# Mapping research over 50-years on odontogenic keratocyst: Global data and bibliometric profile analysis

Mapeamento da investigação ao longo de 50 anos sobre o queratocisto odontogénico: Análise global de dados e perfil bibliométrico

Cartografía de la investigación a lo largo de 50 años sobre el queratoquiste odontogénico: Datos globales y análisis del perfil bibliométrico

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#### **Abstract**

Oral and maxillofacial pathology is a traditional matter of study. The objective of this research is to perform a bibliometric analysis of odontogenic keratocyst over a length of fifth years. A bibliographic search (retrospective study) in line with the STROBE guidelines and the concepts of the Leiden Manifesto was performed on the Web of Science using the term "odontogenic keratocyst". A citation analysis as so authorship, and year of publication were performed. A graphical representation of the keywords was created with VOSviewer. These steps are essential to create this list and relate it to all published articles on the subject. A ranking was created in which the top 51 most frequently referred articles. Variables had been discussed individually. The USA leads the number of publications, followed by Brazil and England. A massive variety of publications refer to recurrence after treatment and its association with tumoral antigen expression. The importance of choosing suitable keywords was discussed. The bibliometric indicators validate records to evaluate the overall performance of studies productiveness and the quality of research output. This work provides a treasured reference for maxillofacial surgeons, oral pathologists, academics and researchers.

Keywords: Bibliometrics; Citation analysis; Scientometrics; Top-cited articles; Keratocyst.

#### Resumo

A patologia bucomaxilofacial é um assunto tradicional de estudo. O objetivo deste trabalho é realizar uma análise bibliométrica do queratocisto odontogênico durante um período de cinco anos. Foi realizada uma pesquisa bibliográfica (estudo retrospectivo) de acordo com as diretrizes STROBE e os conceitos do Manifesto de Leiden na Web of Science utilizando o termo "odontogenic keratocyst". Foi efetuada uma análise das citações quanto à autoria e ao ano de publicação. Foi criada uma representação gráfica das palavras-chave com o VOSviewer. Estes passos são essenciais para criar esta lista e relacioná-la com todos os artigos publicados sobre o tema. Foi criado um ranking com os 51 artigos mais citados. As variáveis foram discutidas individualmente. Os EUA lideram o número de publicações, seguidos do Brasil e da Inglaterra. Uma enorme variedade de publicações refere-se à recorrência após o tratamento e à sua associação com a expressão de antígenos tumorais. Foi discutida a importância da escolha de palavras-chave adequadas. Os indicadores bibliométricos validam os registos para avaliar o desempenho global da produtividade dos estudos e a qualidade dos resultados da investigação. Este trabalho constitui uma referência preciosa para cirurgiões maxilofaciais, patologistas orais, académicos e investigadores.

Palavras-chave: Bibliometria; História; Patologia bucal; Cistos odontogênicos.

#### Resumen

La patología oral y maxilofacial es una materia de estudio tradicional. El objetivo de este trabajo es realizar un análisis bibliométrico del queratoquiste odontogénico a lo largo de cinco años. Se realizó una búsqueda bibliográfica

(estudio retrospectivo) en línea con las directrices STROBE y los conceptos del Manifiesto de Leiden en la Web of Science utilizando el término "odontogenic keratocyst". Se realizó un análisis de las citas por autoría y año de publicación. Se creó una representación gráfica de las palabras clave con VOSviewer. Estos pasos son esenciales para crear esta lista y relacionarla con todos los artículos publicados sobre el tema. Se creó una clasificación en la que figuraban los 51 artículos más citados. Las variables se habían analizado individualmente. Estados Unidos encabeza el número de publicaciones, seguido de Brasil e Inglaterra. Una enorme variedad de publicaciones se refieren a la recurrencia tras el tratamiento y su asociación con la expresión de antígenos tumorales. Se discutió la importancia de elegir palabras clave adecuadas. Los indicadores bibliométricos validan los registros para evaluar el rendimiento global de la productividad de los estudios y la calidad de los resultados de la investigación. Esta obra constituye una valiosa referencia para cirujanos maxilofaciales, patólogos orales, académicos e investigadores.

