

Potential Recurrence of Odontogenic Keratocyst Post-Surgery, Rapid Review

Cindy Ayu Widyandhika¹, Jamas Ari Anggraini², Harmas Yazid Yusuf³

¹ Program Study of Undergraduate, Faculty of Dentistry, Padjadjaran University, Indonesia

² Department of Oral Biology, Faculty of Dentistry, Padjadjaran University, Indonesia

³ Department of Oral and Maxillofacial Surgery, Faculty of Dentistry, Universitas Padjadjaran, Bandung Indonesia

Corresponding: e-mail: cindy18001@mail.unpad.ac.id

ARTICLE INFO : Received, Jan 2, 2023; Revised, May 12, 2023; Accepted, June 9, 2023; Published June 20, 2023
ORCID : Widyandhika CA (0009-0005-6150-8961); Anggraini JA (0009-0002-3420-7128); Yusuf HY (0000-0001-7970-1181)
DOI : [10.24815/jds.v8i1.33454](https://doi.org/10.24815/jds.v8i1.33454)

ABSTRACT

Background: Odontogenic keratocyst is one of the most frequent cysts found in the jaw. Odontogenic keratocyst, or OKC, is a benign but locally aggressive cyst with a higher recurrence rate than other odontogenic cysts after surgical treatment. The OKC recurrence rate is reported to be around 2.5% to 62.5%. **Objective:** This rapid review analyzes the most frequent recurrence potential factors after surgical procedures. **Material and Methods:** Articles search by using a combination of the keywords "odontogenic keratocyst" "keratocystic odontogenic tumor," "potential," "probability," "recur," "relapse," "recurrence" in three databases; PubMed, ScienceDirect, and Google Scholar. Article inclusion criteria include articles that have data on the recurrence of odontogenic keratocyst after surgical treatment; in the English language; issued within the last 10 years (2012-2022); and using observational research with all types of study designs (cohort, cross-sectional, and case-control). This study used the PRISMA protocol (Preferred Reporting Items for Systematic Review and Meta-Analysis). **Results:** Thirteen articles with an observational study design were included in this rapid review, consisting of thirteen articles with retrospective cohort design. This study recorded two hundred fifty-three cases of recurrence out of 1263 cases of OKC, with a mean follow-up period of 53.6 months. Recurrences were found more often in male patients than female patients (1.58:1), especially in the third and fourth decades of age. The multilocular form of the lesion (n = 52), especially on the mandible, was more common than the unilocular form of the lesion (n = 83). Teeth preserved in the sites of involvement caused incomplete cyst removal and increased the risk of recurrence. **Conclusion:** This rapid review showed that the percentage of potential OKC recurrences ranged from 7.1% to 45%, with an average recurrence rate of 22.5%. The presence of a daughter or satellite cyst and residual teeth or teeth preserved in the lesion area that was not removed in the form of a large multilocular cyst affects the potential for recurrence.

Keywords: Potential recurrence, Rapid Review, Odontogenic keratocyst, Surgery

ABSTRAK

Latar belakang: Keratokista odontogenik merupakan salah satu kista yang paling sering ditemukan pada rahang. Keratokista odontogenik atau OKC, adalah kista jinak, tetapi agresif secara lokal, dan memiliki tingkat kekambuhan yang lebih tinggi daripada kista odontogenik lainnya setelah perawatan bedah. Tingkat kekambuhan OKC dilaporkan sekitar 2,5% hingga 62,5%. **Tujuan:** Tinjauan cepat ini bertujuan untuk menganalisis faktor potensi kekambuhan yang paling sering setelah prosedur pembedahan. **Materi dan Metode:** Pencarian artikel dengan menggunakan kombinasi kata kunci "odontogenic keratocyst" "keratocystic odontogenic tumor" "potential" "probability" "recur" "relapse" "recurrence" pada tiga database; PubMed, ScienceDirect, dan Google Scholar. Kriteria inklusi artikel meliputi artikel yang memiliki data kekambuhan keratokista odontogenik setelah perawatan bedah; dalam bahasa Inggris; diterbitkan dalam 10 tahun terakhir (2012-2022); dan menggunakan penelitian observasional dengan semua jenis desain penelitian (cohort, cross-sectional, dan case-control). Studi ini dilakukan dengan menggunakan protokol PRISMA (Item Pelaporan Pilihan untuk Tinjauan Sistematis dan Analisis Meta). **Hasil:** Tiga belas artikel dengan desain penelitian observasional dimasukkan dalam kajian cepat ini, yang terdiri dari tiga belas artikel dengan desain kohort retrospektif. Studi ini mencatat dua ratus lima puluh tiga kasus kekambuhan dari total 1263 kasus OKC, dengan masa tindak lanjut rata-rata 53,6 bulan. Kekambuhan lebih sering ditemukan pada pasien laki-laki dibandingkan pasien perempuan (1,58:1), terutama pada usia dekade ketiga dan keempat. Bentuk lesi multilokular (n = 52), terutama pada mandibula, lebih umum daripada bentuk lesi unilokular (n = 83). Gigi yang diawetkan di tempat yang terkena menyebabkan peningkatan kista yang tidak sempurna dan meningkatkan risiko kekambuhan. **Kesimpulan:** Tinjauan cepat ini menunjukkan bahwa persentase potensi kekambuhan OKC berkisar antara 7,1% hingga 45%, dengan tingkat kekambuhan rata-rata 22,5%. Adanya kista anak atau satelit, serta sisa gigi atau gigi yang diawetkan di area lesi yang tidak diangkat berupa kista multilokular besar, mempengaruhi potensi kekambuhan.

