author, year)	Country	Number of cases in the maxilla	The most common cysts (N)	Time period (years)
(Izgi et al., 2021).[8]	Turkey	165	Radicular cyst (95) Dentigerous cyst (22) Keratocyst (13)	(2008 – 2018)
(Villasis-Sarmiento et al., 2017).[9]	Mexico	337	Radicular cyst (215) Dentigerous cyst (110) Residual cyst (5)	(2000 – 2013)
(Kambalimath et al., 2014).[10]	India	76	Radicular cyst (57) Dentigerous cyst (7) Keratocyst (3)	(2001 – 2011)
(Del Corso et al., 2014).[11]	Italia	374	Radicular cyst (205) Keratocyst (48) Dentigerous cyst (21)	(1992 – 2012)
(Bataineh et al., 2004).[12]	Jordan	313	Radicular cyst (149) Dentigerous cyst (67) Residual cyst (59) LPC (34)	(1989 – 2001)

With regard to odontogenic tumors, almost all tumors were benign with only two malignant cases such finding is in accordance with all studies in (table 2).[14,15,16,17] Most tumors were of epithelial origin which is similar with Nalabolu et al.[14]

The most common tumor in this study was odontoma that is similar to previous studies.[15,16,17] Odontoma represents a hamartous lesion rather than a true neoplasm and is believed to be the most common odontogenic tumor like lesion; however, it is usually asymptomatic and diagnosed accidentally on radiograph. This could be the reason for the small number of cases in this study since patients do not seek treatment until obvious symptoms appear.[14] Geographic, genetic and environmental factors can also affect the prevalence of odontomes.[14] Similar to other studies, this lesion showed a high prevalence in the second decade of life.[14,16]

Ameloblastoma was the second most common tumor which was in agreement with Taghavi et al.[17] and with other studies. [15,16] If keratocyst was excluded, a male predilection was shown with male to female ratio of 5:1 which disagrees with Zwahlen and Grätz literature review,[18] that reported no gender predilection. Although maxillary ameloblastomas are rare compared to those of the mandible; they still ranked high among OGTS in this study. This tumor is aggressive, usually showing extensive extension into the adjacent structures such as nasal cavity and maxillary sinus,[19] and can clinically mimic a sinonasal malignancy. Ameloblastoma can arise primary from the surface epithelium of the maxillary sinus; however, such a diagnosis is rare and can't be made without excluding an extension from the surrounding maxillary bone first.[20]

Odontogenic myxoma is a benign mesenchymal tumor. It is reported as the third most common odontogenic neoplasm of jaw after odontoma and ameloblastoma.[17] In this study, it showed a similar prevalence to ameloblastoma (18.8%) and two cases were reported initially as sinus lesions. This lesion usually arises within the maxillary bone and invades the adjacent sinus; however sinonasal counterparts have been rarely reported in the literature.[21] There are different theories regarding the etiology of this lesion in sinus. Myxomas may arise from modified fibroblasts associated with tooth germ in which these altered cells can hyper secrete mucin.[22] The association between myxoma and impacted teeth has provided support for this theory. However, evidence exists that argues against this odontogenic theory a study by Slootweg and Wittkampf.[23] demonstrated that the extracellular matrix of myxomas differs histologically from the extracellular matrix of dental and periodontal tissues. Also, the presence of reported cases of myxomas in remote sites from odontogenic tissues such as sphenoid sinus.[24]

This study also reported other rare lesions in less numbers such as calcifying odontogenic cyst (2.5%), Pindborg tumor (7.8%), unicystic ameloblastoma (3.1%), ameloblastic fibroma (1.6%), cementoblastoma (1.6%) and one case of the newly described entity primordial odontogenic tumor that

was published previously by the author (B.A.) as the first Iraqi case report.[25]

The anterior maxilla was more commonly affected in both cysts and tumors; a finding that is similar with Izgi et al.[8]

Table 4: The characteristics of maxillary odontogenic tumors in comparison studies

Study (author, year)	Country	Number of cases in the maxilla	The most common tumors (N)	Time period (years)
(Nalabolu et al., 2017).[14]	India	45	Keratocyst (17) Ameloblastoma (16) Odontoma (6) Adenomatoid odontogenic tumor (6)	(2002 – 2014)
(AlSheddi et al., 2015).[15]	Saudi Arabia	52	Odontoma (15) Keratocyst (14) Ameloblastoma (6)	(1984 – 2010)
(Avelar et al., 2008).[16]	Brazil	78	odontoma (31) keratocyst (17) Ameloblastoma (9) myxoma (8)	(1992 – 2007)
(Taghavi et al., 2013).[17]	Iran	50	Odontoma (16) Ameloblastoma (13) Myxoma (11)	(2000 – 2010)

