

## ORIGINAL ARTICLE

**Clinical and Radiologic Spectrum of Odontogenic Keratocyst: Descriptive Study**NAVEED KHAN<sup>1</sup>, ABDUR REHMAN<sup>1</sup>, ALAMGIR<sup>1</sup>, ZAINAB JADOON<sup>1</sup>, SHAGUFTA NAEEM<sup>2</sup>, SHABANA NAZ<sup>2</sup><sup>1</sup>Department of Dentistry, Ayub Medical College, Abbottabad- Pakistan.<sup>2</sup>Department of Pathology, Ayub Medical College, Abbottabad- Pakistan.Correspondence to Dr Abdur Rehman, E-mail: [mani79\\_pak@yahoo.com](mailto:mani79_pak@yahoo.com), Tel+92-333-5033123**ABSTRACT****Background:** Odontogenic keratocyst is relatively common developmental odontogenic lesion occurring with a frequency of approximately 10-14% of all jaw cyst.**Aim:** To find the clinic-radiologic pattern of odontogenic keratocyst cases.**Study design:** Descriptive cross sectional.**Methodology:** Present study was conducted at Pathology and diagnostic Medicine, Khyber Medical University Peshawar in which 83 patients were enrolled. An OPG x-ray was done for all the patients to see the nature of the lesion. Surgical specimens were stored in formalin solution. After gross examination, they were evaluated histopathologically for diagnosis of Odontogenic keratocyst according to WHO criteria<sup>2</sup>. Data was collected through performa by non-probability convenient sampling following ethical approval. Data was evaluated by using SPSS version 23. Quantitative data was presented as mean  $\pm$  SD. Categorical data was presented by frequency and percentage. Chi square was applied with p-value <0.05 as significant.**Results:** Almost 55.4% were male patients. Almost 45.8% patients were in the 4<sup>th</sup> decade of their life. The mandible was the most common site of involvement accounting for 68.7%. An impacted tooth was associated in 26.5% of cases.**Conclusion:** It was concluded that OKC involved mandible while occurring in 4<sup>th</sup> decade of life among males commonly. There was an association with an impacted tooth, therefore to rule out the presence of odontogenic keratocyst, all impacted teeth should be assessed.**Key words:** Impacted Tooth, Multilocular, Odontogenic Keratocyst and Satellite Cyst.**INTRODUCTION**

Odontogenic keratocyst is relatively common developmental odontogenic lesion occurring with a frequency of approximately 10-14% of all jaw cyst. It arises from the rest of dental lamina. In the past the term primodial cyst was used for odontogenic keratocyst.<sup>1</sup> It is generally consider as a developmental odontogenic cyst, but some researcher consider it as a tumour, and in 2005 it was reclassified as keratocystic odontogenic tumour.<sup>2</sup> But in 2017, "WHO suggest that there is less conclusive evidence to support a neoplastic nature of this pathology. The odontogenic keratocyst remains the most suitable terminology. Apart from "PATCH gene" mutation, There is involvement of MCC and TP53 genes<sup>3</sup>.

According to literature review, it has been reported that this disorder has highest incidence young age while affecting males commonly. This disease usually involve mandible. In Mandible, the third molar area and the ascending ramus are the more common site of involvement<sup>4</sup>. Maxillary lesions causes more morbidity. Extension into the soft tissue, adjacent bones, osseous expansion and destruction are reported. Patients may experience pain, swelling, limited mouth opening and paresthesia or anesthesia of the involved area. However the OKC can be completely asymptomatic and are diagnosed as incidental radiographic findings<sup>5</sup>.

High rate of recurrence are more common for OKC. Upto 62.5% of recurrence have been reported.<sup>6</sup> Satellite or daughter cyst formation, collagenase activity, high rate of mitotic activity and the remnants of dental lamina rests in the cystic wall are the reasons for the recurrence.

Radiographically OKC can presents as unilocular or multilocular radiolucency. Bigger unilocular OKCs can be mistaken for ameloblastomas<sup>7</sup> so careful assessment is mandatory. OKC can be found in two forms. 1-Associated with impacted tooth. This type attain a large size and are benign, less aggressive and less recurrent. 2-Unassociated with an impacted tooth. This type is usually multilocular, more aggressive and possess more recurrent potential<sup>8</sup>.

