



<u>Original article</u>

Clinico-pathological analysis of Odontogenic Tumors over 28-year-period in Benghazi, Libya

Samar S. Saleh Hamed, Nagla S. El-Hasi

Department of oral pathology, oral medicine, oral diagnosis and Radiology, Faculty of dentistry, University of Benghazi, Libya.

ABSTRACT:

Background: Odontogenic tumors (OTs) are rare lesions that unique to the jaws constituting about < 1% of all oral tumors. They are a complex group of heterogeneous behavior that range from tumor like lesions, benign tumors to malignant neoplasms with potential to metastases.

Aim of study: To describe the relative incidence of odontogenic tumors (According to World Health Organization classification 2022) at Oral Pathology Department in Benghazi and compare the finding with the literatures.

Methods: A retrospective study of 106 OTs was documented for the demographic data. Statistical analysis was carried out by software *SPSS*.

Results: OTs constituted 1.2% of all diagnosed oral lesions with 97% of them were benign tumors. Ameloblastoma was the most common type (37.7%) followed by odontomas (24.5%). The peak incidence was around the third decade with male: female ratio 1:1.12. mandible was the most common site (64%).

Conclusion: OTs are relatively uncommon lesions among our sample that is similar to other literatures with some variations.

Keywords: Odontogenic tumors, retrospective study, incidence, World Health Organization classification, Benghazi.

Corresponding author:

Nagla El hasi: Department of oral medicine, oral pathology, oral diagnosis and Radiology, Benghazi, Libya. E.mail: <u>nagla.saed@uob.edu.ly</u>

INTRODUCTION:

Odontogenic tumors (OTs) are uncommon lesions that are unique to the gnathic bones. They are accounted for < 1% of all oral tumors.^{1, 2} From a biological point of view, the majority of these lesions are benign neoplasms and some exhibit malignant behavior with metastatic capacity, while the rest represent as hamartomas lesions.¹

Odontogenic tumors can be originated either from the odontogenic epithelium such as dental lamina, reduced enamel epithelium, rests of Serres or rests of Malassez, or from odontogenic mesenchymal tissues such as dental follicle, dental papilla, pulp or periodontal ligament, or from both.³ Several classification schemes based on the origin of the tumor have been devised.⁴ The first histological typing of OTs "histological typing of OTs" was published by the World Health Organization (WHO) in 1971 and was reviewed and updated in 1992 and 2005.⁵

In 2017, the WHO updated the classification to reclassify the "keratocystic odontogenic tumor" and "calcifying cystic odontogenic tumor" as odontogenic cysts. Furthermore, primordial odontogenic tumors were included as mixed tumor and cemento-ossifying fibroma as mesenchymal tumors. In addition, the fibro-odontoma was included as a benign variant of the odontoma.⁶ Recently, the 5th edition of the World Health Organization classification was available online (Table 1). The most important update was adding Adenoid ameloblastoma as a new entity to the epithelial odontogenic neoplasms.⁷

Retrospective studies have been conducted around the world that reported variable geographic distribution. These variations are attributed to the high cultural and genetic diversity.^{8, 9} Knowledge of the clinical presentation of OTs and their epidemiology are necessary to understand the characteristics and behaviour of these lesions and can be valuable in developing a clinical differential diagnosis.^{6, 10} Libyan Journal of Dentistry (LJD) Volume 7, Issue 1, 2023

This study aims to investigate the frequency and distribution of histologically diagnosed odontogenic tumors at the department of oral pathology in Benghazi, and compare data with the literatures.