Kata kunci: Potensi kekambuhan, Odontogenic Keratokista, Pembedahan, Rapid review

1. Introduction

Odontogenic keratocyst is one of odontogenic cyst type.¹ Odontogenic keratocyst found in 12-14% from all *odontogenic cyst*.^{2,3} Odontogenic keratocyst or OKC, OKC is a cyst in the jaw that is benign, uni- or multi-cystic, which can originate from the dental lamina or the basal layer of the oral epithelium.⁴ OKC has unique characteristics, like being locally aggressive, having a high recurrence rate, and having a distinctive histological picture.³ OKC can be affected by genetic changes or mutations. The human homolog *Drosophila Patched* gene (PTCH) is the most important reported in OKC.^{5,6} PTCH mutations are also the underlying cause of Nevoid Basal Cell Carcinoma Syndrome (NBCCS).^{7,8} The PTCH mutation associated with NBCCS is characterized by multiple OKCs in the jaws.⁹

OKC is an odontogenic lesion with a higher recurrence rate and is more aggressive than other odontogenic cysts after surgical treatment.¹⁰ The types of surgical treatment that can be performed for odontogenic keratocysts vary, including curettage, enucleation, marsupialization or decompression, and resection.¹¹ These treatments can be done with or without combinations to prevent recurrence, such as in combination with peripheral ostectomy, the addition of Carnoy's solution, or the addition of liquid nitrogen cryotherapy.^{3,12}

The OKC recurrence rate is reported to be around 2.5% to 62.5%.¹³ Various factors, such as incomplete cyst removal, remnants of epithelial or satellite cysts at the bone margins, and the aggressive nature of OKC, cause this recurrence.¹⁴ Incomplete removal of the cyst results in the formation of a new cyst from the rest of the lamina of another tooth which is the remnants of the epithelium or satellite cyst (daughter cyst) at the bone margin.¹⁴ Woolgar et al. also stated that removing incomplete cyst layers and new cysts originating from satellite cysts or remnants of odontogenic epithelium that were left behind or not removed during surgery could trigger recurrences.¹⁵ Presence of vital teeth in the lesion area that was not removed during the surgical procedure can also cause recurrences.^{1,16}

So far, Research on OKC recurrence has focused on the methods of management performed, but research on OKC recurrence associated with recurrence factors is still lacking. Thus, the authors are interested in conducting a literature study that analyzes the potential recurrence factors that most often occur after odontogenic keratocyst surgical procedures by performing a rapid review. This study aims to determine the factors that influence the potential for recurrence of odontogenic keratocyst, which most often occurs after surgery.

2. Material and Methods

This research was conducted using the rapid review method, which refers to the PRISMA guidelines (Preferred Reporting Items for Systematic Review and Meta-Analysis).¹⁷ This study was initiated by formulating research questions using PICO: (1) Population: patients with OKC recurrence who have undergone surgical treatment; (2) Intervention: surgical treatment; (3) Comparison: non-surgical treatment; and (4) Outcome: recurrence potential. The research was conducted from August to November 2022. The databases used in this research were PubMed ScienceDirect, and Google Scholar.

2.1. Inclusion and Exclusion Criteria

The inclusion criteria in this study were articles having data on odontogenic keratocyst recurrence after surgical treatment, in English, published within the last ten years (2012-2022), and using an observational study type with all types of study designs (cohort, cross-sectional, and case-control). Incomplete articles (only contain abstracts or discussions), cannot be accessed or locked, articles with a literature review study design, and articles with non-human subjects (in vitro, in vivo in experimental animals) are excluded.

2.2. Search Strategy and Study Selection

Searching for articles in this study uses "Boolean Operators," which is a search method that combines the words "OR", "AND", and "NOT" as synonyms. Searches were conducted through the PubMed, ScienceDirect, and Google Scholar databases, using keywords that included odontogenic keratocyst: "(keratocystic AND odontogenic AND tumor)" OR "(odontogenic AND keratocyst)" and recurrence potential: "(potential OR probability) AND "(recur OR relapse OR recurrence). Filters for

the year of publication (2012 – 2022) are used in each database, as well as additional filters for types of articles in ScienceDirect, research articles, and in PubMed, observational studies. All identified article results from the three databases were then selected manually. The first step is to check for duplicates and eliminate duplicate articles. Next is filtering for articles with irrelevant titles and abstracts, then a second screening for articles that cannot be accessed in full text or locked. Next, get the articles to be read in its entirety.

2.3. Data Extraction and Risk of Bias Assessment

Data extraction included the author's name, year of publication, journal name, type of study design, study period, number of odontogenic keratocyst cases, gender, average age, type of treatment, follow-up (months), number of recurrences, as well as the potential for replication. The quality assessment of the articles included in this rapid review study was assessed using the JBI Critical Appraisal Tools, which were adjusted to the research design of each article. To determine each article, there are 8-11 questions with the answers "Yes", "No", or "Unclear". One person carried out this risk assessment of bias by scoring 1 for each question with a "Yes" answer and a score of 0 for the other answers. The risk of bias from articles is categorized based on the total score range of "Yes" responses. Articles with a total score range of "Yes": (1) 1-4 have a high risk of bias, (2) 5-8 have a moderate risk of bias, and (3) 9-11 have a low risk of bias.¹⁸

3. Result and Discussion

3.1. Study Selection

The results of the article search process found 1,497 articles identified from PubMed (n = 2), ScienceDirect (n = 225), and Google Scholar (n = 1,270). Next, 335 articles were omitted due to duplication checks, so 1,162 articles were obtained. After the first screening was done by reading the title and abstract, 15 relevant articles were obtained. The second screening is done by reading the contents of the entire text. A total of 2 articles were omitted because; of review articles (n = 1), and not in English (n = 1), so 13 articles were obtained for review. The process of finding and selecting articles to be included in this study is summarized in **Figure 1**.