Histopathologically there is a thin, uniform epithelial lining with a thickness of 6-8 cells. Basal palisading and a corrugated parakeratin layer is seen. Recurrence of the OKC is indicated by hyalinisation especially in young individuals<sup>9</sup>. There is a natural ability in epithelial lining of OKC for spread and growth<sup>10</sup>. Odontogenic keratocyst can presents as single or solitary lesion or multiple lesions<sup>11</sup>.

This study presented the prevalent clinico-radiologic pattern for presentation of Odontogenic keratocyst in our population. Its results helped dentists to treat this disease at early stage with better management plan.

**METHODOLOGY**

This was a descriptive cross sectional study in which 83 cases were selected for data collection. After IRB permission it was carried at Institute of Pathology& Diagnostic Medicine Khyber Medical University Peshawar. An OPG (Orthopantomogram) x-ray was done for all the patients to see the nature of the lesion. Surgical specimens were stored in formalin solution. After gross examination, they were evaluated histo-pathologically for diagnosis of Odontogenic keratocyst according to WHO criteria<sup>2</sup>. Data was collected through performa by non-probability convenient sampling following ethical approval. Unwilling participants alongwith participants having carcinoma were excluded. Both genders with age ranging from 14-65years were included presenting with odontogenic keratocyst.

**Statistical analysis:** Data was evaluated by using SPSS version 23. Quantitative data was presented as mean  $\pm$  SD. Categorical data was presented by frequency and percentage. Chi square was applied with p-value <0.05 as significant.

**RESULTS**

Parameters like age (years), gender and site of involvement were presented as frequency and percentage. Age ranged from 14-65 years with mean $\pm$ SD (38.12 $\pm$ 10.13). Association of odontogenic keratocyst with impacted tooth and radiological variant was shown in table-2.

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Table-1: Baseline Parameters of enrolled population (n=83)

| Age in Groups (Years) | Frequency | Percentage (%) |
|-----------------------|-----------|----------------|
| Less than 20          | 3         | 3.6            |
| 20 --- 30             | 14        | 16.9           |
| 31 --- 40             | 38        | 45.8           |
| 41 --- 50             | 23        | 27.7           |
| 51 --- 60             | 3         | 3.6            |
| Over 60               | 2         | 2.4            |
| <b>Gender</b>         |           |                |
| Male                  | 46        | 55.4           |
| Female                | 37        | 44.6           |
| <b>Site</b>           |           |                |
| Mandible              | 57        | 68.7           |
| Maxilla               | 26        | 31.3           |

Table-2: Association of OKC with Impacted Tooth, Radiological Variant and Gender

| Association                 | Frequency | Percent (%) |
|-----------------------------|-----------|-------------|
| Yes                         | 22        | 26.5        |
| No                          | 61        | 73.5        |
| <b>Radiological variant</b> |           |             |
| Unilocular                  | 40        | 48.2        |
| Multilocular                | 43        | 51.8        |

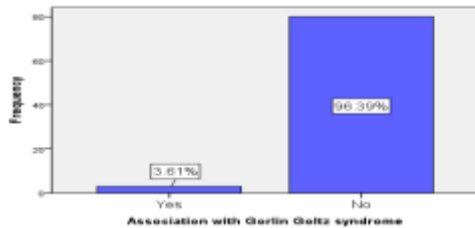
Insignificant association of twist expression with gender was shown in table-3 with p-value of >0.05.

Table-3: Association of twist expression with gender

| Score of Twist expression | Male      |         | Female    |         | Total     |         |
|---------------------------|-----------|---------|-----------|---------|-----------|---------|
|                           | Frequency | Percent | Frequency | Percent | Frequency | Percent |
| Negative                  | 20        | 43.5%   | 12        | 32.4%   | 32        | 38.6%   |
| Mild positive             | 8         | 17.4%   | 9         | 24.3%   | 17        | 20.5%   |
| Moderate positive         | 6         | 13.0%   | 10        | 27.0%   | 16        | 19.3%   |
| Strong positive           | 12        | 26.1%   | 6         | 16.2%   | 18        | 21.7%   |

Association of odontogenic keratocyst with Gorlin Goltz Syndrome was shown in figure-1.

Fig. 1: Association of OKC with Gorlin Goltz syndrome



**DISCUSSION**

Odontogenic keratocyst is a common odontogenic lesion of the maxillofacial area causing significant morbidity and in some cases mortality.

Our results were in line with many other previous studies. Parameters like gender distribution and age distribution was in the similar manner as reported in previously studies.<sup>12,13</sup> In our study males were predominant victims and usually were in their forties. Paradoxical to age distribution among enrolled participants in one study age ranged from 5 to 86 years<sup>14</sup>. However the mean age in our study was 38.12 years that was little high in comparison to 28.2 years. Paradoxical to our results, one study reported females as predominant victims than males<sup>15</sup>.

