



Recurrence and Prognosticators of Recurrence in Odontogenic Keratocyst of the Jaws

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Abstract

Introduction The incidence of recurrence of OKC varied from 2.5 to 62%. Studies have linked recurrence to treatment methods and also clinical and pathological features. The aim of this study was to evaluate the 5-year recurrence and the factors associated with recurrence in odontogenic keratocysts of the jaws.

Methods A retrospective review of records was done from the Institute's Medical Records Directory from 2010 to 2021. The following data were obtained of the lesion; age at presentation, gender, site, subsite, radiographic presentation (locality), radiographic borders, presence or absence of satellite cysts, inflammatory infiltrate, and treatment rendered presence or absence of cortical perforation and soft-tissue extension and presence or absence of recurrence. Kaplan Meir estimator was used to evaluate recurrence rate and log rank test was used to compare the survival amongst groups. Cox regression analysis was used to evaluate the odds ratio to find out the possible factors influencing risk of

recurrence. A p value of <0.05 was considered statistically significant at 95% confidence interval.

Results In our study cohort, 27.2% of patients had recurrence. Posterior maxillary lesions, multilocular lesions, lesions with scalloped borders, presence of soft-tissue extension and cortical perforation, presence of satellite cysts and inflammatory infiltrate and enucleation with peripheral osteotomy were significantly associated with recurrence. However, soft-tissue extension, cortical perforation, multilocular lesions and presence of satellite cysts were independent risk factors.

Conclusion There is still debate on the best treatment modality for the management of OKCs. More studies are required to quantify the results.

Keywords Odontogenic keratocyst · Keratocystic odontogenic tumor · Jaw cyst · Recurrence · Survival · Outcome

Introduction

Background: Odontogenic keratocyst (OKC) is reclassified as a cyst of developmental origin according to the updated 2017 WHO classification of Head and Neck Tumors [1]. The 2005 WHO classification of Head and Neck tumors classified OKC as a tumor and named it as keratocystic odontogenic tumor owing to its aggressive behavior, solid variance, gene mutation and higher degree of recurrence [2]. However, the 2017 WHO classification reclassified it as a cyst in view of its resolution following conservative treatments and presence of genetic mutations in other odontogenic/non neoplastic lesions like dentigerous cysts [3]. In the early 1970s the reported recurrence rates were in the order of 40–60% on account of unfamiliarity with the aggressiveness

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of the lesion. Enucleation and curettage was the mainstay of treatment [4].

The incidence of recurrence of OKC varied from 2.5 to 62% with great variation in reporting, treatment and followup. The reported recurrence rate with only enucleation varied from 17 to 56% and when adjunctive treatment modalities like decompression or Carnoy's solution was used the recurrence rate varied from 1 to 8.7% [1, 5–7]. In 1976 Brannon proposed three mechanisms for OKC recurrence; incomplete removal of the lining, growth of new cysts from daughter cysts or rests left behind after surgery and development of new OKC in adjacent area [8]. The features that were considered to predict recurrence were higher level of cell proliferative activity in the epithelium, budding in the basal layer of the epithelium, parakeratinization of the surface layer, supraepithelial split of the epithelial lining, subepithelial split of the epithelial lining and presence of remnants/cell rests as well as daughter cysts [1]. Although these reasons have been cited there still remains debate on the exact cause for recurrence. Studies have linked recurrence to treatment methods and also clinical and pathological features (larger size, daughter cysts, cortical perforation, and association with dentition) [9–13]. The treatment modalities available include; enucleation (with primary closure, pack open, chemical fixation or cryosurgery), marsupialization (only or followed by enucleation) and resection [14, 15]. Owing to the variation in reporting, management, followup and lack of universal guidelines there still exists variation in reporting of outcome following management of OKCs. Hence we have undertaken a retrospective study to evaluate the recurrence free survival (RFS) and the factors associated with recurrence in non-syndromic odontogenic keratocysts of the jaws.

Methods

Study Design and Setting

A retrospective review was done from the Institute's Medical Records Directory from 2010 to 2021. Ethical clearance was obtained from the ethical committee of the Institute. The following study was conducted in accordance with STROBE guidelines [16].

Participants

Records were reviewed from January 2010 to December 2021 from the Medical Records Directory. Biopsy-proven patients of odontogenic keratocyst were taken into consideration. Patients with adequate preoperative data including imaging, morphological analysis and solitary lesions in the jaws were considered. Inadequate data, multiple jaw lesions,

syndromic patients, inability to review pathological reports, or archive them were the exclusion criteria. We collected 5-year (60 months) followup data from the archives. Recurrence free survival was defined as the time frame from the final diagnosis with histological report till the occurrence of relapse or last visit to the department. Patients with less than 5-year followup or who could not be contacted for further review and recurrences after 5 years were excluded.

Evaluation and Outcome

From the medical records, the following data were obtained of the lesion; age at presentation, gender, site, subsite, radiographic presentation (locularity), radiographic borders, presence or absence of satellite cysts, inflammatory infiltrate, and treatment rendered, presence or absence of cortical perforation and soft-tissue extension and presence or absence of recurrence. Authors PT and MM were the principal investigators in the study. Data collection and assessment of data was done by authors RNB and ST.

Data Analysis

The statistical analysis was performed using IBM SPSS software, version 21.0 (IBM, Armonk, NY, USA). Kaplan Meir estimator was used to evaluate recurrence rate and log rank test was used to compare the survival amongst groups. Cox regression analysis (univariate and multivariate) was used to evaluate the odds ratio to find out the possible factors influencing risk of recurrence. A *p* value of <0.05 was considered statistically significant at 95% confidence interval.

Results

Based on our selection criteria, a total of 180 patient records were accessed for final analysis. The mean age of presentation was 31.82 ± 0.27 years. The median age of presentation was 31 years (25–38). The male:female ratio in our study was 1:0.59. In our study cohort, a total of 49 patients (27.2%) had recurrence and 131 (72.7%) patients did not experience any recurrence at 5-year followup. Overall the power of the study was estimated to be 80%.