Maxillary sinus is vulnerable to diseases of odontogenic origin due to its proximity with developing teeth and root apices. Diagnosis of such lesions in this location is challenging as routine radiographs do not offer characteristic features and show overlaps of various adjacent structures.[26] Most odontogenic cysts and tumors in this location are associated with an ectopic tooth inside the sinus with the commonest being dentigerous cyst.[27,28] which is similar to this study findings. Many theories exist regard the etiology of those ectopic teeth including trauma, developmental and pathologic conditions. The most likely scenario is the presence of impacted tooth (mostly canine or molars) in close anatomic relation to the sinus wall and as the cyst grows and enlarges it displacing the tooth inside the sinus space.[4] Other odontogenic cysts had also been reported infrequently to involve the maxillary sinus. Radicular cyst can grow to large sizes, breach the sinus wall and occupy the entire sinus space.[6] Keratocysts can also involve the maxillary sinus with a controversy regarding its origin; it might arise from the entrapment of dental lamina within the sinus during normal development due to the close anatomic relation,[29] or from normal expansion of an adjacent intraosseous cyst, or in relation to an ectopic tooth inside the sinus.[5]

CONCLUSION

There are some geographic similarities and differences with regard to the prevalence and distribution of odontogenic cysts and tumors of maxilla. The presence of these lesions within the maxillary sinus is uncommon and different theories exist in the literature regarding their origin; nevertheless, they should be included in the differential diagnosis of sinus lesions especially if there was an association with an ectopic or adjacent impacted tooth.

Conflict of interest: None.

REFERENCES

1. Jonathan B, Mandible and Maxilla, in: Goldblum JR, Lamps LW, McKenney JK, Myers JL, Rosai and Ackerman's Surgical Pathology, Elsevier Health Sciences, Philadelphia, 2017: 212-234.
2. Jones AV, Craig GT, Franklin CD. Range and demographics of odontogenic cysts diagnosed in a UK population over a 30- year period. J Oral Pathol Med. 2006;35:500-7.
3. Neville BW, Damm DD, Allen CM, *et al*. Oral and maxillofacial pathology, fourth ed., Elsevier Health Sciences, Philadelphia, 2015.
4. Buyukkurt MC, Omezli MM, Miloglu O. Dentigerous cyst associated with an ectopic tooth in the maxillary sinus: a report of 3 cases and review of the literature. Oral Surg Oral Med Oral Pathol Oral Radiol Endod. 2010;109:67-71.
5. Sheethal HS, Rao K, Umadevi HS, *et al*. Odontogenic keratocyst arising in the maxillary sinus: A rare case report. J Oral Maxillofac Pathol. 2019; 23:74.
6. Kumar N, Dadhich A. Unusually large radicular cyst presenting in the maxillary sinus. BMJ Case Rep. 2020; 13.