Our findings depicted that mandible was the common site affected /involved by OKC. Similar finding was documented by another study that reported mandible as common affected site in jaw.<sup>16</sup> It was reported that posterior part of the mandible was more frequently involved than the maxilla. Other studies reported that mandible was involved in 60-80% cases by OKC<sup>17,18</sup>. To our best knowledge, no study showed that maxilla is the most common site. It is hoped that involvement of maxilla will cause more morbidity and mortality due to more vital structures in structures involvement through its local extension. Maxilla is more commonly involved by syndromic OKC.

According to our results association of OKC with an impacted tooth was 26.5% which was similar to the findings of other studies that showed 27 % association with an impacted tooth.<sup>19,20</sup> However, low frequency of maxillary impacted teeth was associated with OKC that involved maxillary antrum.<sup>21</sup> Thus all

impacted teeth follicles lining must be sent for histopathological findings to exclude OKC.

It was found that syndromic odontogenic keratocyst was present in 3 cases (3.6%) while majority of the cases (96.4%) had solitary or non-syndromic odontogenic keratocyst. Syndromic patients had multiple OKC and this was a similar finding as reported previously<sup>22,23</sup>. One study reported that no association was seen between multiple OKC and Gorlin-Goltz syndrome<sup>24</sup>. It showed that careful assessment is mandatory to confirm the presence of Gorlin – Goltz, syndrome in the case of multiple OKC. Thus patient should be referred to specialist for its proper management. This syndrome is usually associated with malignancies and its common presentation is OKC. Recurrence rate of syndromic OKC is higher than the non syndromic OKC. Nevoid basal cell carcinoma syndrome is diagnosed approximately at age 13 years, but for this syndrome average age of diagnosis is 20years<sup>25</sup>.

**Limitations of study:** The limitations included single centre study with limited resources and finance.

**CONCLUSION**

It was concluded that OKC involved mandible while occurring in 4<sup>th</sup> decade of life among males commonly. Multilocular odontogenic keratocyst was more common than unilocular variety. Some odontogenic keratocyst were associated with impacted teeth, therefore to rule out this lesion all impacted teeth follicles should be sent for histopathological examination. Patients with multiple odontogenic keratocyst must be assessed for the presence of Gorlin Goltz syndrome.

**Author’s contribution:** NK&AR: Overall supervision and Write up and literature review, A&ZJ: Statistics application, analysis literature review, help in write up, SN&SN: Literature review help in write-up.

**Conflict of interest:** None

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**REFERENCES**

- Nayak MT, Singh A, Singhvi A, Sharma R. Odontogenic keratocyst: What is in the name?. Journal of natural science, biology, and medicine. 2013 Jul;4(2):282.
- Suluk-Tekkeşin M, Wright JM. The World Health Organization classification of odontogenic lesions: a summary of the changes of the 2017 (4th) edition. Turk Patoloji Derg. 2018;34(1):1-18
- Lunawat SD, Kunte VR, Bhoosreddy AR, Gade LP, Patil RS. Odontogenic keratocyst: a rare presentation in anterior maxilla. J Coll Physicians Surg Pak. 2020;30(11):1226-35.
- Boffano P, Cavarra F, Agnone AM, Bruccoli M, Ruslin M, Forouzanfar T, Ridwan-Pramana A, Rodríguez-Santamarta T, de Vicente JC, Starch-Jensen T, Pechalova P. The epidemiology and management of