The most common site of involvement was posterior mandible (62.2%) followed by posterior maxilla (19.4%), anterior mandible (15.0%) and anterior maxilla (3.3%). Multilocularity was present in a total of 53.9% of patients. Most of the lesions had well-defined borders with scalloped margin being present in 38.9% of patients. Cortical perforation was present in 45% of patients and soft-tissue extension was present in 44.4% of patients. On histological analysis satellite cysts were present in a total of 44 patients and inflammatory infiltrate was present in 50.6% of patients

(Table 1). Three treatment procedures were principally carried out in our patients; I(enucleation with peripheral ostectomy), II(marsupialization followed by enucleation and peripheral ostectomy) and resection. Unilocular lesions with no sinus communication, no cortical perforations were treated by enucleation followed by peripheral ostectomy. Larger lesions with multilocularity, sinus communication were initially decompressed followed by enucleation and peripheral ostectomy. Enucleation after marsupialization was done 6–15 months following the primary procedure. During this followup period after marsupialization regular iodoform dressing was done. Patients undergoing iodoform

dressing were evaluated every 7–10 days for wound irrigation and pack change. In none of the maxillary lesions resection was done. Most of the posterior lesions had sinus involvement and marsupialization followed by enucleation was done. Resection was done in cases with multiple cortical perforations, extensive soft-tissue extension, pathological fracture or extensive involvement of ramus condyle area.

The overall 5-year recurrence free survival (RFS) in our cohort was 68.7%. A number of factors were evaluated which might affect recurrence (Figs. 1, 2, 3, 4, 5,6,7, 8, 9 and 10). The 5-year RFS was 72.7% for mandibular lesions and 55.5% for lesions in the maxilla. Multilocular lesions

Table 1 Log rank test of the predictor variables to recurrence free survival

	Frequency	Percentage	5-year survival	p value
Recurrence free survival			68.7% (over all)	
Predictor variable				
Sex				
Male	113	62.8%	74.0%	0.058
Female	67	37.2%	60.0%	
Site				
Mandible	139	77.2%	72.7%	0.053
Maxilla	41	22.8%	55.5%	
Sub Site				
Ant. mandible	27	15.0%	81.8%	0.21
Post. mandible	112	62.2%	70.7%	
Ant. maxilla	6	3.3%	60.0%	
Post. maxilla	35	19.4%	55.4%	
Radiographic presentation				
Unilocular	83	46.1%	87.6%	<0.001
Multilocular	97	53.9%	55.9%	
Borders				
Well defined	110	61.1%	84.5%	<0.001
Scalloped	70	38.9%	46.4%	
Soft-tissue extension				
Absent	100	55.6%	86.9%	<0.001
Present	80	44.4%	48.2%	
Cortical perforation				
Absent	99	55%	85.1%	<0.001
Present	81	45%	49.2%	
Satellite cysts				
Absent	136	75.6%	77.9%	<0.001
Present	44	24.4%	42.6%	
Treatment				
I (enucleation + peripheral ostectomy)	74	41.1%	41.0%	<0.001
II (decompression-enucleation + peripheral ostectomy)	67	37.2%	80.0%	
III (resection)	39	21.7%	100.0%	
Inflammatory infiltrate				
Absent	81	49.4%	86.5%	<0.001
Present	99	50.6%	53.3%	
Total	180	100	68.7%	