3.2. Description of The Study Characteristics

The total number of articles reviewed in this rapid review totaled 13 articles. The characteristics of the studies reviewed are summarized in **Table 1**. A total of thirteen articles had a retrospective cohort study design.^{19,20,29-31,21-28} A total of four articles were conducted in Brazil^{19,23,26,30}, two articles in China^{25,27}, two articles in Turkey^{20,22}, and five other articles each conducted in America³¹, Norway²⁹, Saudi Arabia²⁸, Iran²⁴, and Poland²¹. The highest number of OKC cases was reported by Fidele et al.²⁷ 565 cases, and the lowest number of OKC cases was reported by Cunha et al.³⁰ 24 cases. Meanwhile, the highest number of recurrences, namely 59 cases, was reported by Fidele et al.²⁷, and the lowest number of recurrences was reported by Ege et al.²⁰ with 7 cases.

Odontogenic keratocyst recurrence post-surgery cases distribution is summarized in **Table 3**. Twelve articles reviewed^{19-29,31} reported OKC recurrence distribution post-surgery to male and female gender comparisons. Daroit et al.²⁶ and Kaczmarzyk et al.²¹ reported quite large comparisons between men and women, 3.5:1 and 3:1. Thirteen articles,¹⁹⁻³¹ reported OKC recurrence distribution by age, which the lowest reported average age was 18.8 years, and the highest reported average age was 48 years. Twelve articles^{19-27,29-31} reported the type of surgical treatment performed before recurrence. Enucleation without combination is the most frequently performed type of surgical treatment (n = 91). Sites of recurrence are reported by nine articles reviewed.^{20-22,24-29} Mandible is the most reported site of recurrence (n = 139). Seven articles^{20,21,23-25,27,28} reported the reproduction shape, with multilocular lesions recorded as the most common form of the lesion (n = 92). Eleven articles²¹⁻³¹ reported the mean follow-up period of the patients. The most extended average follow-up period reported by Kaczmarzyk et al.²¹ was 101,88 months.

The results of the risk of bias assessment using the Joanna Briggs Institute (JBI) Critical Appraisal presented in **Table 2** showed 7 cohort retrospective articles had a moderate risk of bias and 6 cohort retrospective articles had a low risk of bias. None of those articles gained a high score based on the JBI assessment. The range score of all articles is between six and nine.