Palabras clave: Bibliometría; Análisis de citas; Cienciometría; Artículos más citados; Queratoquiste.

### 1. Introduction

Oral and maxillofacial pathology is a common subject matter. Besides to some pathology journals, many articles are published in different journals including maxillofacial surgery, dentistry, otorhinolaryngology and medicine, amongst others. Oral and maxillofacial tumors and cysts are substantially studied worldwide.

Among the odontogenic cysts, the odontogenic keratocyst is one of the best studied. A crucial reason that explains this regularly studied pathology is the excessive rate of recurrence (Cakarer et al. 2018). Another important point concerning odontogenic keratocysts is their classification. The World Health Organization has used different classifications as tumor and cyst over the years (Soluk-Tekkesin et al. 2020). This classification has modified through the years and is likewise controversial withinside the literature.

A contemporary method of analyzing literature on a particular topic is to perform a bibliometric evaluation. Thus, the purpose of this research is to carry out a bibliometric analysis of odontogenic keratocysts, assessing diverse statistics available withinside the literature. A useful list of the top 51 most cited articles on the subject has been created. Bibliometrics may be very beneficial for researchers, academics, and scholars.

### 2. Methodology

The present bibliometric evaluation is a retrospective study that accompanied the Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) Statement (Vandenbroucke et al. 2014) and the standards of The Leiden Manifesto (Hicks et al. 2015). The information for this study has been taken from the Web of Science database. The following attributes have been used as data elements: citation analysis, contribution by country and author, source names, and productivity per year. No publication year and language restrictions have been applied.

All articles featuring odontogenic keratocyst have been screened. The search was further refined using the categories of country, author, affiliation, journal and year. Subsequently, all relevant works published withinside the Web of Science have been retrieved and examined according to the number of citations. Some types of research such as conference papers, letters, and abstracts have been excluded. The data has been analyzed on the same day to avoid the risk of bias.

The gathered information was transferred to Microsoft Excel. Information was compiled to retrieve bibliometric indicators. After refining the findings, all files and outcomes have been collected. All values retrieved from Web of Science have been collected and analyzed. Other analyzed variables were country and institution affiliation, publication language, Hirsch index (h-index), citation, and authorship. A beneficial list of the top 51 most cited articles has been created.

Statistical tests have been performed and considered significant only with a 95% confidence interval. Charts have been created with Microsoft Excel. The VOSviewer free software (Leiden University, The Netherlands) was used to create a graphical illustration of some vital elements, a visual form of bibliometric evaluation. The IRB ethics approval for the study was exempted due to the fact the information came from previous published art.

#### 3. Results and Discussion

An overall of 780 articles were retrieved, with 96 excluded based on study. The range of publications at the concern is on an upward trend (R2 = 0.7008). The large majority was published in English language (n = 759; 97.31%) followed by German (n = 10), French (n = 4), Spanish (n = 3), Italian (n = 2), Hungarian (n = 1) and Polish (n = 1). An overall of 13,213 citations were found with an average of 16.93 per article. The average citation of open access articles (n = 205) was 8.66, even as that of subscription articles (n = 575) averaged 19.89.

Pearson correlation tests were performed. There was a weak correlation between the year of publications and the number of articles (r = 0.3687) in regards to the number of citations (r = 0.1960). This discrepancy may be visible in a chart that groups this data (Figure 1). The citation density (citations divided by the number of articles) has fallen sharply over the last 10 years.

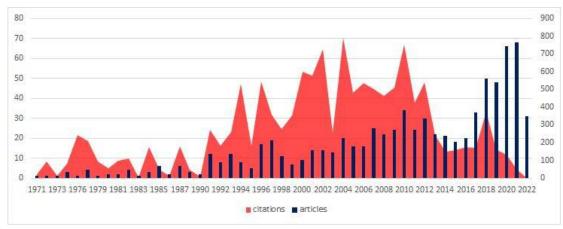


Figure 1 - Comparison between the number of articles (blue first axis) and citations (red secondary axis).

Source: Own authorship.