Fig. 1 Kaplan Meier estimator of sex as a prognosticator of recurrence

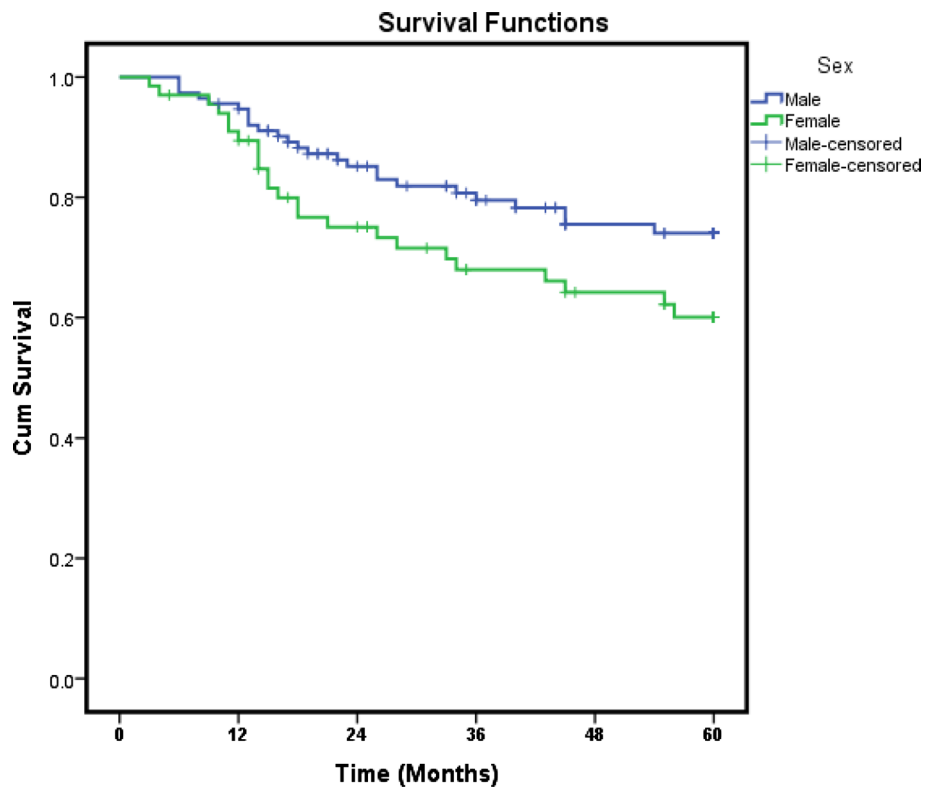
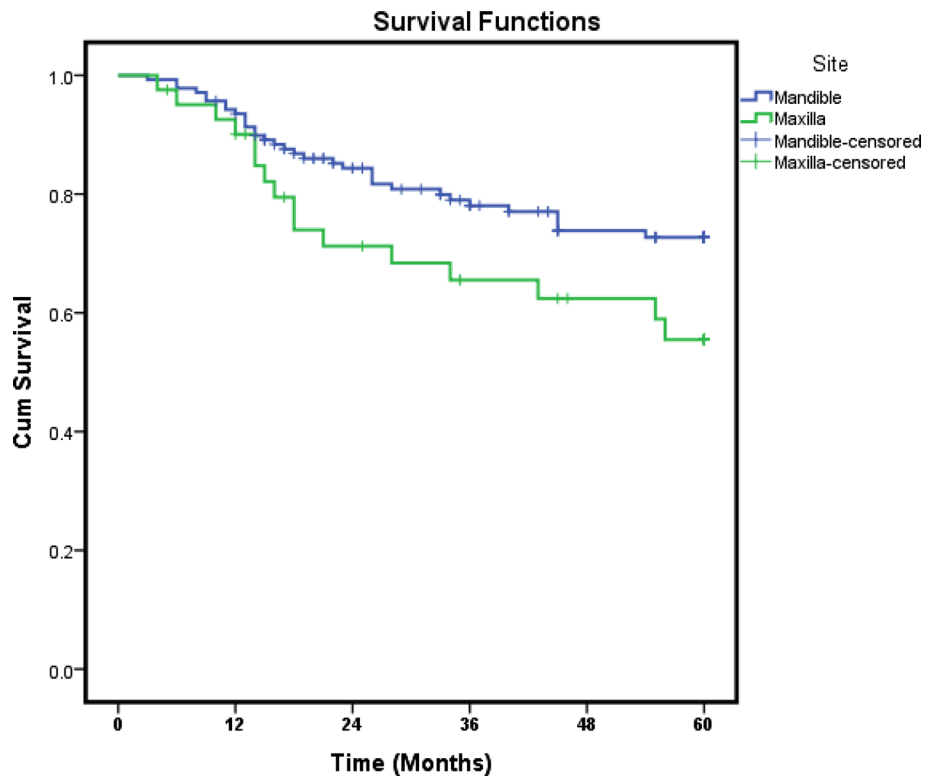


Fig. 2 Kaplan Meier estimator of tumor site as a prognosticator of recurrence



had a 5-year recurrence free survival of 55.9% compared to 87.6% of unilocular lesions. Lesions with scalloped margin had a 5-year recurrence free survival of 46.4% compared

to 84.5% of lesions with well-defined borders. Soft-tissue extension was associated with RFS of 48.2% compared to 86.9% RFS of lesions without soft-tissue extension. Cortical

Fig. 3 Kaplan Meier estimator of tumor sub-site as a prognosticator of recurrence

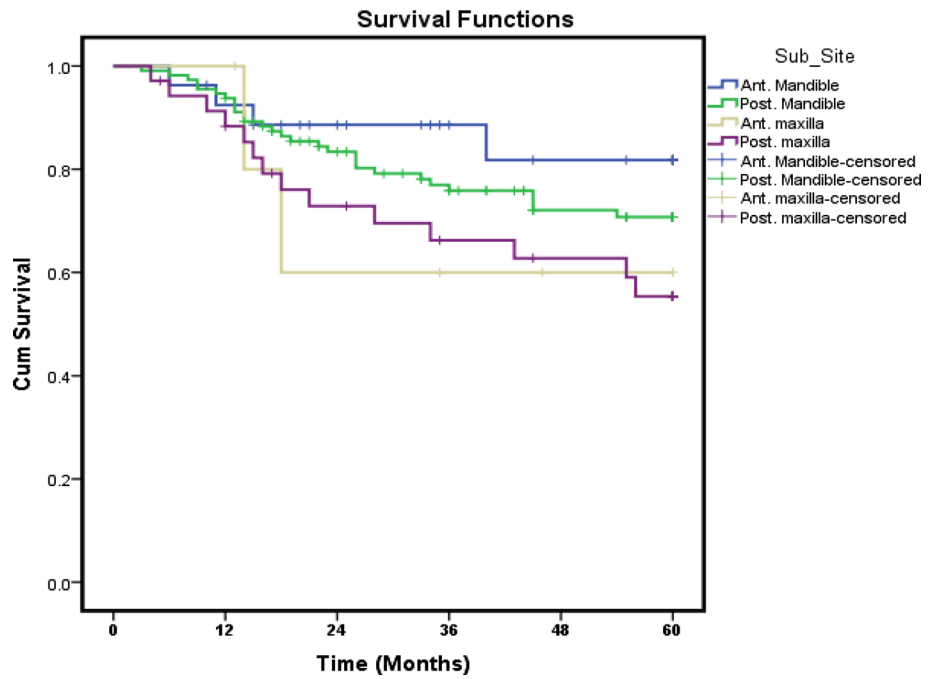
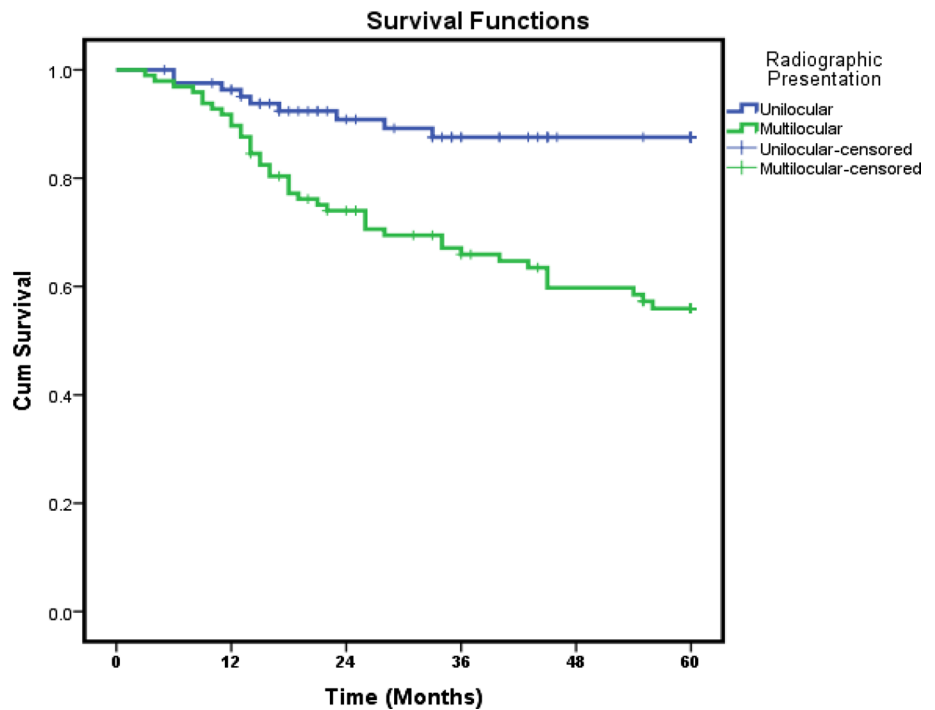