Benign epithelial odontogenic tumors Benign mesenchymal odontogenic tumors Odontogenic fibroma Adenomatoid odontogenic tumor Squamous odontogenic tumor Cementoblastoma Calcifying epithelial odontogenic tumor Cemento-ossifying fibroma Ameloblastoma, unicystic Odontogenic myxoma Ameloblastoma, extraosseous/peripheral Ameloblastoma, conventional Adenoid ameloblastoma Metastasizing ameloblastoma Benign mixed epithelial & mesenchymal Malignant odontogenic tumors odontogenic tumors Sclerosing odontogenic carcinoma Odontoma Ameloblastic carcinoma Primordial odontogenic tumor Clear cell odontogenic carcinoma Ameloblastic fibroma Ghost cell odontogenic carcinoma Dentinogenic ghost cell tumor Primary intraosseous carcinoma, NOS Odontogenic carcinosarcoma Odontogenic sarcomas

Table 1: 2022 WHO classification of odontogenic tumors

METHODS:

The archival records of the oral pathology department- University of Benghazi were revised retrospectively from January 1990 to December 2018. A total of 106 cases were diagnosed as OTs during this period. The histopathological diagnosis was based on 2022 WHO histopathologic classification.

All collected cases were reviewed and analyzed for the demographic features including, age of patient, gender, tumor location, and histopathological type. This study was taken out with permission from the institutional authorities. Statistical analysis was carried out using SPSS.

RESULTS:

A total of 106 OTs cases were diagnosed from 1990 to 2018 which constituted 1.2 % of all registered biopsies (8995 diagnostic samples). Of the cases 103 (97%) were benign OTs while only 3 cases were diagnosed as malignant OTs (Table 2).

Ameloblastoma was the most frequently diagnosed odontogenic tumor 40 (37.7 %) followed by odontoma 26 (24.5%) and adenomatod odontogenic tumors 10 (9.4%). Male to female ratio of all registered OTs was 1:1.12 (table2).

Diagnosis	No.	%	Male	Female	M: F
			No(%)	No(%)	Ratio
AME	40	37.7	25 (62.5)	15 (37.5)	1.6:1
ΑΟΤ	10	9.4	4 (40)	6 (60)	1:1.5
СЕОТ	5	4.7	2 (40)	3 (60)	1 : 1.5
OD					
compound OD (12)	26	24.5	10 (38.5)	16 (61.5)	1:1.6
complex OD (13)					
ОМ	7	6.6	4 (57.1)	3 (42.9)	1.3 : 1
OF	4	3.8	2 (50)	2 (50)	1:1
СВ	1	0.9	-	1 (100)	0:1
AF	2	1.9	2 (100)	-	1:0
COF	8	7.5	-	8 (100)	1:0
AC	3	2.8	1	2	1:2
Total	106	100	50 (47.2%)	56 (52.8%)	1:1.12

 Table 2: Relative frequency and gender distribution of Odontogenic Tumors 1990-2018

AME=Ameloblastoma, AOT=Adenomatoid odontogenic tumor, CEOT=Calcifying epithelial odontogenic tumor, OD=Odontoma, OM= Odontogenic myxoma, OF= Odontogenic fibroma, CB=Cementoblastoma, AF=Ameloblastic fibroma, COF= Cemento-ossfiying-fibroma, AC=Ameloblastic carcinoma.

The peak incidence of OTs was in the third decade. Patient age ranged widely between 5 -75 with a mean age of 26.51 years. Age was not reported in two cases. (table 3)

Of a total of 106 lesions in our series, the location was reported in 103 cases. The mandible showed the highest prevalence with overall 66 cases (64%) while 35 cases of OTs (34%) were identified in the maxilla (Mandible: maxilla ratio was 1.9:1). Only two cases (1.9%) were found peripherally in the gingiva. (table 4).

According to ameloblastoma, unicystic type comprised 21 cases (53.8%), multicystic variant were

17 (43.6%), with only one case (2.6%) of peripheral ameloblastoma. The histological pattern of one case was not specified (Figure 1). Male to female ratio was 1.6:1 and the mean age of occurrence was (30.88) (Tables 2- 3). Regarding the site, the posterior mandible was the most affected site (71.4%) (Figure 2). Regarding odontoma (26 cases), 13 cases were diagnosed as complex odontoma and 12 cases were identified as compound odontoma. Male to female ratio was (1:1.6) with the mean age of 17.56 years. The percentage of occurrence in the mandible and maxilla were (60%), (40%) respectively (tables 2-3 and4)