There were 77 nations and 298 distinctive institutions were represented withinside the publications. Countries from all inhabited continents were represented. The USA leads the number of publications (n = 148), followed by Brazil (n = 101) and England (n = 72). The University of São Paulo leads as institutional affiliation (n = 25), followed by The League of European Research Universities (n = 21) and Federal University of Minas Gerais (n = 19). Table 1 summarizes the primary ten examples of each.

Table 1 - Top ten countries and institutions with more publications on the topic and the number of publications (n).

	Countries	n		Institutions	origin	n
1	USA	148	1	Universidade de São Paulo	Brazil	25
2	Brazil	101	2	League of European Research Universities	Europe	21
3	England	72	3	Universidade Federal de Minas Gerais	Brazil	19
4	India	66	4	Harvard University	USA	18
5	Japan	64	5	University of London	England	15
6	China	49	6	Universidade Federal do Rio Grande do Norte	Brazil	14
7	Italy	47	7	Universitá Politecnica delle Marche	Italy	11
8	Turkey	33	7	Peking University	China	11
9	Germany	26	7	Universidade Estadual de Campinas	Brazil	11
10	South Korea	24	7	University of Sheffield	England	11
			7	Wuhan University	China	11

Source: Own authorship.

An overall of 2,854 authors were located. Professor Ricardo Santiago Gomez, from the Federal University of Minas Gerais is the more prolific author on odontogenic keratocyst (n = 15) with a total of 340 publications, an h-index of 34 and 6,590 citations. A list of the more prolific authors at the concern has been compiled (Table 2).

**Table 2 -** Top eleven more prolific authors on the topic with publications and number of citations on odontogenic keratocyst and career h-index.

author	origin	publications	Citations	H-index
Gomez, RS	Brazil	15	450	34
Li, TJ	China	14	487	20
Gomes, CC	Brazil	11	198	23
Rubini, C	Italy	10	155	21
Diniz, MG	Brazil	9	157	20
de Souza, LB	Brazil	8	79	18
Browne, RM	USA	7	294	30
High, AS	England	7	158	23
Matthews, JB	USA	7	252	48
Robinson, PA	USA	7	158	19
Santarelli, A	Italy	7	56	22

Source: Own authorship.

Out of 780 articles, 638 have at least one citation. The most cited article is "Relative incidence of odontogenic-tumors and oral and jaw cysts in a Canadian population" by Dale et al (Daley et al. 1994) from the University of Western Ontario, Canada. Prof. Dale has a total of 78 publications, 1,812 citations and an H-index of 24. A list of the 51 more cited publications was compiled (Table 3). Twenty-nine of these 51 articles were published more than 20 years ago (56.86%). The Pearson correlation between citation density and year of publications was negative (r = -0.4357).

Table 3 - Top 51 more cited publications on odontogenic keratocyst ranked according to the number of citations (cit).