- odontogenic keratocysts (OKCs): A European multicenter study. *Journal of Cranio-Maxillofacial Surgery*. 2022 Jan 1;50(1):1-6.
5. Borghesi A, Nardi C, Giannitto C, Tironi A, Maroldi R, Di Bartolomeo F, et al. Odontogenic keratocyst: imaging features of a benign lesion with an aggressive behaviour. *Insights into imaging*. 2018;9(5):883-97.
  6. Fidele NB, Yueyu Z, Zhao Y, Tianfu W, Liu J, Sun Y, Liu B. Recurrence of odontogenic keratocysts and possible prognostic factors: Review of 455 patients. *Medicina oral, patologia oral y cirugia bucal*. 2019 Jul;24(4):e491.
  7. Ramadevi S, Kumar MN, Khartik K. Odontogenic Keratocyst-A Case Report. *Indian Journal of Mednodent and Allied Sciences*. 2015;3(2):127-30.
  8. Marx RE. Jaw cysts, benign odontogenic tumors of the jaws, and fibro-osseous diseases. In *Current therapy in oral and maxillofacial surgery 2012 Dec 1* (pp. 390-410). Elsevier Inc..
  9. Cottom HE, Bshena FI, Speight PM, Craig GT, Jones AV. Histopathological features that predict the recurrence of odontogenic keratocysts. *Journal of oral pathology & medicine*. 2012;41(5):408-14.
  10. Meng Y, Zhao Y-N, Zhang Y-Q, Liu D-G, Gao Y. Three-dimensional radiographic features of ameloblastoma and cystic lesions in the maxilla. *Dentomaxillofacial Radiology*. 2019;48(6):20190066
  11. Arshad F. Syndromic odontogenic keratocyst: A case report and review of literature. *Journal of International Society of Preventive & Community Dentistry*. 2016;6(1):84-8.
  12. Titinchi F. Novel recurrence risk stratification of odontogenic keratocysts: A systematic review. *Oral Diseases*. 2022 Oct;28(7):1749-59.
  13. Mohanty S, Dabas J, Verma A, Gupta S, Urs A, Hemavathy S. Surgical management of the odontogenic keratocyst: a 20-year experience. *International Journal of Oral and Maxillofacial Surgery*. 2021;50(9):1168-76.
  14. Ansari MK, Alam S, Meraj F, Ahmed SS, Munir SA. Clinicopathological analysis of 847 odontogenic cysts in North Indian population examined over 10 years' period: A retrospective study. *Indian Journal of Oral Health and Research*. 2020 Jul 1;6(2):50.
  15. Du H, Wang S, Li M, Zhang DQ, Li G. CT/cone-beam CT image characteristics of ameloblastoma, odontogenic keratocyst and dentigerous cyst associated with the impacted mandibular third molar. In *2021 IEEE International Conference on Medical Imaging Physics and Engineering (ICMIPE) 2021 Nov 12* (pp. 1-5). IEEE.
  16. Ravi J, Wadhwan V, Gotur SP. Orthokeratinized versus parakeratinized odontogenic keratocyst: Our institutional experience. *Journal of Oral and Maxillofacial Pathology: JOMFP*. 2022;26(1):60.
  17. Alshibib AL, Al-jumaily HA, Alquisi AF. The Recurrence Rate of Odontogenic Keratocyst after Enucleation with Peripheral Osteotomy. *SRP*. 2020;11(4):690-4.
  18. Kammer Pv, Rivero Erc. Epidemiologic profile of syndromic and nonsyndromic odontogenic keratocyst cases: a 12-year retrospective study. *Oral Surgery, Oral Medicine, Oral Pathology And Oral Radiology*. 2020;129(1):E134-E5.
  19. Ogunsalu C, Daisley H, Kamta A, Kanhai D, Mankee M, Maharaj A. Odontogenic keratocyst in Jamaica: a review of five new cases and five instances of recurrence together with comparative analyses of four treatment modalities. *West Indian Med J*. 2007;56(1):90-5.
  20. Fidele N-B, Yueyu Z, Zhao Y, Tianfu W, Liu J, Sun Y, et al. Recurrence of odontogenic keratocysts and possible prognostic factors: Review of 455 patients. *Medicina oral, patologia oral y cirugia bucal*. 2019;24(4):e491.
  21. Mortazavi H, Baharvand M. Jaw lesions associated with impacted tooth: A radiographic diagnostic guide. *Imaging science in dentistry*. 2016;46(3):147-57.
  22. Baral R, Ojha B, Bajracharya D. Lesions Associated with Impacted Tooth. *J Kantipur Dent Coll*. 2020;1(1):25-31.
  23. Bachani L, Lingappa A. Odontogenic Keratocyst associated with impacted maxillary 3rd molar and involving the maxillary antrum: a rarity. *IP International Journal of Maxillofacial Imaging*. 2021;2(2):77-81.
  24. Garlapati K, Ignatius AV, Ajaykartik K, Suvarna C. Pathologies of impacted teeth: A cone-beam computed tomography diagnosis. *Indian Journal of Dental Sciences*. 2019;11(2):116.
  25. Kitisubkanchana J, Reduwan NH, Poomsawat S, Pornprasertsuk-Damrongsri S, Wongchuensoontorn C. Odontogenic keratocyst and ameloblastoma: radiographic evaluation. *Oral radiology*. 2021;37(1):55-65.