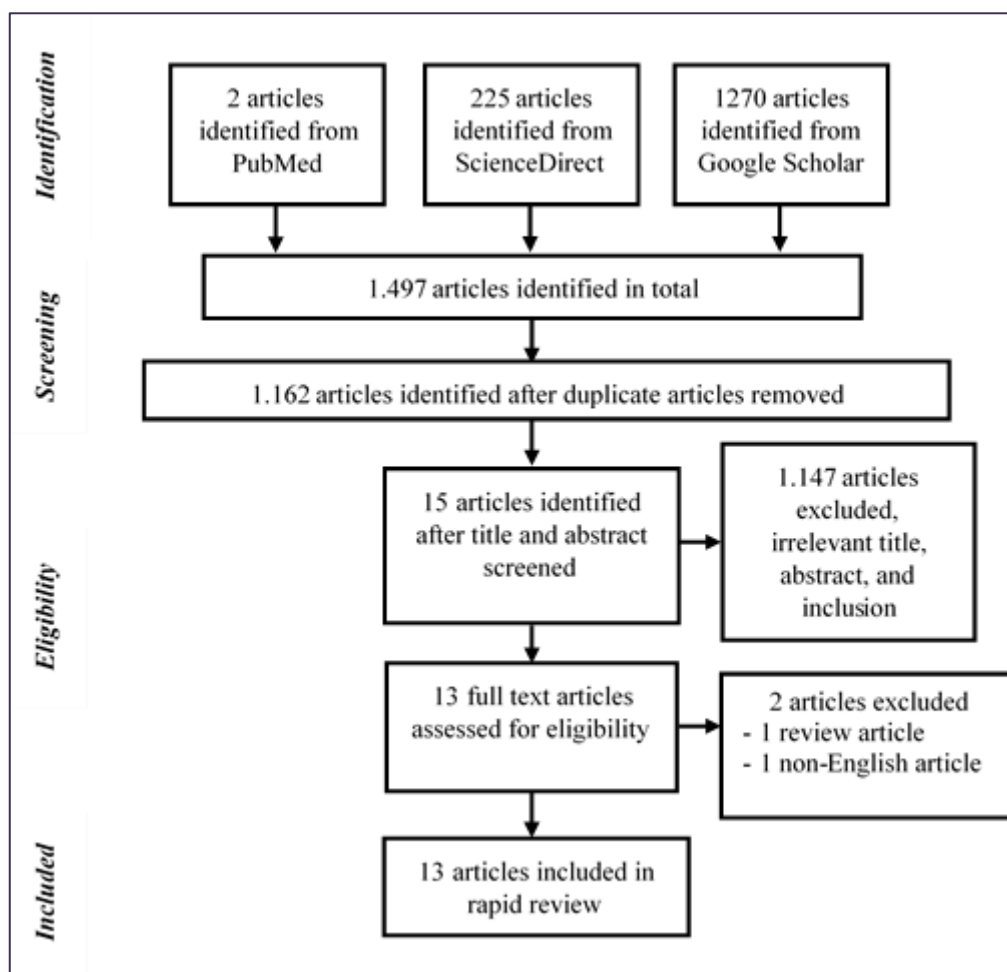


Figure 1.. PRISMA Flow Diagram Article Search Results

Based on Table 1, in 1,263 cases of odontogenic keratocyst, there were 253 cases of recurrence. It indicates a relatively high risk of recurrence in cases of odontogenic keratocyst. Odontogenic keratocyst recurrence is more common in male patients than in females.^{20–22,25–29,31} However, there was no significant relationship between gender and recurrence. In addition, the patient's age is an essential factor affecting recurrence. Most articles report that recurrences are more common in the third and fourth decades of life.^{19–21,23–25,27–31} The age factor is closely related to recurrence. Berge et al.²⁹ showed an exciting finding, patients with certain health conditions (such as asthma, hypertension, or heart disease), had a 2.73 times higher risk of recurrence than patients with good health conditions. However, no further association was found between health conditions and clinical findings.

The multilocular form of the lesion is associated with a high potential for recurrence. Tabrizi et al.²⁴ demonstrated a significant difference in recurrence between the lesion's multilocular ($n = 23$) and unilocular ($n = 3$) forms. Kaczmarzyk et al.²¹ and de Franca et al.²³ also showed a higher recurrence rate in the multilocular form of the lesion. Furthermore, Fidele et al.²⁷ also noted that the multilocular form of the lesion is associated with a high recurrence rate. Kinard et al.³¹ reported a high prevalence of unilocular lesions but a 4.7-fold higher recurrence potential for multilocular lesions. More extensive lesions (more than 4 cm in diameter), according to Tabrizi et al.²⁴ and Kaczmarzyk et al.²¹ also have a high recurrence potential with a poor prognosis. Recurrence with the multilocular form of the lesion is also frequently associated with Nevroid Basal Cell Carcinoma Syndrome (NBCCS).^{8,32}

Table 1. Study Characteristics Reviewed

No	Study (Year)	Location	Journal	Title	Study Period	Study Design	OKC Cases	OKC Case Recurrences	Recurrence percentage (%)
1	Akay et al. ²² (2015)	Turki	Ege Journal of Medicine	Keratocystic odontogenic tumor: A retrospective study of 64 cases	2006-2014	Cohort retrospective	64	9	14.1%
2	Cunha et al. ³⁰ (2016)	Brazil	Oral Surgery, Oral Medicine, Oral Pathology, and Oral Radiology	Clinicopathologic features associated with recurrence of the odontogenic keratocyst: a cohort retrospective analysis	2002-2012	Cohort retrospective	24	8	33%
3	Daroit et al. ²⁶ (2018)	Brazil	Journal of oral and maxillofacial surgery: official journal of the American Association of Oral and Maxillofacial Surgeons	Does Surgical Fragmentation of Odontogenic Keratocyst Capsule Interfere With the Recurrence Rate?	1991-2013	Cohort retrospective	34	9	26.4%
4	Ege et al. ²⁰ (2019)	Turki	Annals of Medical Research	Clinical and radiographic features of odontogenic keratocysts and evaluation of recurrence rates after treatment: A retrospective study	2015-2018	Cohort retrospective	41	7	17.1%
5	Kinard et al. ³¹ (2019)	Amerika	Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology	How well do we manage the odontogenic keratocyst? A multicenter study	2001-2013	Cohort retrospective	231	42	18.18%
6	de França et al. ²³ (2021)	Brazil	European Archives of Oto-Rhino-Laryngology	Recurrence-free survival and prognostic factors of odontogenic keratocyst: a single-center retrospective cohort	2007-2019	Cohort retrospective	40	18	45%
7	Borges et al. ¹⁹ (2021)	Brazil	Advances in Oral and Maxillofacial Surgery	Retrospective study of therapeutic approaches, recurrence and prevalence of cases of odontogenic keratocysts at a general hospital	2014-2020	Cohort retrospective	39	8	20.5%
8	Berge et al. ²⁹ (2016)	Norwegia	Oral Surgery, Oral Medicine, Oral Pathology and Oral Radiology	Pattern of recurrence of nonsyndromic keratocystic odontogenic tumors	1975-2012	Cohort retrospective	92	27	29.3%
9	Bello. ²⁸ (2016)	Arab Saudi	Medicina Oral, Patologia Oral y Cirugia Bucal	Keratocystic odontogenic tumor: A biopsy service's experience with 104 solitary, multiple and recurrent lesions	1984-2014	Cohort retrospective	104	9	8.7%
10	Zhao et al. ²⁵ (2016)	China	Journal of Dentomaxillofacial Radiology	Recurrent keratocystic odontogenic tumours: report of 19 cases	1990-2008	Cohort retrospective	257	19	7.4%