Title	cit
Relative incidence of odontogenic-tumors and oral and jaw cysts in a Canadian population (Daley et al. 1994)	252
Odontogenic keratocyst (Brannon 1976)	243
Nevoid basal cell carcinoma (Gorlin) syndrome (Gorlin 2004)	220
Odontogenic keratocyst – clinicopathologic study of 312 cases. 2. Histologic features (Brannon 1977)	183
Odontogenic keratocyst: Review of 256 cases for recurrence and clinicopathologic parameters (Myoung et al. 2001)	164
Systematic review of the treatment and prognosis of the odontogenic keratocyst (Blanas et al. 2000)	159
The odontogenic keratocyst – a benign cystic tumor (Ahlfors et al. 1984)	150
Odontogenic cysts, odontogenic tumors, fibroosseous, and giant cell lesions of the jaws (Regezi 2002)	149
The aggressive nature of the odontogenic keratocyst: is it a benign cystic neoplasm? Part 2. Proliferation and genetic studies (Shear 2002b)	148
PTCH gene mutations in odontogenic keratocysts (Barreto et al. 2000)	147
Marsupialization as a definitive treatment for the odontogenic keratocyst (Pogrel and Jordan 2004)	142
The aggressive nature of the odontogenic keratocyst: is it a benign cystic neoplasm? Part 1. Clinical and early experimental evidence of aggressive behaviour (Shear 2002a)	133
A retrospective review of treatment of the odontogenic keratocyst (Morgan et al. 2005)	126
An analysis of oral and maxillofacial pathology found in children (Jones and Franklin 2006) over a 30-year period	125
Γreatment of large odontogenic keratocysts by decompression and later cystectomy - A long-term follow-up and a nistologic study of 23 cases (Marker et al. 1996)	122
New tumour entities in the 4th edition of the World Health Organization Classification of Head and Neck tumours: odontogenic and maxillofacial bone tumours (Speight and Takata 2018)	115
Recurrence of keratocysts and decompression treatment - a long-term follow-up of 44 cases (Brøndum and Jensen 1991)	115
Keratocystic odontogenic tumour: Reclassification of the odontogenic keratocyst from cyst to tumour (Madras and Lapointe 2008)	112
Nevoid basal cell carcinoma syndrome. Clinical findings in 37 Italian affected individuals (Muzio et al. 1999)	107
Developmental odontogenic cysts - an update (Shear 1994)	105
Γhe odontogenic keratocyst - orthokeratinized variant (Wright 1981)	94
Recurrence of the odontogenic keratocyst in relation to clinical and histological features - 20-year follow-up-study of 72 patients (Vedtofte and Prætorius 1979)	94
Analysis of clinical and histopathologic parameters of odontogenic keratocyst (Payne 1972)	94
Expression and induction of collagenases (MMP-8 and-13) in plasma cells associated with bone-destructive lesions (Wahlgren et al. 2001)	90
Epithelial-cell proliferation in odontogenic keratocysts - a comparative immunocytochemical study of ki67 in simple, recurrent and basal-cell nevus syndrome (BCNS)-associated lesions (Li et al. 1995)	90
Conservative treatment protocol of odontogenic keratocyst: A preliminary study (Maurette et al. 2006)	89
Malignant epithelial odontogenic tumors (Eversole 1999)	84
Dedifferentiation of odontogenic keratocyst epithelium after cyst decompression (August et al. 2003)	83
Odontogenic keratocyst: To decompress or not to decompress? A comparative study of decompression and enucleation versus resection/peripheral ostectomy (Kolokythas et al. 2007)	80
The primordial cyst (odontogenic keratocyst) - its tumor-like characteristics and behavior (Partridge and Towers 1987)	80
Primary intraosseous squamous cell carcinoma arising in an odontogenic cyst - a clinicopathologic analysis of 116 reported cases (Bodner et al. 2011)	79
Nevoid basal cell carcinoma syndrome: molecular biology and new hypotheses (Cohen 1999)	77
Expression of cell cycle and apoptosis-related proteins in sporadic odontogenic keratocysts and odontogenic keratocysts associated with the nevoid basal cell carcinoma syndrome (Lo Muzio et al. 1999)	73

The role of endotoxin and cytokines in the pathogenesis of odontogenic cysts (Meghji et al. 1996)  71 The aggressive nature of the odontogenic keratocyst: is it a benign cystic neoplasm? Part 3. Immunocytochemistry of cytokeratin and other epithelial cell markers (Shear 2002c)  A systematic review of the recurrence rate for keratocystic odontogenic tumour in relation to treatment modalities (Kaczmarzyk et al. 2012)  The Odontogenic Keratocyst: A Cyst, or a Cystic Neoplasm? (Li 2011)  69 Malignant odontogenic tumors: A 22-year experience (Goldenberg et al. 2004)  68 Cell proliferation markers in the odontogenic keratocyst: effect of inflammation (de Paula et al. 2000)  68 Management of odontogenic keratocysts of the jaws: A ten-year experience with 120 consecutive lesions (Pitak-Arnnop et al. 2010)  Cell proliferation, apoptosis and apoptosis-related factors in odontogenic keratocysts and in dentigerous cysts (Kichi et al. 2005)  Expression of proliferating cell nuclear antigen in ameloblastomas and odontogenic cysts (Piattelli et al. 1998)  66 A retrospective study of paediatric oral lesions from Thailand (Dhanuthai et al. 2007)  64 Conservative management of a large odontogenic keratocyst: Report of a case and review of the literature (Giuliani et al. 2006)  Characterization and management of the keratocystic odontogenic tumor in relation to its histopathological and biological features (Mendes et al. 2010)  Dentigerous cyst: a retrospective clinicopathological analysis of 2082 dentigerous cysts in British Columbia, Canada (Zhang et al. 2010)  Basal cell nevus syndrome (High and Zedan 2005)  62  The World Health Organization Classification of Odontogenic Lesions: A Summary of the Changes of the 2017 (4th) Edition (Soluk-Tekkeşin and Wright 2018)  Cystic Lesions of the Jaws - A Clinicopathological Study of 322 Cases and Review of the Literature (Manor et al. 2012)  Marsupialization inhibits interleukin-1 alpha expression and epithelial cell proliferation in odontogenic keratocysts		
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	Cystic Lesions of the Jaws - A Clinicopathological Study of 322 Cases and Review of the Literature (Manor et al. 2012)	61
	Marsupialization inhibits interleukin-1 alpha expression and epithelial cell proliferation in odontogenic keratocysts (Ninomiya et al. 2002)	61