Diagnosis	Age Range	Mean Age (SD)	0-9	10-19	20-29	30-39	40-49	50-59	60-69	70-79
AME (40)	13-75	30.88±13.48	-	8	12	11	6	1	1	1
AOT (10)	10-30	17.9± 6.19	-	7	3	-	-	-	-	-
CEOT(5)	5-70	45± 27.99	1	-	-	-	-	2	-	1
OD (26)	9-40	17.56±7.67	2	15	7	-	1	-	-	-
OM (7)	11-55	24.57±14.89	-	4	1	1	-	1	-	-
OF (4)	20-56	35.5±15.17	-	-	1	2	-	1	-	-
CB (1)	00	00	-	-	-	1	-	-	-	-
AF (2)	6-15	10.50±6.36	1	1	-	-	-	-	-	-
COF (8)	9-60	32.5±16.16	1	-	2	3	-	1	1	-
AC (3)	22-50	31.67±15.89	-	-	2	-	-	1		-
Total (106)	5-75	26.65±14.53	5	35	28	18	7	7	2	2

Table 3: Age group distribution of Odontogenic tumors by life decade

Table 4: Site distribution of Odontogenic tumors 1990-2018

tumortuno	Mandible	Maxilla	Gingiva	
tumor type	No, %	No, %	No, %	
AME (39)	34 (87.2 %)	4(10.3%)	1(2.6%)	
AOT (10)	1(10%)	9(90%)	-	
CEOT (4)	-	4(100%)	-	
OD (25)	15(60%)	10(40%)	-	
OM (7)	4(57.1%)	3 (42.9%)	-	
OF (4)	1(25%)	2(50%)	1(25%)	
CB (1)	1(100%)	-	-	
COF (8)	6(75%)	2(25%)	-	
AF (2)	1(50%)	1(50%)	-	
AC (3)	3(100%)	-	-	
Total (103)	66 (64%)	35 (34%)	2 (1.9%)	



Figure (1): the frequency of histological types of ameloblastoma with gender predominance



Figure (2): distribution of anatomical sites of ameloblastoma in the jaws

DISCUSSION:

Odontogenic tumors are relatively uncommon lesions with diverse clinical and histopathological features that are derived from tooth forming tissue.^{11, 12} This study was done in Benghazi University to document the prevalence of odontogenic tumors.

The relative frequency of OTs in our sample was 1.2% of the total biopsied specimens recorded in Oral Pathology Department in Dental faculty of Benghazi University in a period between 1990 to 2018. This low

frequency is similar to those reported in other studies.^{1, 13-17} while higher incidence was conducted by some researchers.^{11, 18-21}

This data confirms that benign tumors are the most frequently seen (97%) while malignant OTs representing only (3%) that in agreement with the previous literatures.⁶, ⁸, ¹⁵, ²²⁻²⁵

We have observed predominance of OTs in female (52.8%) more than male (47.2%) with male: female ratio 1:1.12 that are corroborating in other studies.^{6, 9,}

^{11, 14, 26, 27} However, the male predilection conducted by other researchers.^{11, 18, 28, 29}

The peak incidence of OTs was in the third decades with the mean age of 26.51 years. This result was similar to that found in the other literatures.^{11, 16, 18, 21, 27, 30-33} Less mean age less than a decade was shown by some authors.^{23, 25} A remarkable preference for mandible was documented (64%) which concurs with other papers. ^{1, 6, 9, 11, 13, 15-19, 34}

Ameloblastoma was by far the most frequent tumor in this data with percentage (37.7%). This is concordance with many studies.^{6, 11, 13, 19, 26, 35} The second most common tumor was odontoma (24.5%) and this was similar to a result reported by silva et.al.⁶ However, this differ from the previous papers which considered Amloblastoma and odontogenic Keratocyst were more frequent.^{1, 5, 21, 28, 32, 36, 37} The explanation for this marked variations was the WHO 2005 classification which classified OKC as Odontogenic tumors.