7. El-Naggar AK, Chan JK, Grandis JR, *et al*, WHO classification of head and neck tumours. International Agency for Research on Cancer (IARC), France, 2017.
8. Izgi E, Mollaoglu N, Simsek MB. Prevalence of odontogenic cysts and tumors on turkish sample according to latest classification of world health organization: A 10-year retrospective study. Niger J Clin Pract. 2021;24:355.
9. Villasis-Sarmiento L, Portilla-Robertson J, Melendez-Ocampo A, *et al*. Prevalence and distribution of odontogenic cysts in a Mexican sample. A 753 cases study. J Clin Exp Dent. 2017; 9:e531.
10. Kambalimath DH, Kambalimath HV, Agrawal SM, *et al*. Prevalence and distribution of odontogenic cyst in Indian population: a 10 year retrospective study. J Clin Exp Dent. 2014;13:10-5
11. Del Corso G, Righi A, Bombardi M, *et al*. Jaw cysts diagnosed in an Italian population over a 20-year period. Int J Surg Pathol. 2014;22:699-706.
12. Bataineh AB, Ma'amon AR, Qudah MA. The prevalence of inflammatory and developmental odontogenic cysts in a Jordanian population: a clinicopathologic study. Quintessence Int. 2004;35:815-9.
13. Müller S. Developmental Odontogenic Lesions Associated with the Crown of an Impacted Tooth: A Guide to the Distinct Histologic Features Required for Classification. Head Neck Pathol. 2021;15:107-12.
14. Nalabolu GR, Mohiddin A, Hiremath SK, *et al*. Epidemiological study of odontogenic tumours: An institutional experience. J Infect Public Health. 2017;10:324-30.
15. AlSheddi MA, AlSenani MA, AlDosari AW. Odontogenic tumors: analysis of 188 cases from Saudi Arabia. Ann Saudi Med. 2015;35:146-50.
16. Avelar RL, Antunes AA, Santos TD, *et al*. Odontogenic tumors: clinical and pathology study of 238 cases. Braz J Otorhinolaryngol. 2008;74:668-73.
17. Taghavi N, Rajabi M, Mehrdad L, *et al*. 0-year retrospective study on odontogenic tumors in Iran. Indian J Dent Res. 2013;24:220.
18. Zwahlen RA, Grätz KW. Maxillary ameloblastomas: a review of the literature and of a 15-year database. J Craniomaxillofac Surg. 2002;30:273-9.
19. Evangelou Z, Zarachi A, Dumollard JM, *et al*. Maxillary ameloblastoma: a review with clinical, histological and prognostic data of a rare tumor. In Vivo. 2020;34:2249-58.
20. Schafer DR, Thompson LD, Smith BC, *et al*. Primary ameloblastoma of the sinonasal tract: a clinicopathologic study of 24 cases. Cancer. 1998;82:667-74.
21. Mewar P, González-Torres KE, Jacks TM, *et al*. Sinonasal myxoma: a distinct lesion of infants. Head Neck pathol. 2020;14:212-9.
22. White DK, Chen SY, Mohnac AM, *et al*. Odontogenic myxoma: a clinical and ultrastructural study. Oral Surg Oral Med Oral Pathol. 1975;39:901-17.
23. Slootweg PJ, Wittkampf AR. Myxoma of the jaws: an analysis of 15 cases. J Maxillofac Surg. 1986;14:46-52.
24. Moore BA, Wine T, Burkey BB, *et al*. Sphenoid sinus myxoma: case report and literature review. Ochsner J. 2008;8:166-71.
25. Bashar A, Venus M, Omar M, *et al*. Primordial odontogenic tumor: A case report with a novel clinical presentation. J Oral Maxillofac Surg Med Pathol. 2021;33:239-42.
26. Mehra P, Jeong D. Maxillary sinusitis of odontogenic origin. Curr Allergy Asthma Rep. 2009;9:238-43.
27. Pierse JE, Stern A. Benign cysts and tumors of the paranasal sinuses. Oral Maxillofac Surg Clin North Am. 2012;24:249-64.
28. Press SG. Odontogenic tumors of the maxillary sinus. Curr Opin Otolaryngol Head Neck Surg. 2008;16:47-54.
29. Cioffi GA, Terezhalmy GT, Del Balso AM. Odontogenic keratocyst of the maxillary sinus. Oral Surg Oral Med Oral Pathol. 1987;64:648-51.

المستخلص:

الخلفية: المعرفة حول انتشار وتوزيع الأمراض في موقع معين أمر مهم عند صياغة التشخيص التفريقي. كان الهدف من هذه الدراسة هو وصف الانتشار والسمات الإكلينيكية للأكياس والأورام السنية التي تصيب الفك العلوي ومناقشة الظهور غير المعتاد لتلك الآفات داخل الجيوب الأنفية الفكية.المواد والطرق: تم إجراء تحليل رجعي متعدد المراكز على تقارير التشخيص النسيجي للمرضى الذين تم تشخيصهم بالأكياس والأورام السنية في الفك العلوي من عام 2010 إلى عام 2020. تم جمع البيانات فيما يتعلق بالعمر والجنس والموقع.

النتيجة: تم تحديد 384 حالة، 320 (83.3٪) حالة كيسات سنية و 64 (16.6٪) أورام سنية. كان متوسط العمر 30.5 سنة مع انحراف معياري 16.2 سنة. المرضى الذكور كانوا اكثر عرضة للاصابة (220 ، 57.3٪). كان الكيس الجذري Radicular cyst هو الكيس الأكثر شيوعًا (العدد = 205 ، 64.1٪) ، بينما كان الورم الأكثر شيوعًا هو الورم السني odontoma (العدد = 14 ، 21.9٪) وكان كيس الأسنان Dentigerous cystهو الآفة الأكثر شيوعًا الموجودة داخل الجيب الفكي.

الاستنتاج: تشير هذه الدراسة إلى أن هناك بعض أوجه التشابه والاختلاف الجغرافي فيما يتعلق بتوزيع الاكياس والأورام السنية في الفك العلوي وزيادة الوعي بمظهرها داخل الجيوب الأنفية الفكية خاصةً إذا كان هناك ارتباط مع سن مجاور مطمور.