Fig. 4 Kaplan Meier estimator of radiographic presentation as a prognosticator of recurrence



perforation was associated with RFS of 49.2% and presence of inflammatory infiltrate had RFS of 53.3%. Lesions treated with resection had a RFS of 100% compared to 41% RFS of lesions treated with enucleation-peripheral osteotomy alone and 80% RFS of lesions treated in a staged manner. Presence of satellite cysts had a RFS of 42.6% (Table 1).

On univariate analysis lesions on the posterior maxilla, radiographic presentation, lesion border, soft-tissue

extension, cortical perforation, treatment, satellite cysts and inflammatory infiltrate were significantly associated with recurrence. Recurrence was present in 41.2% of patients with multilocular lesions compared with 10.8% of patients with unilocular lesions ($p < 0.001$). With regard to lesions margins, recurrence was present in 50% of patients with scalloped margin compared to 12.7% of patients with well-defined margins ($p < 0.001$). Presence of soft-tissue

Fig. 5 Kaplan Meier estimator of radiographic border as a prognosticator of recurrence

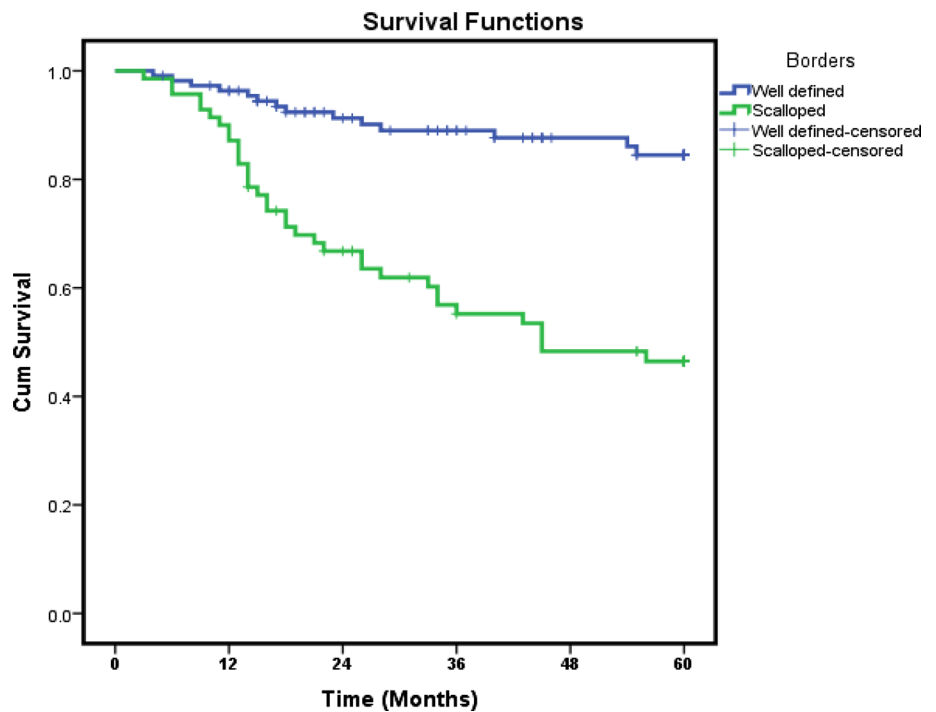
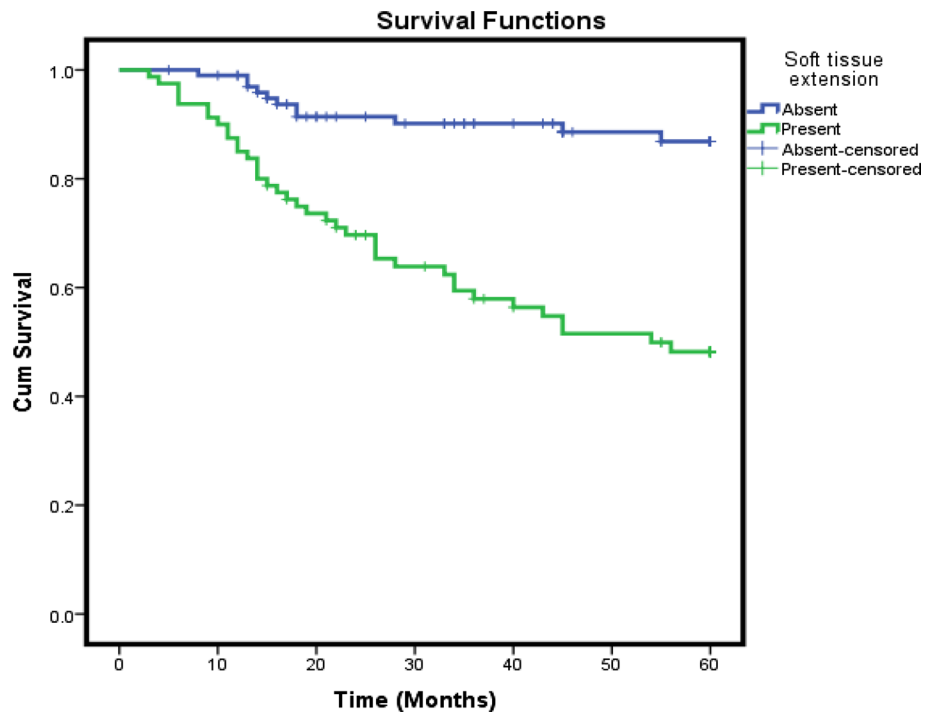