Of all ameloblastoma cases, 53.8% were unicystic type, 43.6% were multicystic variant while the peripheral ameloblastoma only constituted 2.6%. these results are similar to a study done by Filipe, et al.¹⁴ Most studies showed that multicystic type was the most frequent type. ^{1, 6, 18, 26, 38}

Ameloblastoma was more common in mandible (87.2%) than maxilla (10.3%). This result was in agreement with previous studies.^{8, 21, 26, 33, 39} The peak incidence of ameloblastoma was 30.88 which is consistent with other studies.^{40, 41} The occurrence of tumors in a younger age group was observed in other reports. ^{9, 11, 15, 16, 21, 32} In this research, ameloblastoma was shown male predilection (62.5%) compared with female (37.5%) in support of recent studies.^{1, 9, 11, 15, 18, 32}

Regarding odontoma, the peak of incidence was in the second decade that seems similar to previous reports.^{15, 21, 33} Moreover, similar male to female ratio (1:1.6) was documented by chrysomali et.al.¹ Some studies reported equal distribution in the mandible and maxilla.^{1, 33} However, in this sample higher percentage was noticed in the mandible.

AOT represented the third most common tumor in our data (9.4%), similar percentage was pointed out by the literature taken out in India.⁴² while slightly lower percentage reported by sharma et. Al.⁴³

In 2017, WHO classification recognized COF as odontogenic tumor ⁶ that was the cause of missing data about it in the most previous studies. In our data, 8 cases were reported (7.5%) representing the forth common odontogenic tumor.

The odontogenic myxoma was reported as the third most common tumor by de Medeiros et.al.⁶ and the second one in other publications ^{11, 25, 31}. However, in

our sample it represented the fifth common tumor (6.6%).

In this sample, 5 and 4 cases were reported as CEOT and odontogenic fibroma respectively. the less frequent tumors were ameloblastic carcinoma (3 cases), ameloblastic fibroma (2 cases) and cementoblastoma (one case).

CONCLUSION:

With the comparison with previous study, we found some variations in the profile of incidence and prevalence of the OTs and this due to different changes and updates among WHO classification. Moreover, the geographic variations and study design also play a crucial role in the epidemiology. The present study reflects not only the differences in the distribution of OTs but also similarities among the previous population samples assessed around the world.

To sum up, our data documented the odontogenic cases in Benghazi and was compared with previous literatures. Epidemiological studies are important because they allow to know more precisely the occurrence of these lesions in the diverse population, which help to identify the groups at risk with a view of the most common clinical features related to them.

REFERENCES:

1. Chrysomali E, Leventis M, Titsinides S, Kyriakopoulos V, Sklavounou A. Odontogenic tumors. Journal of Craniofacial Surgery. 2013;24(5):1521-5.

2. El-Naggar AK, Chan JK, Grandis JR. WHO classification of head and neck tumours2017.

3. Cawson RA, Odell EW. Cawson's essentials of oral pathology and oral medicine e-book: Elsevier Health Sciences; 2017.

4. Wright JM, Tekkesin MS. Odontogenic tumors: where are we in 2017? Journal of Istanbul University Faculty of Dentistry. 2017;51(3 Suppl 1):S10.

5. Sharma PN, Ranka RK, Chaudhary MS, Gawande MN, Hande AH, Zade PF. Odontogenic tumors: A review of 93 cases in the Vidharba region of Maharashtra. Journal of Oral and Maxillofacial Pathology: JOMFP. 2020;24(1):185.

6. da Silva L-P, Pinto L-P, de Souza L-B. Clinicopathological analysis of odontogenic tumors over 22 years period: experience of a single center in northeastern Brazil. Medicina oral, patologia oral y cirugia bucal. 2018;23(6):e664.

7. Soluk-Tekkesin M, Wright JM. The World Health Organization Classification of Odontogenic Lesions: A Summary of the Changes of the 2022 (5 th) Edition. Turkish Journal of Pathology. 2022;38(2):168-84.

