

Radiological and clinical evaluation of maxillofacial cysts and tumors in patients referred to Hospitals in Kermanshah during 2008-2012

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Abstract

Background: Maxillofacial cysts and tumors are major causes of jawbone destruction; and may occasionally result in early mortality of patients. This study purposed to radiologically and clinically evaluate maxillofacial cysts and tumors among Iranian patients.

Methods: A retrospective study was conducted on medical records of patients referred to surgical departments of two hospitals (Bisotun and Imam Khomeini) in Kermanshah, Iran, from 2008 to 2012. Totally, 347 records were reviewed and 85 cases with intraosseously maxillofacial lesions were selected. The information including age, gender, type of lesion and its characteristics (number of lesions, anatomic location, radiolucency, and lesion border) were collected. Data were presented by descriptive statistics using SPSS 20 software.

Results: Overall occurrence of maxillofacial lesions was 24.5% (85/347) including 41 cysts, 14 benign tumors, 10 malignant tumors, and 20 bone diseases in jaws. The most common cysts, benign tumor, malignant tumor, and jawbone disease was radicular cyst (41.5%), ameloblastoma (57.1%), squamous cell carcinoma (75%), and central giant

cell granuloma (65%), respectively. The posterior region of mandible and anterior part of maxilla were respectively identified as the first and second most common anatomic locations for development of the maxillofacial lesions. The peak age of patients was third and fourth decade for the majority of lesions but seventh and eighth decade for squamous cell carcinoma.

Conclusions: Our findings are generally consistent with those reported in the literature. However, there are a few dissimilarities that may be due to racial and/or environmental differences.

Key words: Maxillofacial, cysts, benign tumor, malignant tumor

Introduction

Jawbones as the main hard tissue of orofacial region can be the sites for development of various conditions such as cysts, neoplasms, or systemic bone diseases. Approximately, 90% of metastatic tumors of the orofacial region occur in jaw bones, especially the mandible [1].

Jaw cyst is described as a pathological cavity that contains fluid, semi-fluid, or gaseous substance. Its frequency has been reported from 7.8% to 36% of jawbone biopsies [2]. Radicular cyst, dentigerous cyst, and odontogenic keratocyst (OKC) are the three most common cysts in jawbones [3].

Odontogenic tumors are a heterogeneous group of tumors with specific location in the mandible and maxilla. These tumors are benign (e.g. ameloblastoma, myxoma, odontoma, and fibroma) or malignant such as squamous cell carcinoma (SCC). SCC has been reported as the most prevalent malignant tumor in jaw-bones of adult patients. While, Burkitt's lymphoma and rhabdomyosarcoma are common among children and adolescents[4].

Cystic and tumoral lesions in mandible and maxilla are important because of resulting in morbidities such as jaw swelling, pain and sensory disturbances [5]; displacement of tooth [6]; occlusal alterations; and failure of eruption of teeth [7]. Moreover, a large portion of primary intraosseous malignancies of the jaws are originated from pre-existing cysts more commonly odontogenic cysts [8]. Aggressive lesions of jaws may also invade and destroy the walls of maxillary sinus, floor of the orbit and nasal wall[9]. Early death of patients with mandibular and maxillary malignancies may occur from infection or metastatic spread [10].

The overall and relative frequency of cystic and tumoral lesions in jawbones differs among various populations that may be attributed to variations in geographic or cultural settings. The aim of this study was to evaluate maxillofacial cysts and tumors among Iranian patients according to age, gender, anatomic area, and radiological features.

Materials and Methods

This descriptive cross-sectional study was performed by using medical records of patients referred to the surgical department of two hospitals (Bisotun and Imam Khomeini) in Kermanshah city, Iran, from 1 January 2008 to 31 December 2012.

A total of 367 records were reviewed and 85 cases with intraosseously maxillofacial lesions confirmed by histopathology diagnosis were selected. An oral-maxillofacial radiologist examined all associated radiographs. Demographic information (age and gender) as well as type of lesion and its characteristics (number of lesions, anatomic location, radiolucency, and lesion border) were collected.