Fig. 6 Kaplan Meier estimator of soft-tissue extension as a prognosticator of recurrence



extension was associated with 47.5% recurrence compared to 11% of patients without soft-tissue extension ($p < 0.001$). Presence of cortical perforation was associated with 45.7% recurrence compared to 12.1% recurrence in patients without cortical perforation ($p < 0.001$). Presence of satellite cysts and inflammatory infiltrate were significantly associated with recurrence ($p < 0.001$, $p < 0.001$). Enucleation with

peripheral ostectomy was significantly associated with recurrence compared to staged procedure. No recurrence occurred in patients who were treated with resection (Table 2).

On multivariate analysis presence of soft-tissue extension, presence of cortical perforation, multilocular lesions and presence of satellite cysts were found as independent risk factors for recurrence. The OR (odds ratio) of having

Fig. 7 Kaplan Meier estimator of cortical perforation as a prognosticator of recurrence

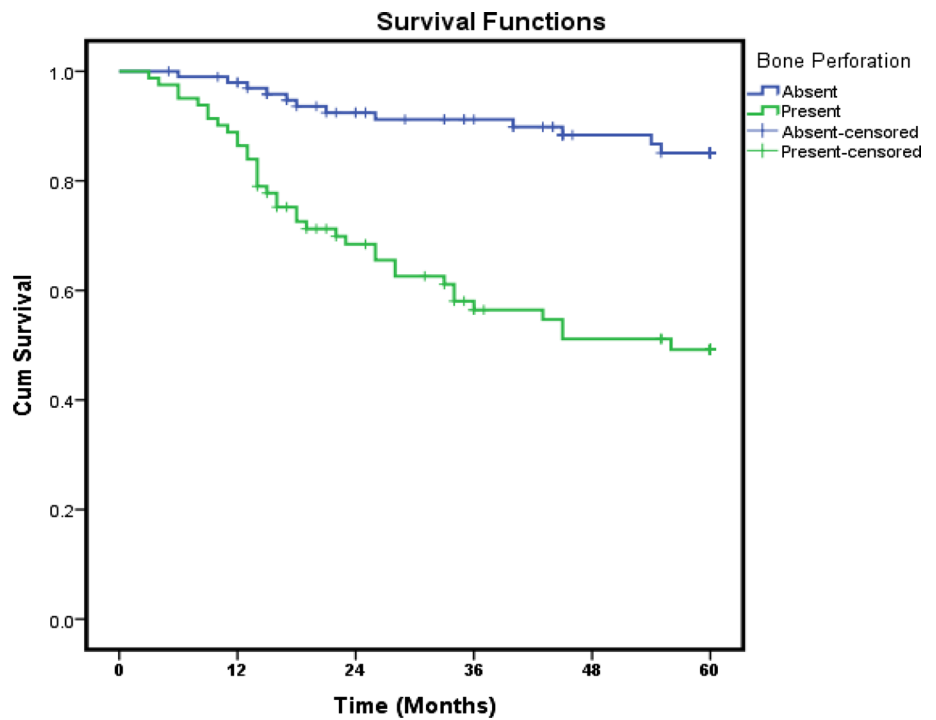
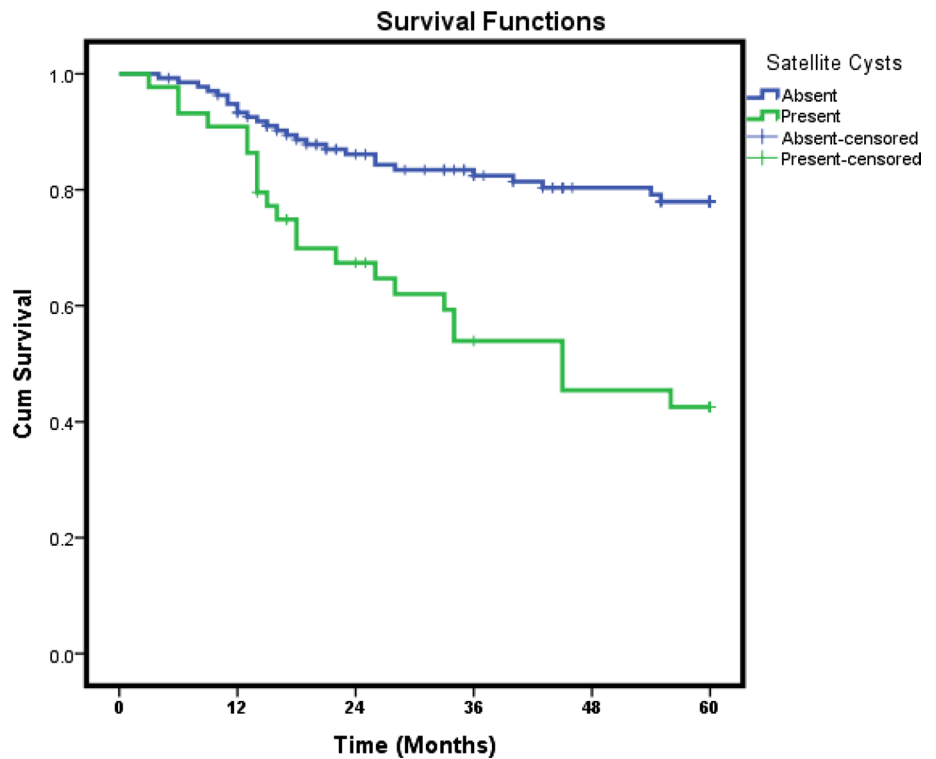