No	Study (Year)	Location	Journal	Title	Study Period	Study Design	OKC Cases	OKC Case Recurrences	Recurrence percentage (%)
11	Tabrizi et al. ²⁴ (2014)	Iran	Journal of Craniofacial Surgery	Correlation of radiographic features and treatments with the frequency of recurrence in odontogenic keratocysts of the mandible	2005-2011	Cohort retrospective	91	26	28.6%
12	Kaczmarzyk et al. ²¹ (2018)	Polandia	Clinical oral investigations	Investigation of clinicopathological parameters and expression of COX-2, bcl-2, PCNA, and p53 in primary and recurrent sporadic odontogenic keratocysts	1997-2015	Cohort retrospective	41	12	29.3%
13	Fidele et al. ²⁷ (2019)	China	Medicina oral, patologia oral y cirugia bucal	Recurrence of odontogenic keratocysts and possible prognostic factors: Review of 455 patients	2003-2015	Cohort retrospective	565	59	10.4%

Table 2. JBI risk of bias assessment of synthesized articles

Study	Q1 ^a	Q2	Q3	Q4	Q5	Q6	Q7	Q8	Q9	Q10	Q11	Score	Risk of Bias ^b
Akay et al. ²²	✓	?	✓	✓	x	✓	✓	✓	✓	x	✓	8	Moderate
Cunha et al. ³⁰	✓	?	✓	✓	x	✓	✓	✓	✓	✓	✓	9	Low
Daroit et al. ²⁶	✓	?	✓	✓	x	✓	✓	x	✓	x	✓	7	Moderate
Ege et al. ²⁰	✓	✓	✓	✓	✓	✓	✓	?	?	?	✓	8	Moderate
Kinard et al. ³¹	✓	?	✓	✓	x	✓	✓	x	✓	✓	✓	7	Moderate
de França et al. ²³	✓	?	✓	✓	x	x	✓	✓	✓	x	✓	7	Moderate
Borges et al. ¹⁹	✓	?	✓	✓	x	✓	✓	✓	✓	x	x	6	Moderate
Berge et al. ²⁹	✓	?	✓	✓	x	✓	✓	✓	✓	✓	✓	9	Low
Bello. ²⁸	✓	?	✓	✓	x	x	✓	x	✓	✓	✓	7	Moderate
Zhao et al. ²⁵	✓	?	✓	✓	x	✓	✓	✓	✓	✓	✓	9	Low
Tabrizi et al. ²⁴	✓	✓	✓	✓	x	x	✓	✓	✓	✓	✓	9	Low
Kaczmarzyk et al. ²¹	✓	?	✓	✓	x	✓	✓	✓	✓	✓	✓	9	Low
Fidele et al. ²⁷	✓	✓	✓	✓	x	x	✓	✓	✓	✓	✓	9	Low

Note : ^aQ1 – Q10 are questions 1 to 11 for articles with a cohort study based on Joanna Briggs Institute (JBI) Critical Appraisal. ^bThe risk of bias is categorized based on total amount of "yes" answers, which: 1) 1-4 : High; 2) 5-8 : Moderate; 3) 9-11 : Low. "✓" indicate yes, "x" indicate no, "?" indicate unclear.

Table 2 Potential Recurrence OKC Post-surgery Case Distribution

Study (Year)	Recurrence Case	Gender (M:F)	Age (x)	Treatment	Site	Shape	Follow-up (months) (x)	Recurrence Potential
Akay et al. ²² (2015)	9	1,25:1	20-60 (18,8)	Enucleation (n = 4) Marsupialization (n = 5)	Mandible (n = 9)	N/A	57,12	Mandible is the most recurrence site. A daughter cyst related to recurrence was found in 4 cases. Rapid follow-up can minimize the second surgery,
Cunha et al. ³⁰ (2016)	8	N/A	8-80 (32,1)	Decompression + Enucleation (n = 8)	N/A	N/A	60,5	Based on the microscopic appearance, recurrence is associated with residual tooth germs and epithelial islands in the cyst capsule. Radiographically there is residual tooth germ which causes lesions between the roots of the teeth.
Daroit et al. ²⁶ (2018)	9	3,5:1	6-63 (22,2)	Enucleation (n = 6) Enucleation + cryotherapy (n = 3)	Mandible (n = 7) Maxilla (n = 2)	N/A	21,6	Residual cyst capsule fragments do not affect recurrence.