8. Sekerci A-E, Nazlım S, Etoz M, Denız K, Yasa Y. Odontogenic tumors: a collaborative study of 218 cases diagnosed over 12 years and comprehensive review of the literature. Medicina oral, patologia oral y cirugia bucal. 2015;20(1):e34.

9. Avelar RL, Antunes AA, Santos TdS, Andrade ESdS, Dourado E. Odontogenic tumors: clinical and pathology study of 238 cases. Revista Brasileira de Otorrinolaringologia. 2008;74:668-73.

10. Regezi JA, Sciubba JJ, Jordan RC. Oral pathology: clinical pathologic correlations: Elsevier Health Sciences; 2016.

11. Jing W, Xuan M, Lin Y, Wu L, Liu L, Zheng X, et al. Odontogenic tumours: a retrospective study of 1642 cases in a Chinese population. International journal of oral and maxillofacial surgery. 2007;36(1):20-5.

12. Philipsen HP, Reichart PA. Classification of odontogenic tumours. A historical review. Journal of oral pathology & medicine. 2006;35(9):525-9.

13. da Silva L-P, Serpa M-S, Jefferson-da-Rocha Tenório G-J, do Nascimento F, de Souza-Andrade E-S, Veras-Sobral A-P. Retrospective study of 289 odontogenic tumors in a Brazilian population. Medicina oral, patologia oral y cirugia bucal. 2016;21(3):e271.

14. Jaeger F, de Noronha MS, Silva MLV, Amaral MBF, Grossmann SdMC, Horta MCR, et al. Prevalence profile of odontogenic cysts and tumors on Brazilian sample after the reclassification of odontogenic keratocyst. Journal of Cranio-Maxillofacial Surgery. 2017;45(2):267-70.

15. Osterne RLV, de Matos Brito RG, Alves APNN, Cavalcante RB, Sousa FB. Odontogenic tumors: a 5year retrospective study in a Brazilian population and analysis of 3406 cases reported in the literature. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. 2011;111(4):474-81.

16. da-Costa DOP, Mauricio AS, de-Faria PAS, da-Silva LE, Mosqueda-Taylor A, Lourenço SQC. Odontogenic tumors: a retrospective study of four Brazilian diagnostic pathology centers. Medicina oral, patologia oral y cirugia bucal. 2012;17(3):e389.

17. Koivisto T, Bowles WR, Rohrer M. Frequency and distribution of radiolucent jaw lesions: a retrospective analysis of 9,723 cases. Journal of endodontics. 2012;38(6):729-32.

18. Luo H-Y, Li T-J. Odontogenic tumors: a study of 1309 cases in a Chinese population. Oral oncology. 2009;45(8):706-11.

19. Elarbi M, El-Gehani R, Subhashraj K, Orafi M. Orofacial tumors in Libyan children and adolescents. A

descriptive study of 213 cases. International journal of pediatric otorhinolaryngology. 2009;73(2):237-42.

20. Mamabolo M, Noffke C, Raubenheimer E. Odontogenic tumours manifesting in the first two decades of life in a rural African population sample: a 26 year retrospective analysis. Dentomaxillofacial Radiology. 2011;40(6):331-7.

21. Siriwardena B, Tennakoon T, Tilakaratne W. Relative frequency of odontogenic tumors in Sri Lanka: Analysis of 1677 cases. Pathology-Research and Practice. 2012;208(4):225-30.

22. Mosqueda-Taylor A, Ledesma-Montes C, Caballero-Sandoval S, Portilla-Robertson J, Rivera LMR-G, Meneses-García A. Odontogenic tumors in Mexico: a collaborative retrospective study of 349 cases. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. 1997;84(6):672-5.

23. Ochsenius G, Ortega A, Godoy L, Peñafiel C, Escobar E. Odontogenic tumors in Chile: a study of 362 cases. Journal of Oral Pathology & Medicine. 2002;31(7):415-20.

24. Santos JN, PEREIRA PINTO L, Figueredo CRLVd, Souza LBd. Odontogenic tumors: analysis of 127 cases. Pesquisa Odontológica Brasileira. 2001;15:308-13.

