Review Article

A systematic review on odontogenic cysts and tumours

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Abstract

Background: There are still certain gaps in the research that need to be filled despite the fact that numerous studies have looked into the transformation of odontogenic cysts into neoplastic lesions. To identify pertinent research that had been published and to synthesise the available data and provide an overview of the current body of knowledge, this review also sought to do so.

Materials and Methods: Adopting the Preferred Reporting Items of Systematic Reviews and Meta-Analyses (PRISMA) guidelines, a search strategy was implemented across several online databases to search for relevant articles as per the defined selection criterion.

Results: After the search strategy was complete, 31 studies were chosen. Men tended to have more cancer than women. Swelling and discomfort were the primary pathology-related complaints. Although two cases were not detailed, radiologically, well-defined and poorly defined borders were reported in 18 and 11 participants, respectively. Squamous cell carcinoma with good differentiation (n = 12) was the most common cancer kind. More than 74% of patients were still living 6 months to 10 years following follow-up, four (12.90%) experienced recurrences and/or metastases and two (6.45%) experienced a disease-related mortality between 2 months and a year.

Conclusion: Prompt surgical follow-ups and cautious excision of odontogenic cysts are essential to avoiding neoplastic change and recurrence. Future research is required to look at possible reasons why odontogenic cysts can convert neoplastically.

Keywords: Neoplasm, neoplastic cell transformation, OKC

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INTRODUCTION

Several research works have documented the prevalence of odontogenic cysts and tumours, concentrating on the more severe conditions, such as ameloblastoma and keratocystic odontogenic tumours (KCOT). Odontogenic lesions can be distinguished between cysts (a diseased cavity lined with epithelium) and tumours (a solid tissue mass, not always malignant). On radiographic imaging, intrabony lesions of the jaws are most frequently found accidentally. A prolonged inflammatory process in the bone of the surrounding root apex and a proliferating epithelial

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remnant of Malassez cause a radicular cyst. Fluid expanding the dental follicle peri-coronally during development is the cause of dentigerous cysts.[1,2]

The dental lamina and other odontogenic epithelial sources are where KCOT develops. With a more aggressive growth pattern than other odontogenic pathoses, this lesion has a greater recurrence rate. Ameloblastomas can develop from any odontogenic epithelium, including the dental lamina (during pre-odontogenesis), epithelial rests of Malassez and Serres (during post-eruption), reduced enamel epithelium (during post-odontogenesis) and possibly the basal layer of the overlying epithelium (during embryogenesis and pre-odontogenesis, the primitive source of dental lamina).[3] Therefore, it seems sense that ameloblastomas can appear differently on radiographs. An odontogenic myxoma is an intraosseous tumour composed of myxomatous fibrous extracellular matrix derived from remains of mesenchymal tissue.[4] The origin of adenomatoid odontogenic tumours is the dental lamina in the gubernacular cord of growing permanent teeth. Ameloblastic fibromas share with ameloblastomas a common origin in the enamel organ or dental lamina; however, the specimen does not contain any dental hard tissue.[5,6]

When radiographic transparencies are an accidental discovery on panoramic radiographs, it is crucial to know the frequency of odontogenic cysts and tumours so that patients can receive a fair assessment of their likely diagnoses and avoid over-emphasising rare but aggressive lesions.[7-9] The present systematic review was conducted to provide data for those odontogenic cysts transformed into neoplastic lesions. This is significant for all healthcare professional in providing a global incidence for their patients.

MATERIALS AND METHOD

Following the Preferred Reporting Items of Systematic Reviews and Meta-Analyses (PRISMA) statement and the Cochrane Collaboration's guidelines for systematic reviews, we planned and conducted our systematic review. A thorough computerised search of the published data was performed from the registry's inception until July 2023, covering research in PubMed (MEDLINE) and Scopus. The search terms "KOT," "OKC," "KERATOCYSTIC ODONTOGENIC TUMOR," OR "ODONTOGENIC KERATOCYST," and "malignant transformation" or "cancer" were used in both PubMed (MEDLINE) and Scopus. Furthermore, a manual search and screening were conducted to find further studies in the references given in the retrieved reports and relevant reviews.