Study (Year)	Recurrence Case	Gender (M:F)	Age (x̄)	Treatment	Site	Shape	Follow-up (months) (x̄)	Recurrence Potential
Ege et al. ²⁰ (2019)	7	1,4:1	13-78 (36,68)	Enucleation (n = 5) Decompression (n = 1) Decompression + Enucleation (n = 1)	Mandible (n = 6) Maxilla (n = 1)	Unilocular (n = 4) Multilocular (n = 3)	N/A	There was no significant difference in recurrence between unilocular and multilocular lesions.
Kinard et al. ³¹ (2018)	42	1,18:1	39	Enucleation (n = 12) Enucleation + Peripheral ostectomy (n = 13) Decompression (n = 17)	N/A	N/A	18	Remaining cysts left behind from decompression increase the potential for recurrence.
de França et al. ²³ (2021)	18	1:1,3	34,7	Enucleation + Carnoy's solution/curettage (n = 12) Ostectomy + Carnoy's solution (n = 6)	N/A	Unilocular (n = 8) Multilocular (n = 10)	81,32	Satellite cysts have the potential for recurrence.
Borges et al. ¹⁹ (2021)	8	1:1,12	21-40 (33,1)	Enucleation + curettage (n = 3) Enucleation + Peripheral ostectomy (n = 3) Decompression + Enucleation + ostectomy (n = 2)	N/A	N/A	N/A	Post-enucleation recurrence is higher.
Berge et al. ²⁹ (2016)	27	1,8:1	48	Enucleation (n = 23) Marsupialization (n = 4)	Mandible (n = 24) Maxilla (n = 3)	N/A	53	Mandible perforation, a thin, keratinized cyst capsule, increases the potential for recurrence. Patients with unfavorable health conditions (asthma, hypertension, and heart disease), have a higher recurrence potential.
Bello. ²⁸ (2016)	9	2:1	18-55 (30,7)	N/A	Mandible (n = 9)	Unilocular (n = 9)	26	Satellite cyst in unilocular cyst has no significant effect on recurrence potential.
Zhao et al. ²⁵ (2016)	19	1,71:1	18-45 (30,5)	Enucleation (n = 12) Enucleation + Carnoy's solution (n = 7)	Mandible (n = 15) Maxilla (n = 4)	Unilocular (n = 10) Multilocular (n = 9)	53,2	Recurrence was related to residual tooth germ at the involved site (not significant).
Tabrizi et al. ²⁴ (2014)	26	1:1,6	27,55	Enucleation (n = 17) Marsupialization (n = 9)	Mandible (n = 26)	Unilocular (n = 3) Multilocular (n = 23)	55,08	Multilocular lesions have a higher recurrence potential than unilocular lesions. Radiographic appearance can predict recurrence.
Kaczmarczyk et al. ²¹ (2018)	12	3:1	39	Enucleation (n = 12)	Mandible (n = 9) Maxilla (n = 3)	Unilocular (n = 5) Multilocular (n = 7)	101,88	3 out of 9 cases of recurrence, daughter cyst was found. Larger size of cyst, multilocularity, and cortical perforation increase the potential for recurrence.
Fidele et al. ²⁷ (2019)	53	1,18:1	7-81 (28,4)	Enucleation (n = 48) Marsupialization + Enucleation (n = 4) Resection (n = 1)	Mandible (n = 34) Maxilla (n = 19)	Unilocular (n = 22) Multilocular (n = 31)	62,4	The area of the tooth involved in the cyst area and not extracted, has a higher tendency for recurrence. Daughter cysts and multilocular lesions increase the chance of recurrence.

Mandible is the part of the jaw that has the highest recurrence rate. The recurrence rate of the mandible is 2.3 times higher than that of the maxilla, especially in the posterior aspect of the mandible.³¹ Recurrences in the posterior mandible are generally associated with unerupted teeth.²² The high recurrence is also caused by teeth around the lesion area that are not removed, causing recurrences. Fidele et al.²⁷, stated that teeth involved with the lesion or the lesion's location and not extracted had a higher tendency for recurrence. Cunha et al.³⁰ recommended the extraction of teeth with root involvement with OKC lesions. However, some patients choose to retain vital teeth, which can complicate complete enucleation and cyst removal.

Enucleation without combination was the most frequently performed surgical method (n = 91) and had the highest recurrence rate compared to other surgical treatment types. This is due to the difficulty of eradicating the cyst due to the cyst's thin layer, so the cyst's removal is incomplete.^{29,33,34} Borges et al.¹⁹, in their study, did not recommend enucleation without combination and curettage without combination in the choice of surgical treatment. This is due to the high recurrence rate of enucleation and curettage. The small sample in Borges et al.¹⁹ study showed that adding Carnoy's solution could reduce the potential for recurrence. This study also concluded that decompression and peripheral ostectomy in combination with Carnoy's solution had a lower recurrence potential than enucleation alone.

De Franca et al.²³, showed that marsupialization and decompression without a combination also has a high recurrence rate. Kinard et al.³¹, stated that residual cysts left after decompression could increase the potential for recurrence. In cases of OKC involving dentition, Zhao et al.²⁵ recommended marsupialization and/or decompression as the first stage of the surgical procedure before enucleation as the second stage. Cysts involving perforations of the alveolar bone are recommended for segmental or marginal resection. This surgery also has a meager recurrence rate, resulting in facial deformities and loss of masticatory function that usually require reconstructive surgery.^{25,35,36}

Incomplete removal of the cyst is associated with the presence of residual tooth germs and epithelial islands in the cyst lining.^{25,30} These residual tooth germs and epithelial islands can form daughter or satellite cysts which will enlarge and play a role in the potential for recurrence, especially in multilocular lesions.^{21-24,27} Residual of these daughter or satellite cysts leads to the formation of new cysts at the bony margins. The presence of a daughter or satellite cyst and the appearance of these epithelial islands can be a sign of more aggressive surgery. Bello et al.²⁸ stated that daughter or satellite cysts in unilocular lesions were found to have little potential for recurrence.²⁸ The follow-up period recorded they were ranged from 18 months to 100 months. Akay et al.²² recommended more rapid follow-up to detect recurrences early.²² Berge et al.²⁹ also recommended long-term follow-up. Follow-up is done by assessing clinical and radiographic results at 4, 6, and 8 years, followed by a range of follow-ups for another 3-4 years.²⁹

This research has several limitations. First, the study was only conducted by one reviewer. Furthermore, the language used in the articles was limited to English, and the types of articles obtained were limited to the retrospective cohort design. In addition, some of the articles reviewed have a moderate risk of bias. The result of this rapid review is that the potential for odontogenic keratocyst recurrence is multifactorial, with a recurrence percentage of 7.1% to 45%, with an average of 22.5%. Seven of the thirteen cases studied showed the presence of daughter or satellite cysts, residual tooth germ, and teeth in the lesion area that were not extracted, offering a high recurrence rate of odontogenic keratocyst. The multilocular cyst shape and large size can increase the potential for recurrence.