25. Fernandes AM, Duarte ECB, Pimenta FJGS, Souza LN, Santos VR, Mesquita RA, et al. Odontogenic tumors: a study of 340 cases in a Brazilian population. Journal of oral pathology & medicine. 2005;34(10):583-7.

26. Saghravanian N, Jafarzadeh H, Bashardoost N, Pahlavan N, Shirinbak I. Odontogenic tumors in an Iranian population: a 30-year evaluation. Journal of oral science. 2010;52(3):391-6.

27. Goteti SH. Odontogenic tumors: A review of 675 cases in Eastern Libya. Nigerian Journal of Surgery. 2016;22(1):37-40.

28. Sriram G, Shetty RP. Odontogenic tumors: a study of 250 cases in an Indian teaching hospital. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. 2008;105(6):e14-e21.

29. Johnson NR, Savage NW, Kazoullis S, Batstone MD. A prospective epidemiological study for odontogenic and non-odontogenic lesions of the maxilla and mandible in Queensland. Oral surgery, oral medicine, oral pathology and oral radiology. 2013;115(4):515-22.

30. Barnes L, Eveson JW, Sidransky D, Reichart P. Pathology and genetics of head and neck tumours: IARC; 2005.

31. Ladeinde AL, Ajayi OF, Ogunlewe MO, Adeyemo WL, Arotiba GT, Bamgbose BO, et al. Odontogenic tumors: a review of 319 cases in a Nigerian teaching hospital. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. 2005;99(2):191-5.

32. Tawfik MA, Zyada MM. Odontogenic tumors in Dakahlia, Egypt: analysis of 82 cases. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. 2010;109(2):e67-e73.

33. Varkhede A, Tupkari JV, Sardar M. Odontogenic tumors: a study of 120 cases in an Indian teaching hospital. 2011.

34. Avelar RL, Primo BT, Pinheiro-Nogueira CB, Studart-Soares EC, de Oliveira RB, de Medeiros JR, et al. Worldwide incidence of odontogenic tumors. Journal of Craniofacial Surgery. 2011;22(6):2118-23.

35. Naz I, Mahmood MK, Akhtar F, Nagi AH. Clinicopathological evaluation of odontogenic tumours in pakistan-A seven years retrospective study. Asian Pacific journal of cancer prevention. 2014;15(7):3327-30.

36. Philipsen HP, Reichart PA. Revision of the 1992-edition of the WHO histological typing of odontogenic tumours. A suggestion. Journal of oral pathology & medicine. 2002;31(5):253-8.

37. Mosqueda Taylor A. New findings and controversies in odontogenic tumors. 2008.

38. Ebenezer V, Ramalingam B. A cross-sectional survey of prevalence of odontogenic tumours. Journal of maxillofacial and oral surgery. 2010;9(4):369-74.

39. Lawal AO, Adisa AO, Olusanya AA. Odontogenic tumours: A review of 266 cases. Journal of clinical and experimental dentistry. 2013;5(1):e13.

40. Lima-Verde-Osterne R, Turatti E, Cordeiro-Teixeira R, Barroso-Cavalcante R. The relative frequency of odontogenic tumors: A study of 376 cases in a Brazilian population. Medicina oral, patologia oral y cirugia bucal. 2017;22(2):e193.

41. Reichart P, Philipsen H, Sonner S. Ameloblastoma: biological profile of 3677 cases. European Journal of Cancer Part B: Oral Oncology. 1995;31(2):86-99.

42. Gupta B, Ponniah I. The pattern of odontogenic tumors in a government teaching hospital in the southern Indian state of Tamil Nadu. Oral Surgery, Oral Medicine, Oral Pathology, Oral Radiology, and Endodontology. 2010;110(1):e32-e9.

43. Sharma PN, Ranka RK, Chaudhary MS, Gawande MN, Hande AH, Zade PF. Odontogenic tumors: A review of 93 cases in the Vidharba region of Maharashtra. Journal of Oral and Maxillofacial Pathology. 2020;24(1):185.