Selection process

Articles that described an OKC with malignant transformation inside the cyst lining and included patient-related data were included. Articles were disqualified if they lacked a targeted outcome, had an English writing style or lacked a relevant diagnosis. Two investigators conducted the data extraction process independently, and disagreements were settled through conversation.

Study selection

From the first search, the writers found 836 papers, and they went over the abstracts. Thirty-one items were determined to meet the requirements for inclusion [Figure 1].

The demographic data (age, gender), clinical data (presenting symptoms, history of diagnosed OKC), radiologic assessment, histological investigation, treatment and patient status were the features gathered from the studies to perform the quantitative analysis.

RESULTS

The quality of the studies was assessed using the Cochrane Collaboration approach for assessing the risk of bias in randomised controlled trials (RCTs). The accuracy of the data analysis for this systematic review was ensured using the Newcastle-Ottawa Quality Assessment Form for cohort studies, the Cochrane Collaboration's tool for assessing the risk of bias, the Critical Appraisal Skills Programme, the Grading of Recommendations Assessment Development and Evaluation (GRADE) system for grading evidence and the Oxford Systematic Review Appraisal Sheet.

The results of the research have been given in a narrative overview. Three case series[10-12] and 28 case reports[13-17,10-18,18-61] were reported out of 31 pertinent publications. Figure 1 displays the flow chart of the chosen articles together with the primary justifications for their exclusion. Table 1 displays the primary attributes of the included studies. Cases in this study were compiled into case series, which were subsequently contrasted with other discovered series. It is noteworthy that nine KCOT cases were included in the research conducted by Chantravekin *et al*.[10] and that sixteen KCOT cases, three verrucous carcinoma cases and one spindle cell carcinoma case were included in the study published by Bodner *et al*.[11] Due to the lack of specific and extensive data in this research, none of them could be excluded from any of the categories listed in Table 1. Of the 31 instances that were recovered, men were more likely to have cancer. There was a 52.4-year-old man. Swelling and discomfort were the primary pathology-related