4. Conclusion

The results of this rapid review showed that the percentage of potential OKC recurrences ranged from 7.1% to 45% with an average recurrence rate of 22.5%. The presence of a daughter or satellite cyst as well as residual teeth or teeth in the area of the lesion that were not removed in the form of a large multilocular cyst, affects the potential for recurrence.

5. References

1. Cawson RA OE. Essential of Oral Pathology and Oral Medicine. Oral Pathology and Oral Medicine. 2017. 139, 149-154 p.

2. Rioux-Forker D, Deziel AC, Williams LS, Muzaffar AR. Odontogenic Cysts and Tumors. *Ann Plast Surg*. 2019 Apr;82(4):469–77.
3. Al-Moraissi EA, Dahan AA, Alwadeai MS, Oginni FO, Al-Jamali JM, Alkhutari AS, et al. What surgical treatment has the lowest recurrence rate following the management of keratocystic odontogenic tumor?: A large systematic review and meta-analysis. *J Cranio-Maxillofacial Surg*. 2017;45(1):131–44.
4. Bhargava D, Deshpande A, Pogrel MA. Keratocystic odontogenic tumour (KCOT)—a cyst to a tumour. *Oral Maxillofac Surg*. 2012 Jun 11;16(2):163–70.
5. Kshirsagar R, Bhende R, Raut P, Mahajan V, Tapadiya V, Singh V. Odontogenic keratocyst: Developing a protocol for surgical intervention. *Ann Maxillofac Surg*. 2019;9(1):152.
6. Gomes CC, Diniz MG, Gomez RS. Review of the molecular pathogenesis of the odontogenic keratocyst. *Oral Oncol*. 2009;45(12):1011–4.
7. Lench NJ, Telford EAR, High AS, Markham AF, Wicking C, Wainwright BJ. Characterisation of human patched germ line mutations in naevoid basal cell carcinoma syndrome. *Hum Genet*. 1997;100(5):497–502.
8. Bilodeau EA, Collins BM. Odontogenic cysts and neoplasms. *Surg Pathol Clin*. 2017;10(1):177–222.
9. Karaca C, Dere K, Er N, Aktas A, Tosun E, Koseoglu O, et al. Recurrence rate of odontogenic keratocyst treated by enucleation and peripheral ostectomy: Retrospective case series with up to 12 years of follow-up. *Med Oral Patol Oral y Cir Bucal*. 2018;0–0.
10. Slusarenko da Silva Y, Naclério-Homem M da G. A systematic review on the expression of bcl-2 in the nonsyndromic odontogenic keratocyst: should it be considered a cyst or a tumor? *Oral Maxillofac Surg*. 2020 Sep 3;24(3):277–82.
11. Abdullah WA. Surgical treatment of keratocystic odontogenic tumour: A review article. *Saudi Dent J*. 2011 Apr;23(2):61–5.
12. Andrić M, Brković B, Jurišić V, Jurišić M, Milašin J. Keratocystic odontogenic tumors—clinical and molecular features. *IntechOpen*; 2013.
13. Kim S-H, Oh M-S, Seo Y-S, Kim J-Y, Nam S-H, Lim S-M. Conservative Treatment of Multiple Keratocystic Odontogenic Tumors in a Young Patient with Nevroid Basal Cell Carcinoma Syndrome by Decompression: A 7-year Follow-up Study. *J Clin Pediatr Dent*. 2017 Jan 1;41(4):300–4.
14. Morgan TA, Burton CC, Qian F. A Retrospective Review of Treatment of the Odontogenic Keratocyst. *J Oral Maxillofac Surg*. 2005 May;63(5):635–9.
15. Woolgar JA, Rippin JW, Browne RM. A comparative study of the clinical and histological features of recurrent and nonrecurrent odontogenic keratocysts. *J Oral Pathol Med*. 1987;16(3):124–8.
16. Borg G, Persson G, Thilander H. A study of odontogenic cysts with special reference to comparisons between keratinizing and non-keratinizing cysts. *Sven Tandlak Tidskr*. 1974;67(6):311–25.
17. Moher D, Liberati A, Tetzlaff J, Altman DG. Preferred Reporting Items for Systematic Reviews and Meta-Analyses: The PRISMA Statement. *PLoS Med*. 2009 Jul 21;6(7):e1000097.
18. Goplen CM, Verbeek W, Kang SH, Jones CA, Voaklander DC, Churchill TA, et al. Preoperative opioid use is associated with worse patient outcomes after Total joint arthroplasty : a systematic review and meta-analysis. *BMC Musculoskelet Disord*. 2019;20(234):1–12.
19. Borges LBO, Almeida RS, Da Silva RA, Sato FRL. Retrospective study of therapeutic approaches, recurrence and prevalence of cases of odontogenic keratocysts at a general hospital. *Adv Oral Maxillofac Surg* [Internet]. 2021;2:100047. Available from: <https://www.sciencedirect.com/science/article/pii/S2667147621000340>
20. Ege B, Demirkol M, Yalcin M, Alan H. Clinical and radiographic features of odontogenic keratocysts and evaluation of recurrence rates after treatment: A retrospective study. *Ann Med Res*. 2019;26(9).
21. Kaczmarzyk T, Kisielowski K, Koszowski R, Rynkiewicz M, Gawełek E, Babiuch K, et al. Investigation of clinicopathological parameters and expression of COX-2, bcl-2, PCNA, and p53 in primary and recurrent sporadic odontogenic keratocysts. *Clin Oral Investig*. 2018;22(9):3097–106.
22. Akay C, Tetik A, ZEYTİNOĞLU M. Keratocystic odontogenic tumor: A retrospective study of 64 cases. *Ege J Med*. 2015;54(2):59–64.
23. de França GM, da Silva LBA, Mafra RP, da Silva WR, de Lima KC, Galvão HC. Recurrence-free survival and prognostic factors of odontogenic keratocyst: a single-center retrospective cohort. *Eur Arch Oto-Rhino-Laryngology*. 2021;278(4):1223–31.
24. Tabrizi R, Omid M, Dehbozorgi M, Hekmat M. Correlation of radiographic features and treatments with the frequency of recurrence in odontogenic keratocysts of the mandible. *J Craniofac Surg*. 2014;25(5):e413–7.
25. Zhao Y, Liu B, Cheng G, Wang SP, Wang YN. Recurrent keratocystic odontogenic tumours: report of 19 cases. *Dentomaxillofac Radiol*. 2012;41(2):96–102.
26. NB D, R da RV, Visioli F, FDM M, MG de O, PV R, et al. Does Surgical Fragmentation of