Source: Own authorship.

A total of 286 distinctive journals were retrieved. Oral Surgery Oral Medicine Oral Pathology Oral Radiology was the journal with the greatest number of publications on this topic (n = 57). A list of the ten more productive journals was made (Table 4). A Pearson correlation test performed between the Scimago Journal Ranking (SJR) and the number of citations was considered very poor (r = 0.0401). The Pearson correlation between the number of publications and the SJR turned into negative (r = -0.2614).

**Table 4** - Top ten journals on quantity of publications (n) on odontogenic keratocyst ordered according to the number of citations.

Journal	SJR	n	%	cit	% cit
Oral Surgery Oral Medicine Oral Pathology Oral Radiology	0,570	57	7,31%	2,140	16,20%
Journal of Oral Pathology and Medicine	0,774	53	6,79%	1,351	10,22%
Journal of Oral and Maxillofacial Surgery	0,733	45	5,77%	1,338	10,13%
Oral Oncology	1,421	17	2,18%	837	6,33%
International Journal of Oral and Maxillofacial Surgery	0,975	22	2,82%	561	4,25%
Medicina Oral Patologia Oral y Cirugia Bucal	0,681	22	2,82%	312	2,36%
British Journal of Oral and Maxillofacial Surgery	0,572	15	1,92%	276	2,09%
Dentomaxillofacial Radiology	0,798	17	2,18%	232	1,76%
Oral Diseases	0,762	15	1,92%	217	1,64%
Journal of Craniofacial Surgery	0,436	24	3,08%	83	0,63%

Legend: cit – citations. Note: Oral Surgery Oral Medicine Oral Pathology Oral Radiology, Oral Surgery Oral Medicine Oral Pathology Oral Radiology and Endodontology were funded in one journal, as was the same journal that changed name over the years. Source: Own authorship.

An overall of 259 articles were case reports (n = 33.20%). A considerable number of publications mentioned histopathological features of the odontogenic keratocyst (n = 332, 42.56%). A more expressive number of articles were associated with tumor or neoplasm keywords (n = 482, 61.79%).

After disposing of word "humans", the most frequent keywords in the articles were "odontogenic cysts", "male", "female", "adult" and "middle aged". Figure 2 indicates a graphical illustration of the keywords using a network visualization made using the free software VOSviewer (Leiden University, Netherlands). VOSviewer is a device helpful to assemble and visualize bibliometric networks. The sizes of the circles are associated with the numbers of citations using the determined keywords.

temporal muscle dental implantation, endosseou loss of heterozygosity sensitivity and specificity mandible neoplasm recurrence, local osteotomy mutation gene expression regulation, ne mandibular condyle tomography, x-ray computed recurrence unoenzy**m**e techniques endoscopy maxilla Odontogenic paranasal sinus disease in situ hybridization radiography membrane glycoproteir rna, messenger periapical abscess child cuspid tooth eruption cyst fluid dna-binding proteins molar periodontal cyst periapical diseases child, preschool blacks granuloma, giant cell sex distribution VOSviewer

Figure 2 - Graphic analysis of MeSH keywords.

Source: Own authorship.