The data were analyzed by SPSS 20 (SPSS Inc., Chicago, IL, USA). Descriptive statistics (frequency and percentage) were used to represent the results.

Results

A total of 85 patients had intraosseously maxillofacial lesions including 41 cysts, 14 benign tumors, 10 malignant tumors, and 20 bone diseases presented in jaws. Therefore, overall occurrence of maxillofacial lesions was 24.5% (85/347).

Radicular cyst was the most frequent cystic lesion (41.5%; 9 males, 8 females), followed by OKC (29.3%) and dentigerous cyst (14.6%). The most common benign and malignant tumor was ameloblastoma (57.1%; 5 males, 3 females) and SCC (80%; 5 males, 3 females), respectively. The first and second most common bone diseases in jaws was CGCG (65%; 5 males, 8 females) and cement-ossifying fibroma (25%). (Table - next page)

The peak age of presentation was third and fourth decade for radicular cyst, OKC, ameloblastoma, and osteosarcoma; but seventh and eighth decade for SCC. Of 17 patients with radicular cyst, 14 patients had one cyst and three patients had 2 cysts including a total of 20 cysts. Twelve patients with OKC included 16 cysts (8 patients with 1 cyst and 4 patients with 2 cysts). The other patients presented with one lesion.

Radicular cysts commonly occurred in the anterior region of the maxilla (45%). Posterior part of mandible was detected as the most frequent anatomic location for OKC (68.8%), dentigerous cyst (66.6%), ameloblastoma (75%), SCC (75%), and bone diseases (75%). (Table)

Most cysts were typically observed as radiolucent areas with well-defined borders. Of benign tumors, ameloblastoma and myxoma were radiolucent, but odontoma and osteoma were radio-opaque. Hemangioma presented a mix of radiolucency and opacity. Benign tumors commonly showed well-defined borders. All malignant tumors presented with undefined borders. SCC was radiolucent and osteosarcoma had a mixed radiolucent/radio-opaque view. Most jawbone diseases were characterized with radiolucent areas and well-defined borders.

Discussion

Studies of jawbone cystic and tumoral lesions among the population are essential for identification of individuals at risk, possible factors of development, and for accurate differential diagnosis. Therefore, we aimed to determine clinical and radiographic pattern of maxillofacial cystic and tumoral lesions among patients in Kermanshah, a city in west of Iran.

In this study, the most common cystic lesions were radicular cyst(41.5%), followed by OKC (29.3%) and dentigerous cyst (14.6%) that is consistent with results of other studies[11,12]. However, Rezvani et al reported oral cystic

Table: Frequency distribution of maxillofacial lesions

Maxillofacial lesions	Frequency of patients (%)	Peak age (decade)	location			
			Maxilla		Mandible	
			Anterior	Posterior	Anterior	Posterior
Cysts:						
Radicular cyst*	17 (41.5%)	3rd and 4th	9 (45%)	2 (10%)	4 (20%)	5 (25%)
Odontogenic keratocyst**	12 (29.3%)	3rd and 4th	1 (6.2%)	3 (18.8%)	1 (6.2%)	11 (68.8%)
Dentigerous cyst	6 (14.6%)	< 5th	1 (16.7%)	1 (16.7%)	0	4 (66.6%)
Nasolabial cyst	2 (4.9%)	-	2	0	0	0
Simple bone cyst	2 (4.9%)	-	0	0	0	2
Residual cyst	2 (4.9%)	-	1	0	0	1
Total	41 (100%)	3rd and 4th	14	6	5	23
Benign tumors:						
Ameloblastoma	8 (57.1%)	3rd and 4th	1 (12.5%)	1 (12.5%)	0	6 (75%)
Odontoma	3 (21.4%)	1st and 2nd	0	1	2	0
Myxoma	1 (7.1%)	-	0	1	0	0
Hemangioma	1 (7.1%)	-	0	0	0	1
Osteoma	1 (7.1%)	-	0	1	0	0
Total	14 (100%)	-	1	4	2	7
Malignant tumors:						
Squamous cell carcinoma	8 (80%)	7th and 8th	0	1	1	6 (75%)
Osteosarcoma	2 (20%)	3rd and 4th	1	0	0	1
Total	10 (100%)	-	1	1	1	7
Bone diseases:						
Central giant cell granuloma	13 (65%)	-	-	-	-	-
Cement-ossifying fibroma	5 (25%)	-	-	-	-	-
Periapical cemental dysplasia	1 (5%)	-	-	-	-	-
Fibrous dysplasia	1 (5%)	-	-	-	-	-
Total	20 (100%)	3rd	-	-	-	75%