- Odontogenic Keratocystic Capsule Interfere With the Recurrence Rate? *J Oral Maxillofac Surg*. 2018 Apr;76(4):770–4.
27. 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. *Med Oral Patol Oral Cir Bucal*. 2019;24(4):e491.
 28. Bello I-O. Keratocystic odontogenic tumor: A biopsy service's experience with 104 solitary, multiple and recurrent lesions. *Med Oral Patol Oral Cir Bucal*. 2016;21(5):e538.
 29. Berge TI, Helland SB, Sælen A, Øren M, Johannessen AC, Skartveit L, et al. Pattern of recurrence of nonsyndromic keratocystic odontogenic tumors. *Oral Surg Oral Med Oral Pathol Oral Radiol*. 2016;122(1):10–6.
 30. Cunha JF, Gomes CC, de Mesquita RA, Andrade Goulart EM, de Castro WH, Gomez RS. Clinicopathologic features associated with recurrence of the odontogenic keratocyst: a cohort retrospective analysis. *Oral Surg Oral Med Oral Pathol Oral Radiol*. 2016;121(6):629–35.
 31. Kinard B, Hansen G, Newman M, Dennis P, Haeffs T, Perez S, et al. How well do we manage the odontogenic keratocyst? A multicenter study. *Oral Surg Oral Med Oral Pathol Oral Radiol* [Internet]. 2019;127(4):282–8. Available from: <https://www.sciencedirect.com/science/article/pii/S2212440318312975>
 32. 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 Imaging*. 2018;9(5):883–97.
 33. Hupp JR. *Contemporary Oral and Maxillofacial Surgery* 7th. Vol. 7, Elsevier. 2019. 478–488 p.
 34. Johnson NR, Batstone MD, Savage NW. Management and recurrence of keratocystic odontogenic tumor: a systematic review. *Oral Surg Oral Med Oral Pathol Oral Radiol*. 2013 Oct;116(4):e271–6.
 35. Ebenezer V, Balakrishnan R, Sargunar B. Surgical Treatment of Odontogenic Keratocyst Tumour: A Review Article. *Biomed Pharmacol J*. 2014 Jun 30;7(1):257–61.
 36. Blanas N, Freund B, Schwartz M, Furst IM. Systematic review of the treatment and prognosis of the odontogenic keratocyst. *Oral Surgery, Oral Med Oral Pathol Oral Radiol Endodontology*. 2000 Nov;90(5):553–8.

Authors Contribution

Contribution	Widyandhika CA	Anggraini JA	Yusuf HY
Concepts or ideas	√	√	√
Design	√	√	√
Definition of intellectual content	√		
Literature search	√	√	√
Experimental studies	√		
Data acquisition	√	√	√
Data analysis	√		
Statistical analysis		√	√
Manuscript preparation	√	√	√
Manuscript editing	√	√	√
Manuscript review	√	√	√



JDS (Journal of Syiah Kuala Dentistry Society) is an Open Access Journal licensed under a Creative Commons Attribution-ShareAlike 4.0 International License. This license authorizes the utilization, replication, modification, distribution, and reproduction of the article in any medium or format, provided that due credit is given to the original Author (s) and the source, a link to the Creative Commons license is provided, and any alterations made to the article are duly indicated.

Citation Format: Widyandhika CA, Anggraini JA, Yusuf HY. Potential recurrence of odontogenic keratocyst post-surgery, rapid review. J Syiah Kuala Dent Soc. 2023; 8(1): 92-102.

Publisher's Note: The authors of this article assert that all claims made herein are exclusively their own and may not necessarily reflect the views of their respective affiliated institutions or those of the publisher, editors, and reviewers. The publisher does not provide any guarantee or endorsement for any product subject to evaluation in this article or any claim made by its manufacturer.

Widyandhika CA, Anggraini JA, Yusuf HY. Potential recurrence of odontogenic keratocyst post-surgery, rapid review