This paper aims to perform a bibliometric analysis of the odontogenic keratocyst, including a list of the top 51 most-cited articles. The bibliometric analysis does not intend to be precise (Hicks et al. 2015). Graphical data visualization through bibliometric software and statistical tests were performed to make the information more assimilable. Bibliometric analysis allows researchers and readers to identify the evolution of research in a given area over time and gain a better understanding of that area and some potential errors (Grillo 2021a, 2022; Martelli et al. 2021).

Citation density is the number of citations per publication year and is an essential measure for understanding the scholarly strength and impact of a particular work. Of the ten articles with a higher number of citations, five refer to clinicopathological (Brannon 1976, 1977; Ahlfors et al. 1984; Myoung et al. 2001; Regezi 2002), and two to genetic (Barreto et al. 2000; Shear 2002b) features of keratocysts. With the exception of one article from 2004 (Gorlin 2004), the other nine articles were published more than 20 years ago. These articles can be considered classicals on the subject. For some articles there is a statistically significant correlation of the citation density with the age of the publication (Ahmad et al. 2019; Arshad et al. 2020). This correlation was not found in this paper (r = -0.4357), the same lack of correlation found in another article (Grillo 2021b) on orthognathic surgery, a topic of maxillofacial surgery. Articles tend to be cited more often after 10 years of publication (p < 0.005).

A significant number of articles were published in high or intermediate ranked journals (n = 524, 67.17%). Majority of journals from the field of dentistry/oral surgery area (n = 458, 58.71%). Open access articles were cited less than subscription articles. No connection between journal status and number of citations or number of publications.

As in most other subjects, the USA leads the number of publications, followed by Brazil. Of the 10 most cited articles, four are by Brazilian researchers. This high number can be explained by a considerable number of Brazilian cases and publications on the topic (Servato et al. 2012, 2013; Jaeger et al. 2017). Of the eleven most cited authors, one is considered outstanding ( $n\geq40$ ), and eight as good ( $n\geq20$ ) regarding h-index (Hirsch 2005). H-index of publications was considered

outstanding (n = 56). This index varies according to different types of science. In oral pathology or maxillofacial surgery, this list can be considered as high impact. Despite some critics of the h-index, it is a practical measure of science impact. There is an increasing number of high-quality studies on odontogenic keratocysts. This data can be viewed by comparing total citations, h-index and citation density.

Concerns about the high recurrence index of odontogenic keratocysts can be seen. A massive number of publications evaluating the behavior of the lesion and its association with neoplasms is a constant in the literature. The World Health Organization even classified the odontogenic keratocyst as a tumor in their third edition of the Classification of Head and Neck Tumours. The odontogenic keratocyst was reclassified again as a cyst from the fourth edition (Speight & Takata 2018). A considerable number of articles on tumoral antigen expression and the odontogenic keratocyst can be found in the literature over the years (Slootweg 1995; Shear 2002c; Coşarcă et al. 2016; Slusarenko da Silva & Naclério-Homem 2020; Slusarenko da Silva et al. 2021).

Keywords are crucial to determine the thematic content of an article and to describe the content through a controlled vocabulary (USA: US National Library of Medicine National Institute of Health; Brozoski et al. 2022). The purpose of controlled vocabulary indexing is to facilitate search retrieval, thereby increasing the precision, sensitivity and efficiency of a bibliographic search (Baumann 2016). Network analysis of keywords through a graphical visualization is helpful for selecting an appropriate title and keywords, an effective method for further dissemination (Mondal et al. 2018).

A clear limitation of this study is the inability to use other databases. PubMed does not rely on citation analysis. Google Scholar uses citation analysis, but this can only be analyzed with other softwares. In addition, the citation analysis can be confounded by self-citations.

#### 4. Conclusion

This study was a comprehensive bibliometric analysis of studies on odontogenic keratocysts. Web of Science was used solely to collect data from relevant studies with no restriction on publication year. The bibliometric indicators validate information to assess the performance of research productivity and the quality of research output. This work provides a valuable reference for maxillofacial surgeons, oral pathologists, academics and researchers. Future studies are necessary as the science is very dynamic and this list needs to be updated from time to time. Inclusion of other databases could be helpful in highlighting dynamic sciences such as Maxillofacial Surgery and Pathology.

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