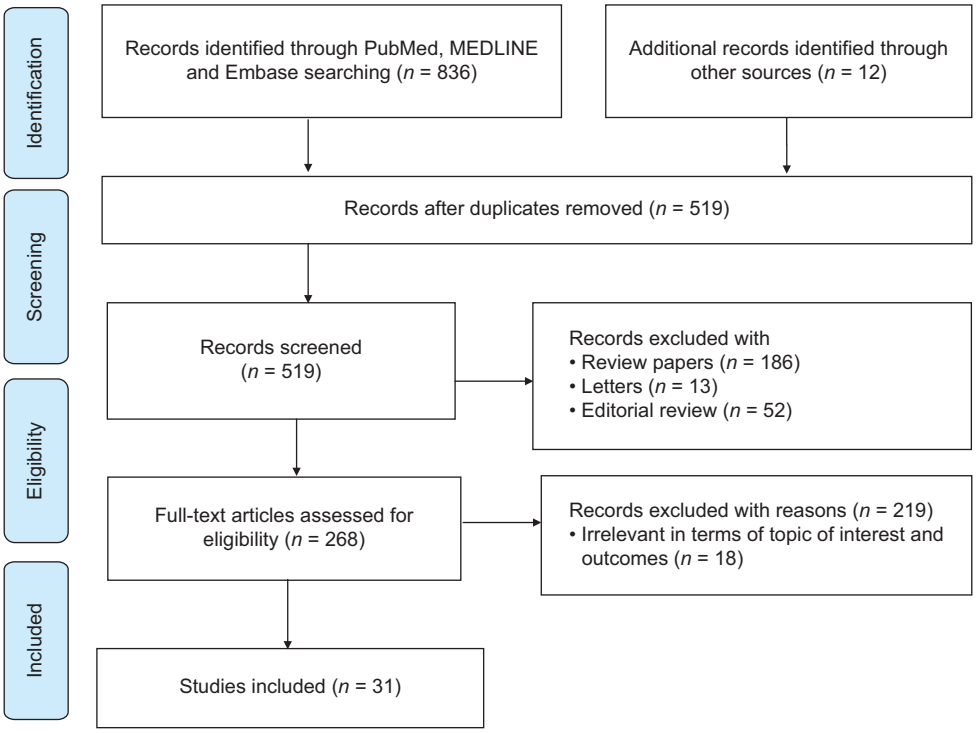

Figure 1: PRISMA flowchart of included studies

Table 1: Characteristics of the included studies

Variables	Present study (*n*=31)	Chantravekin *et al*.[10] (*n*=56)	Bodner *et al*.[11] (*n*=116)
Mean age (in years)	52.4	56.4	60.2
Gender			
Male	22	40	80
Female	9	16	36
Sign and symptoms			
Swelling	22	38	56
Pain	17	18	28
Lymphadenopathy	7	3	0
Sinus tract	6	0	0
Asymptomatic	4	6	13
Location			
Maxilla	6	17	24
Mandible	25	39	92
Radiologic findings			
Well-defined border	18		
Poorly defined border	11	Not available	Not available
Not cited	2		
Radicular resorption	1		
Cortical erosion	12		
IAN displacement	2		
Histopathology findings			
Well-differentiated SCC	12	0	53
Moderately differentiated SCC	3	0	47
Poorly differentiated SCC	2	0	8
Not specified	11	56	0
High-grade MEC	1	0	0
Moderately grade MEC	0	0	0
Low-grade MEC	1	0	0
Not specified	1	0	0
Outcome			
Alive	23	29	62% and 38% at 2 and 5 years, respectively
Recurrence/metastasis	4	0	0
Mortality	2	5	0
Not mentioned	2	15	0

SCC=squamous cell carcinoma, MEC=mucoepidermoid carcinoma

complaints. The maxilla (n = 6) was the least damaged region, whereas the mandible (n = 25) was the most. Although two cases were not detailed, radiologically, well-defined and poorly defined borders were reported in 18 and 11 participants, respectively. Squamous cell carcinoma with good differentiation (n = 12) was the most common cancer kind. More than 50% of the patients needed a neck lymphadenectomy, and a substantial percentage of them received treatment with a surgical excision of the lesion. Over 74% of patients remained alive during a time frame of 6 months to 10 years following follow-up; four patients (12.90%) experienced recurrences and/or metastases within a time frame of 1 to 5 years, and two patients (6.45%) lost their lives to the disease within a time frame of 2 months to 1 year. Regretfully, there was no mention of the status of the two patients. A table containing the information has been displayed [Table 1].

DISCUSSION

Odontogenic cysts arise from inflammation or development of the tooth-forming apparatus' epithelium.[62] The epithelial lining of odontogenic cysts originates from reduced enamel epithelium, remnants of Serres and Malassez epithelial cells and the epithelial cell remnants of Serres. Malassez's epithelial cell rest comes from Serres' surviving deteriorated dental lamina, which initiated tooth development during the sixth week of embryonic development. Reduced enamel epithelium covers the developing tooth crown.[63,64] The breakdown of the epithelial root sheath of a twig also contributes to the formation of dental roots. These remnants will all become embedded in the gingiva and bone. The epithelium that becomes lodged in the bone and gingiva can develop into malignant tumours and odontogenic cysts. A third of all neoplasms have their origin in odontogenic cysts.[65-68]