Fig. 8 Kaplan Meier estimator of satellite cysts as a prognosticator of recurrence



recurrence in the presence of soft-tissue extension was 8.203 compared to its absence (p 0.001). Cortical perforation was an independent risk factor for recurrence with OR 4.823 compared to its absence (p 0.018). The OR of having recurrence in the presence of satellite cysts was 8.004 compared to its' absence (p 0.007). Presence of

multilocular lesion was an independent risk for recurrence with OR 5.675 compared to its' absence (p 0.023) (Table 3).

Fig. 9 Kaplan Meier estimator of treatment as a prognosticator of recurrence

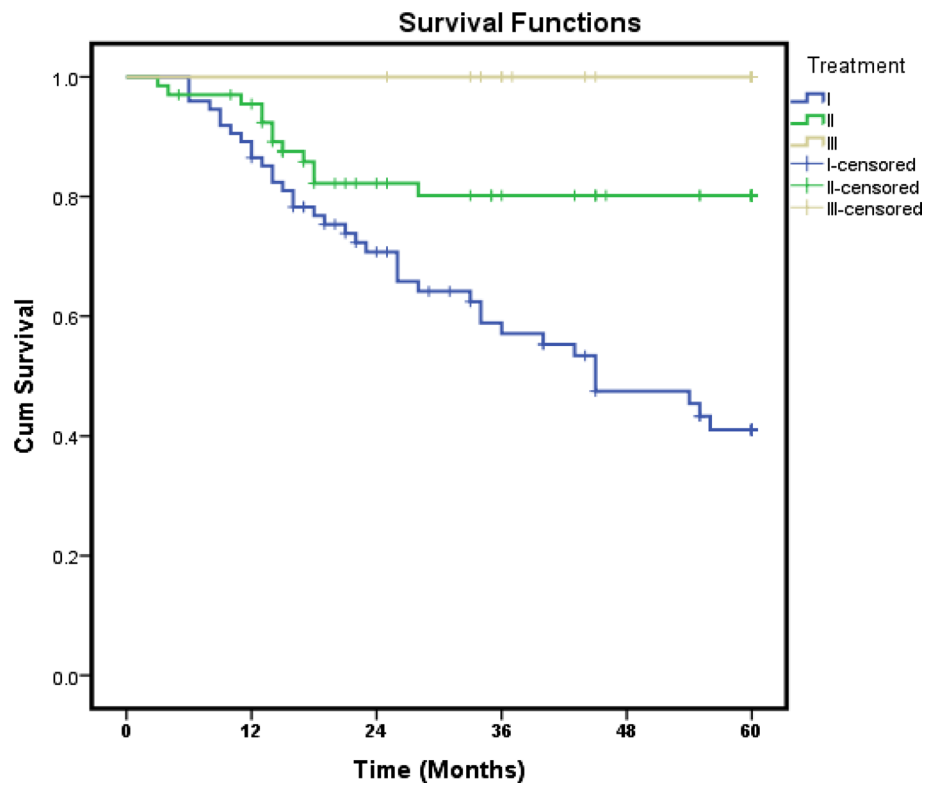


Fig. 10 Kaplan Meier estimator of inflammatory infiltrate as a prognosticator of recurrence

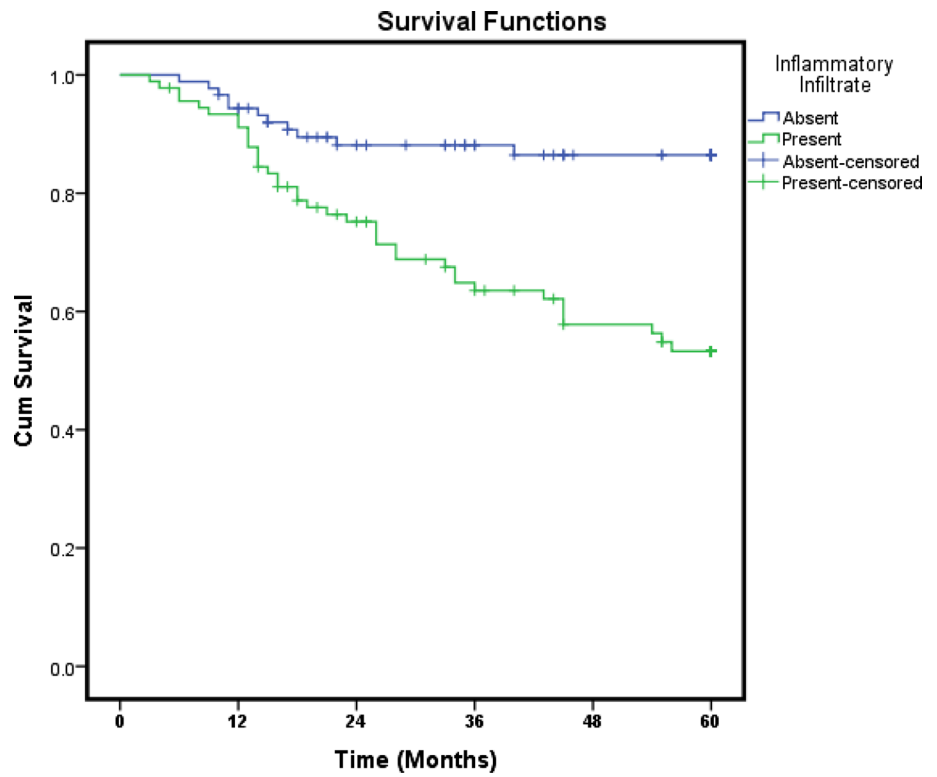


Table 2 Univariate regression to assess the relationship of predictor variables to recurrence