* 14 patients with 1 cyst and 3 patients with 2 cysts

** 8 patients with 1 cyst and 4 patients with 2 cysts

lesions in decreasing frequency as radicular cyst (32.83%), dentigerous cyst (31.34%), and OKC (26.12%) [13]. The study by Baghaei et al showed prevalence of dentigerous cysts 27.2%, radicular cysts 18.6% and OKC 18.6% [14]. This variation indicates that the racial and environmental factors probably influence on development of these lesions.

Radicular cysts were more common among patients in the third and fourth decade of life. This finding is consistent with a study in Italy [3]. Arotiba et al indicated that mean age of occurrence was 26.5 years for radicular cyst [15]. In a study by Meningaud et al, patients with radicular cyst were 38.4 ± 17.9 years [16]. Nevertheless, Fomete et al reported second decade as peak age for radicular cysts among Nigerian patients [17]. Radicular cysts occurred most frequently in the anterior part of the maxilla (45%) as

reported previously by Tortorici et al [3], Ramachandra et al [11], and Koseoglu et al [12].

Peak age incidence of OKC was third and fourth decade of life which agrees with the study on Iranian patients in Mashhad city [18] as well as studies on Indians [11] and Turkish populations [12]. OKC was chiefly located in the posterior part of the mandible (68.8%). This finding is similar to previous studies indicating posterior region of mandible as the main location of OKC [11,19]. Goteti reported OKC location with a mandible-to-maxilla ratio of 1.6:1 [20]. OKC occurs most often intraosseously in the third molar region, angle, and ramus of mandible but rarely in soft tissues such as gum, oral mucosa, or lateral facial deep region [21].

Peak age incidence of dentigerous cysts was third and fourth decade. This is concomitant to most studies in the literature. Contrarily, Fomete et al showed common occurrence of dentigerous cysts in younger patients in second and third decades [17]. Dentigerous cyst was more frequent in the posterior part of mandible (66.6%). This is comparable to a study by Ramchandra et al [11].

The most frequent benign tumors were ameloblastoma (33.4%) followed by odontoma (12.5%). Consistently, ameloblastoma has been reported as the most common benign maxillofacial tumor among patients in Nigeria[22], China [23], and Egypt[24]. However, Tamme et al[25], Santos et al [26], and Ochsenius et al [27] demonstrated the first and second most common benign tumors were odontoma and ameloblastoma, respectively.

The peak age of ameloblastoma was the third and fourth decade of life. Peak incidence of ameloblastoma has been reported third decade in Indians [19] and Libyans [20], and fourth decade in Nigerians [22]. A review of 3677 cases of ameloblastoma by Reichart et al indicated that ameloblastoma tumors occur 10 to 15 years earlier in developing countries (average 27.7 years) than industrialized countries (average 39.1 years) [28]. Ameloblastoma was located more often in the posterior region of mandible (75%) followed by maxilla (25%). A similar, study on Iranian population in Mashhad city by Saghravani et al revealed that the most frequent location of the tumor was the posterior part of mandible (66/88)[29]. Ameloblastoma occurrence with a predilection for mandible has been noted in previous studies [30,31]. Reichart et al reported that ameloblastoma tumors are seen more frequently in the anterior region of the jaws among Blacks (21.6%) compared to Caucasians (12.6%) and Asians (11.9%)[28]. According to Siar et al, unicystic and solid/multicystic ameloblastoma tumors involve predominantly the body and posterior region of mandible, whereas desmoplastic ameloblastoma preferentially involves the anterior part[32].