The neoplastic tumours that emerged from the odontogenic cysts were most likely caused by the epithelial remains of the cysts. Prolonged inflammation, continuous intracystic pressure and partial clearing of the cystic epithelium can cause odontogenic cysts to progress into neoplasms. Jain *et al*.[14] state that the formation of a sinus tract and pus discharge are two instances of odontogenic cyst long-lasting, persistent inflammation symptoms that are assumed to be cancerous. Chronic inflammation may be the source of gene instability in cells due to the production of reactive oxygen species (ROS). ROS are produced by cells, and when they mix with nitric oxide, they can create reactive nitrogen ions as a bridge. Superoxide, hydrogen peroxide and hydroxyl ions are examples of ROS. It is possible for these reactive nitrogen intermediates to initiate the carcinogenesis process by damaging proteins, deoxyribonucleic acid (DNA) and cell membranes. Furthermore, through inducing cell death, cytokine production, keratinisation of the cystic epithelium and aberrations in DNA, proteins and cell membranes, chronic inflammation encourages the transformation of healthy cells into malignant cells. An investigation by Borrás-Ferreres *et al*.,[66] however, demonstrated a neoplastic conversion from a follicular cyst without ongoing inflammation, suggesting the possibility of other physiopathological pathways linked to oncogenes. The exact cause of these modifications is still unknown. Some indicators that odontogenic cysts have advanced to neoplasms include swelling, pain and the presence of the sinus tract. Cystectomy, with or without tooth extraction, may also cause delayed recovery. Usually, radiographically, the early stages of malignant changes are invisible. Unilocular radiolucency, uneven, poorly defined scalloped edges and deteriorated osseous cortical bone, all of which indicate an invasive behaviour, will, however, set them apart. This study on 31 odontogenic cysts revealed that two cases developed into mucoepidermoid carcinoma and seventeen cases into squamous cell carcinoma. The origin of those neoplastic tissues were OKCs, residual cysts, radicular cysts, follicular cysts, calcifying odontogenic cysts and some other unidentified odontogenic cysts.

It was challenging to determine the origin of the cystic lesions because some odontogenic cysts were undetected because the transition was detected later than expected. It should be highlighted that regardless of the patients' age, the transformation was somewhat greater in men. Odontogenic cysts can also develop into neoplasms in younger people; a case report byIsshiki-Murakami *et al*. included an 18-year-old patient as an example.[69]

When a region grows quickly, it is important to take into consideration the malignant transformation of an odontogenic cyst, even though it may not be easily identified radiographically in the early stages.[49] When the osseous cortex is eroded, it typically manifests as an unilocular radiolucency with irregularly scalloped and poorly defined margins, suggesting invasive behaviour.[8-10,20,21,36,49-51,52] Comparable percentages of corticated and weakly defined edges were discovered in this investigation, however. Regretfully, the radiologic pattern was not specified in the remaining case series[10-12] that were included. Despite being a vital diagnostic tool, orthopantomography can be limited in its ability to diagnose some lesions due to image superposition or incomplete data regarding soft tissues.

Malignant lesions that are tiny and asymptomatic might occasionally be misinterpreted. Following a cystectomy, whether or not teeth are extracted, a delayed healing period may be a sign of cancer.[49,50] Nonetheless, there have been reports of certain malignant situations where the soft tissues have fully healed. Consequently, the entire material needs to be histologically analysed.[13,20,45,50] The prognosis is adversely affected by a delayed diagnosis.[42,51] According to the symptoms connected with the total number of instances, swelling, pain and the existence of a sinus tract were the most common associated symptoms. Based on the study's findings, it appears that most patients are still alive 10 years afterwards. In a similar vein, the case series written up by Chantravekin *et al*.[10] and Bodner *et al*.[11] showed greater rates of patient survival at 2 years—85.3% and 62%, respectively. However, we discovered five occurrences of metastasis and recurrence, which these studies did not disclose.

Treatment guidelines for odontogenic cysts that prevent cancer development were nonexistent.[70] Most of the articles we read advised treating odontogenic cysts cautiously, making sure to leave adequate margins.[71-73] In the instance of dentigerous cysts, the impacted tooth in question should also be excised immediately to prevent the residual cystic epithelium from becoming neoplasms. When the cyst is large, marsupialisation is recommended to prevent breaking or harm to important organs or tissues during enucleation, but most of the research indicates that the best course of treatment is cyst enucleation because marsupialisation raises the possibility that cystic cells may persist in the body and become neoplasms.[74-76]

In terms of the implications for research, larger population clinical studies are needed to significantly improve our knowledge of malignant changes brought on by odontogenic cysts.

CONCLUSION

For general practitioners and dentists, this systematic review emphasises the importance of a thorough clinical examination and medical history. Particularly in patients with a known malignant illness, signs of pain and paraesthesia in the face combined with radiographic evidence of radiolucency point to a possible preoperative diagnosis of a metastatic tumour.

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Conflicts of interest
There are no conflicts of interest.

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