Predictor variables	Recurrence absent	Recurrence present	OR	95% CI: Lower	95% CI: Upper	<i>p</i> value
Site						
Mandible	106 (76.3%)	33 (23.7%)	Reference			
Maxilla	25 (61%)	16 (39%)	2.056	0.982	4.305	0.056
Sub_Site						
Ant. mandible	23 (85.2%)	4 (14.8%)	Reference			
Post. mandible	83 (74.1%)	29 (25.9%)	2.009	0.641	6.300	0.232
Ant. maxilla	4 (66.7%)	2 (33.3%)	2.875	0.388	21.289	0.301
Post. maxilla	21 (60%)	14 (40%)	3.833	1.089	13.499	0.036
Borders						
Well defined	96 (87.3%)	14 (12.7%)	Reference			
Scalloped	35 (50%)	35 (50%)	6.857	3.302	14.239	<0.001
Soft-tissue extension						
Absent	89 (89%)	11 (11%)	Reference			
Present	42 (52.5%)	38 (47.5%)	7.320	3.407	15.728	<0.001
Bone perforation						
Absent	87 (87.9%)	12 (12.1%)	Reference			
Present	44 (54.3%)	37 (45.7%)	6.097	2.894	12.845	<0.001
Satellite cysts						
Absent	110 (80.9%)	26 (19.1%)	Reference			
Present	21 (47.7%)	23 (52.3%)	4.634	2.233	9.613	<0.001
Inflammatory infiltrate						
Absent	78 (87.6%)	11 (12.4%)	Reference			
Present	53 (58.2%)	38 (41.8%)	5.084	2.386	10.831	<0.001
Treatment						
III	39 (100%)	0 (0%)				
II	55 (82.1%)	12 (17.9%)	Reference			
I	37 (50%)	37 (50%)	4.583	2.116	9.929	<0.001
Radiographic presentation						
Unilocular	74 (89.2%)	9 (10.8%)	Reference			
Multilocular	57 (58.8%)	40 (41.2%)	5.770	2.589	12.859	<0.001

Discussion

Posterior maxillary lesions, multilocular lesions, scalloped margins on radiograph, soft-tissue extension, cortical perforation, presence of inflammatory infiltrate, presence of satellite cysts and enucleation with peripheral osteotomy were significantly associated with recurrence. However, presences of soft-tissue extension, cortical perforation, multilocular lesions and presence of satellite cysts were independent risk factors for recurrence.

Limitations

1. Strict guidelines were not followed for treatment. Many times the treatment was decided based on patient's expectations which might influence survival. Since it was a retrospective study based on medical records this factor could not be changed.
2. No additional chemical fixation was carried out after enucleation. This might affect recurrence.
3. Adequate data was not available on the nature of basal epithelium of the lining which was taken into consideration in some studies.
4. We only considered 5-year recurrence rates. Long-term followups are needed to actually quantify the results.
5. Our study only evaluated results of non-syndromic OKCs. The recurrence of OKC in syndromic cases was too few to be evaluated at our centre.
6. Due to very few multiple lesions and inadequate data on followup, these lesions were not taken into consideration. Recurrence of multiple lesions and solitary lesions might be different.

Table 3 Multivariate regression analysis of predictor variable to recurrence

Site	OR	95% CI: Lower	95% CI: Upper	<i>p</i> value
Mandible	Reference			
Maxilla	0.496	0.045	5.508	0.568
Sub_Site				
Ant. mandible	Reference			
Post. mandible	0.119	0.013	1.051	0.055
Ant. maxilla	17.144	0.948	309.922	0.054
Post. maxilla	0.496	0.045	5.508	0.568
Borders				
Well defined	Reference			
Scalloped	1.113	0.178	6.939	0.909
Soft-tissue extension				
Absent	Reference			
Present	8.203	2.344	28.699	0.001
Cortical perforation				
Absent	Reference			
Present	4.823	1.311	17.741	0.018
Satellite cysts				
Absent	Reference			
Present	8.004	1.755	36.512	0.007
Inflammatory infiltrate				
Absent	Reference			
Present	1.426	0.295	6.881	0.659
Treatment				
III				
II	Reference			
I	1.751	0.47	6.524	0.404
Radiographic presentation				
Unilocular	Reference			
Multilocular	5.675	1.277	25.225	0.023

Interpretation

Two recent studies focused on the recurrence and prognosticators of recurrence in OKC. The study by França et al. had a total of 18 (45%) recurrent cases over 5-year followup and the study by Fidele et al. had a recurrence rate of 15.09% over 2–12 years followup. Presence of satellite cysts, inflammatory infiltrate and previous decompression or marsupialization was significantly associated with recurrence in the study by França et al. In the study by Fidele et al. preservation of the involved teeth, multilocular lesions and presence of daughter cysts were independent factors for recurrence. Although size > 4 cm was significantly associated with recurrence it was not an independent risk factor. Similar findings were also found in the study by França et al. with regard to lesion size. Enucleation alone had the highest recurrence rate followed by marsupialization-enucleation and resection. Nineteen of their patients had involved teeth with cortical perforation and recurrence was 100% in these cases. Most of the recurrences occurred in first 5 years with

decreasing incidence on subsequent years and the difference was statistically significant [17, 18].