In the present study, the second most common benign tumor was odontoma (12.5%) with peak age of incidence in the first and second decade. Taghavi et al indicated peak age occurrence of second decade for odontoma[33]. Odontoma was located in anterior region of mandible (two cases) and posterior region of maxilla (one case). The study by Isola et al reported that odontoma more likely occurred in mandible than maxilla with ratio about 2:1 [34]. However, Taghavi et al showed a predilection of odontoma for maxilla (59.3%) than mandible [33].

The most common malignant tumor was SCC detected in eight patients (80%). Parkins et al reported that SCC was present in 64% of orofacial malignant tumors from Ghana patients[35]. Similarly, SCC was reported as the most common malignant orofacial tumor among populations in Nigeria[36] and Portugal [37]. The present study showed that SCC commonly presented in patients aged 60+ years. In previous studies conducted by Monteiro et al[37] and Bassey et al[4], oral and maxillofacial SCC was often

found among patients aged 50+ years. SCC was mostly located in posterior part of mandible (75%). Bassey et al found 9 and 6 SCC tumors in mandible and maxilla, respectively [4].

In our study, osteosarcoma was identified in two patients (20%). Peak age of osteosarcoma was third and fourth decades of life that is consistent with a previous report [4]. This is comparable to Ajayi et al that found patients with sarcomas in orofacial region were younger than those with carcinomas [36].

The most systemic bone disease with involvement of jaws was CGCG. This condition is an intraosseous lesion that rarely occurs in the Head and Neck region[38], involves the mandible more than the maxilla and is more common in second and third decade of life [39].

Other conditions in our study sample were nasolabial cyst, simple bone cyst, residual cyst (2 cases each one), myxoma, hemangioma, osteoma (1 case each one), cement-ossifying fibroma (5 cases), periapical cemental dysplasia, and fibrous dysplasia (1 case each). Simple Bone Cyst also known as Traumatic Bone Cyst is a non-neoplastic osseous lesion that appears as a rare pathology, representing approximately 1% of all jaw cysts [40,41]. Hemangioma is a benign neoplasm with endothelial origin that is usually located in soft tissues but rarely develops in bones such as mandible [42].

Conclusion

This study showed that cysts were the most common lesions in the maxillofacial region followed by benign and malignant tumors. The most common cyst, benign tumor, malignant tumor, and jawbone disease was radicular cyst, ameloblastoma, squamous cell carcinoma, and central giant cell granuloma, respectively. The findings are generally consistent with reports in the literature; and some variations may be attributable to racial and/or environmental factors.

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References

1. Rajinikanth M, Prakash AR, Swathi TR, Reddy S. Metastasis of lung adenocarcinoma to the jaw bone. *J Oral Maxillofac Pathol* 2015; 19(3): 385-8.
2. Demirkol M, Ege B, Yanik S, Aras MH, Ay S. Clinicopathological study of jaw cysts in southeast region of Turkey. *Eur J Dent* 2014;8(1):107-11.
3. Tortorici S, Amodio E, Massenti MF, Buzzanca ML, Burrano F, Vitale F. Prevalence and distribution of odontogenic cysts in Sicily: 1986-2005. *J Oral Sci* 2008;50(1):15-8.
4. Bassey GO, Osunde OD, Anyanechi CE. Analysis of 46 cases of malignant jaw tumours in Calabar, Nigeria. *Niger Med J* 2015; 56(4): 240-3.
5. Grisar K, Schol M, Hauben E, Schoenaers J, Politis C. Primary intraosseous squamous cell carcinoma of the mandible arising from an infected odontogenic cyst: A case report and review of the literature. *Oncol Lett* 2016; 12(6): 5327-5331.
6. Priya K, Karthikeyan P, Nirmal Coumare V. Odontogenic Keratocyst: A Case Series of five Patients. *Indian J Otolaryngol Head Neck Surg* 2014; 66(1): 17-21.
7. Sharma S, Goyal D, Ray A, Gupta N. Ameloblastoma in children: should we be radical? *J Indian Soc Pedod Prev Dent* 2011; 29(6 Suppl 2): S74-8.
8. Borrás-Ferreres J, Sánchez-Torres A, Gay-Escoda C. Malignant changes developing from odontogenic cysts: A systematic review. *J Clin Exp Dent* 2016; 8(5): e622-e628.
9. Joshi J, Manjunatha BS, Kumar H, Kumar P. Tumor of the maxilla-odontogenic or glandular? A diagnostic challenge and the role of immunohistochemical markers. *J Cancer Res Ther* 2015; 11(4): 1031.
10. Etetafia MO, Arisi AA, Omoregie OF. Giant ameloblastoma mortality; a consequence of ignorance, poverty and fear. *BMJ Case Rep* 2014 May 22; 2014. pii: bcr2013201251.
11. Ramachandra P, Maligi P, Raghuvver H. A cumulative analysis of odontogenic cysts from major dental institutions of Bangalore city: A study of 252 cases. *J Oral Maxillofac Pathol* 2011; 15(1): 1-5.
12. Koseoglu BG, Atalay B, Erdem MA. Odontogenic cysts: a clinical study of 90 cases. *J Oral Sci* 2004; 46(4): 253-7.
13. Rezvani G, Pardis S, Sharifi E. A descriptive-clinical study of 134 cases of odontogenic cysts during a 10 year period. *Shiraz Univ Dent J* 2009; 10(2): 122-7.
14. Baghaei F, Zargarani M, Najmi H, Moghimbeigi A. A clinicopathological study of odontogenic cysts and tumors in Hamadan, Iran. *J Dent (Shiraz)* 2014; 15(4): 167-72.
15. Arotiba JT, Lawoyin JO, Obiechina AE. Pattern of occurrence of odontogenic cysts in Nigerians. *East Afr Med J* 1998; 75(11): 664-6.
16. Meningaud JP, Oprean N, Pitak-Arnop P, Bertrand JC. Odontogenic cysts: a clinical study of 695 cases. *J Oral Sci* 2006; 48(2): 59-62.
17. Fomete B, Osunde OD, Ogbeifun J, Agbara R, Ononiwu CN. A 10-Year Retrospective Analysis of 64 Cases of Cystic Lesions of the Oral and Maxillofacial Region in a Nigerian Tertiary Hospital. *Oman Med J* 2016; 31(6): 434-438.
18. Imani Moghadam M, Mojeri Khazani T. The evaluation of 41 panoramic radiographic cases of dentigerous cysts and odontogenic keratocysts. *J Mash Dent Sch* 2007; 30(1&2): 1-6. [in persian]
19. Nalabolu GR, Mohiddin A, Hiremath SK, Manyam R, Bharath TS, Raju PR. Epidemiological study of odontogenic tumours: An institutional experience. *J Infect Public Health* 2016 Jul 14. pii: S1876-0341(16)30071-5.
20. Goteti SH. Odontogenic Tumors: A Review of 675 Cases in Eastern Libya. *Niger J Surg* 2016; 22(1): 37-40.
21. Vázquez-Romero MD, Serrera-Figallo ML, Alberdi-Navarro J, Cabezas-Talavera J, Romero-Ruiz MM, Torres-Lagares D, et al. Maxillary peripheral keratocystic odontogenic tumor. A clinical case report. *J Clin Exp Dent* 2017; 9(1): e167-e171.
22. Bassey GO, Osunde OD, Anyanechi CE. Maxillofacial tumors and tumor-like lesions in a Nigerian teaching hospital: an eleven year retrospective analysis. *Afr Health Sci* 2014; 14(1): 56-63.
23. Jing W, Xuan M, Lin Y, Wu L, Liu L, Zheng X, Tang W, Qiao J, Tian W. Odontogenic tumours: a retrospective study of 1642 cases in a Chinese population. *Int J Oral Maxillofac Surg* 2007; 36(1): 20-5.
24. Tawfik MA, Zyada MM. Odontogenic tumors in Dakahlia, Egypt: analysis of 82 cases. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 2010; 109(2): e67-73.
25. Tamme T, Soots M, Kulla A, Karu K, Hanstein SM, Sökk A, Jõeeste E, Leibur E. Odontogenic tumours, a collaborative retrospective study of 75 cases covering more than 25 years from Estonia. *J Craniomaxillofac Surg* 2004; 32(3): 161-5.
26. Santos JN, Pinto LP, de Figueredo CR, de Souza LB. Odontogenic tumors: analysis of 127 cases. *Pesqui Odontol Bras* 2001; 15(4): 308-13.
27. Ochsenius G, Ortega A, Godoy L, Peñafiel C, Escobar E. Odontogenic tumors in Chile: a study of 362 cases. *J Oral Pathol Med* 2002; 31(7): 415-20.
28. Reichart PA, Philipsen HP, Sonner S. Ameloblastoma: biological profile of 3677 cases. *Eur J Cancer B Oral Oncol* 1995; 31B(2): 86-99.
29. Saghravanian N, Salehinejad J, Ghazi N, Shirdel M, Razi M. A 40-year Retrospective Clinicopathological Study of Ameloblastoma in Iran. *Asian Pac J Cancer Prev* 2016; 17(2): 619-23.
30. Lu Y, Xuan M, Takata T, Wang C, He Z, Zhou Z, Mock D, Nikai H. Odontogenic tumors. A demographic study of 759 cases in a Chinese population. *Oral Surg Oral Med Oral Pathol Oral Radiol Endod* 1998; 86(6): 707-14.
31. Siriwardena BS, Tennakoon TM, Tilakaratne WM. Relative frequency of odontogenic tumors in Sri Lanka: Analysis of 1677 cases. *Pathol Res Pract* 2012; 208(4): 225-30.
32. Siar CH, Lau SH, Ng KH. Ameloblastoma of the jaws: a retrospective analysis of 340 cases in a Malaysian population. *J Oral Maxillofac Surg* 2012; 70(3): 608-15.
33. Taghavi N, Rajabi M, Mehrdad L, Sajjadi S. A 10-year retrospective study on odontogenic tumors in Iran. *Indian J Dent Res* 2013; 24(2): 220-4.