A 2019 study by Kinard et al. [19] had an overall 19% recurrence with a median followup of 8 months. In their study maxillary lesions had more recurrence compared to mandible and multilocular lesions were significantly associated with recurrence. On multivariate analysis enucleation alone followed by enucleation with peripheral ostectomy had more recurrences compared to decompression followed by residual cystectomy. OKCs might present as both unilocular and multilocular lesions with unilocular being the predominant [19]. In our study a total of 83 patients had unilocular lesions and 97 had multilocular lesions. Recurrence was more in multilocular lesions and multilocularity was an independent risk factor.

In OKC satellite cysts takes three different forms; rounded keratin-filled cyst lined by flattened or cuboidal cells which can attain a very big size, squamoid structures with central degeneration occupied by epithelial debris and small irregular shaped cysts with lining indistinguishable

from that of the main cyst. Postulation favors basal cell layer budding might be involved in formation of satellite cysts. However, evidence is more in support of satellite cysts being developed from cell rests of Serres [20]. Presence of satellite cysts significantly influenced recurrence in the study by França et al. In the study of Fidele et al. both in univariate and multivariate analysis daughter cysts were significantly associated with recurrence. In our study presence of daughter cysts was significantly associated with recurrence and also an independent risk factor for recurrence. Presence of inflammatory infiltrate has been shown to be associated with recurrence. Inflammation increases the epithelial thickness and signals the proliferation of epithelial lining cells.

Marsupialization followed by a delayed enucleation has been shown to reduce the recurrence as the residual defect reduces in size. The postulation is that the epithelial cyst lining will undergo metaplasia to become undistinguished from the oral mucosa. The idea behind this treatment is that remnants of the epithelial cyst lining might impact recurrence left over during enucleation [21, 22]. A 2019 meta-analysis [23] evaluated the effects of enucleation alone versus marsupialization followed by enucleation in the management of OKCs. Marsupialization followed by enucleation reduced recurrence compared to enucleation alone with an odds ratio of 0.57. However, there was no strong evidence to support the statement and concluded on further studies to evaluate the evidence. Marsupialization and delayed enucleation reduced recurrence in 52% of OKCs compared to enucleation alone (OR 0.39, p 0.10). With peripheral ostectomy, the recurrence rate was reduced in 26% (OR 0.67, p 0.65). Overall staged procedure reduced the recurrence rate in 34% over enucleation (OR 0.57, p 0.17). A 2017 meta-analysis of 6427 cases evaluated the recurrence probability in keratocystic odontogenic tumor [24]. Overall the recurrence was 21.1%; 15.3% in maxilla, 21.5% in mandible, unilocular 14.7%, multilocular 24.4%, marsupialization/decompression 28.7%, decompression + enucleation \pm additional therapy 18.6%, enucleation/curettage 22.5%, enucleation + peripheral ostectomy 18.6%, enucleation + Carnoy's solution 5.3%, enucleation + cryotherapy 20.9%, marginal/segmental resection, 2.2%. The recurrence was not statistically significantly affected by lesion location (maxilla vs. mandible, risk ratio [RR] 0.92, $P=0.32$) or patient's sex (male vs. female, RR 0.94, $P=0.44$), but by locularity (unilocular vs. multilocular, RR 0.67, $P=0.007$). Recurrence risk for surgical managements: marsupialization vs. enucleation (RR 1.65, $P=0.0006$), marsupialization vs. resection (RR 3.17, $P=0.009$), enucleation alone vs. enucleation + peripheral ostectomy (RR 1.66, $P=0.05$), enucleation alone vs. enucleation + Carnoy's solution (RR 1.94, $P=0.03$), enucleation alone vs. enucleation + cryotherapy (RR 0.88, $P=0.56$). Another meta-analysis showed an overall recurrence rate of 19% with conservative treatment [25]. Decompression and

marsupialization alone had a recurrence rate of 18.5 and 18.2% respectively. Decompression followed by enucleation and marsupialization followed by enucleation had a recurrence rate of 11.9 and 17.8% respectively. Enucleation alone had a recurrence of 20.8%. Decompression followed by enucleation had a recurrence of 15.8% compared to 27.2% with enucleation alone (OR 0.48, p 0.0163). Marsupialization followed by enucleation had fewer recurrence compared to enucleation alone; however, the results were not statistically significant. Comparing marsupialization alone versus enucleation also did not show statistical significant results. Similar insignificant difference was obtained for decompression alone compared to enucleation. However, enucleation was favored compared to either decompression or marsupialization alone in reducing recurrence and marsupialization followed by enucleation was also favored. Cutler and Zollinger's modified Carnoy's solution instillation into the cystic cavity fixes the lining and helps in more complete removal. Carnoy's solution penetrates bone to a depth of 1.54 mm [26] and might cause irreversible neurotoxicity, adjacent soft-tissue toxicity, skin and dental follicles in children, irreversible damage to the devitalized osseous margin and no possibility of immediate bone grafting [25]. A recent clinical study has shown that the use of carnoy's solution was not significantly related to the normalness of sensory function at the end of followup and there would be no damage to the nerves if the inferior alveolar nerve is not exposed to the solution more than 3 min [27].

Conclusion

Multilocular lesions, lesions with scalloped borders, presence of soft-tissue extension and cortical perforation, presence of satellite cysts and inflammatory infiltrate and enucleation with peripheral ostectomy were significantly associated with recurrence. However, soft-tissue extension, cortical perforation, enucleation with peripheral ostectomy and marsupialization followed by enucleation + peripheral ostectomy were independent risk factors for recurrence.

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Author Contributions All authors contributed to the study conception and design. All authors read and approved the final manuscript.

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Declarations

Conflict of interest None declared.

Ethical approval CPGIDSH/21/0185-A.

Consent for publication Authors give full consent to publish.

Consent to participate Not required. Retrospective study from medical records.

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