34. Isola G, Cicciù M, Fiorillo L, Matarese G. Association between Odontoma and Impacted Teeth. *J CraniofacSurg* 2017 Jan 18. [Epub ahead of print]
35. Parkins GE, Armah GA, Tettey Y. Orofacial tumours and tumour-like lesions in Ghana: a 6-year prospective study. *Br J Oral MaxillofacSurg* 2009;47(7):550-4.
36. Ajayi OF, Adeyemo WL, Ladeinde AL, Ogunlewe MO, Effiom OA, Omitola OG, et al. Primary malignant neoplasms of orofacial origin: a retrospective review of 256 cases in a Nigerian tertiary hospital. *Int J Oral MaxillofacSurg* 2007;36(5):403-8.
37. Monteiro LS, Albuquerque R, Paiva A, de la Peña-Moral J, Amaral JB, Lopes CA. A comparative analysis of oral and maxillofacial pathology over a 16-year period, in the north of Portugal. *Int Dent J* 2017;67(1):38-45.
38. Garg P, Jain J, De N, Chatterjee K. A central giant cell granuloma in posterior part of maxilla-A case report. *Int J Surg Case Rep* 2017;30:222-225.
39. Janas A, Osica P. Central giant cell granuloma located in the maxilla in a 8-year old boy. *Dev Period Med* 2015;19(2):189-92.
40. Madiraju G, Yallamraju S, Rajendran V, SrinivasaRao K. Solitary bone cyst of the mandible: a case report and brief review of literature. *BMJ Case Rep* 2014;2014. pii: bcr2013200945.
41. Hs CB, Rai BD, Nair MA, Astekar MS. Simple bone cyst of mandible mimicking periapical cyst. *ClinPract* 2012;2(3):e59.
42. Gómez Oliveira G, García-Rozado A, Luaces Rey R. Intraosseous mandibular hemangioma. A case report and review of the literature. *Med Oral Patol Oral Cir Bucal* 2008;13(8):E496-8.