

## Trends in Cleft Lip and Palate Surgery: A Half-Century Bibliometric Analysis and Literature Review

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### ABSTRACT

**Introduction:** Cleft lip and palate (CLP) are significant congenital anomalies requiring comprehensive surgical and multidisciplinary care. This study conducts a bibliometric analysis of CLP surgery research to identify key authors, topics, and journals, highlighting collaboration trends and providing insights into current research and future directions.

**Methods:** The data was acquired online from Scopus on May 22<sup>nd</sup>, 2024 with a search spanning from 1974 to 2024, yielding 7,283 relevant documents. The publication output was then analyzed using VOSViewer to provide the visual representation.

**Results and Discussion:** The results indicate an increase in research studies over the years. Lo, L.J. emerged as the top researcher, with the United States leading in research output. Researchers primarily focused on three areas: surgical techniques, complications, and secondary procedures. Emerging areas such as patient quality of life, velopharyngeal insufficiency, and nonsyndromic clefting are gaining interest for future research.

**Conclusion:** Significant progress in CLP surgery has improved patient outcomes and quality of life over the past fifty years. Continued research and international collaboration are essential to address global disparities and ensure equitable access to advanced CLP care, further enhancing outcomes and quality of life for all patients.

**KEYWORDS:** cleft, cleft lip, cleft lip and palate, surgery, bibliometrics

### ARTICLE DETAILS

**Published On:**  
**14 June 2024**

**Available on:**  
<https://ijmscr.org/>

### I. INTRODUCTION

Cleft lip and palate (CLP) are among the most common congenital anomalies, affecting a significant number of newborns worldwide(1). The management of CLP has evolved considerably over the past fifty years, driven by advancements in surgical techniques, improved understanding of the etiology, and interdisciplinary approaches to patient care(2). This evolution has not only enhanced the functional and aesthetic outcomes for patients but also significantly improved their quality of life. Given the complex nature of CLP, which often requires multiple surgical interventions and long-term follow-up, it is crucial to understand the trends and milestones that have shaped current surgical practices.

A bibliometric analysis offers a comprehensive approach to mapping the progress and identifying pivotal moments in the surgical management of CLP. By analyzing a vast array

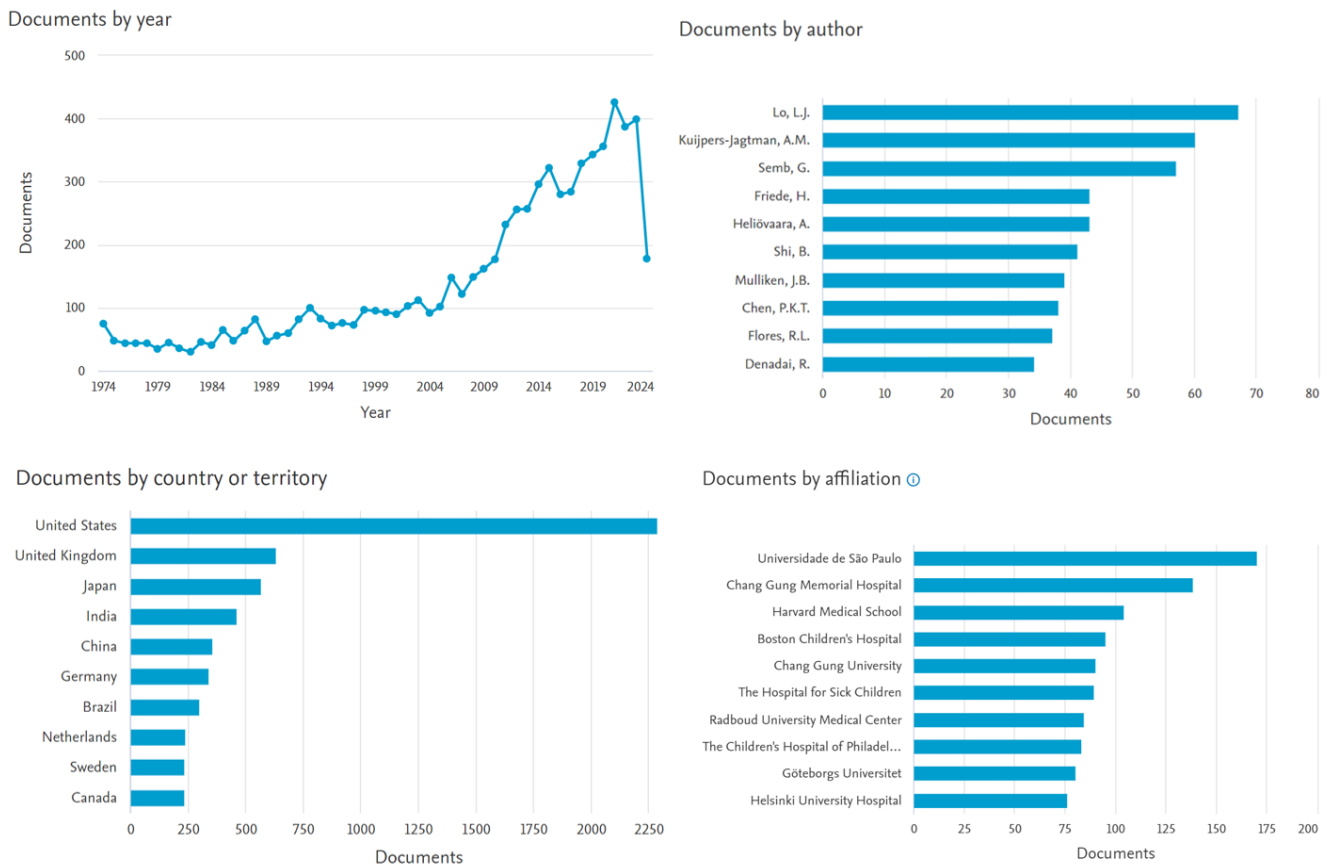
of publications over the past half-century, this study aims to highlight significant trends, influential research, and key breakthroughs that have contributed to the field.

In this paper, we present a detailed bibliometric analysis of the literature on CLP surgery from the last fifty years. Our objective is to identify and discuss the major trends and milestones that have defined the surgical management of CLP. By examining publication patterns, citation metrics, and the emergence of new surgical techniques and technologies, we aim to provide a comprehensive overview of the progress in this field. This analysis not only sheds light on past achievements but also highlights current challenges and future directions for research and clinical practice in CLP surgery.

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## II. MATERIAL AND METHODS

### a. Data Source and Search Strategy



**Figure 1. Number of publications and most productive authors, institutions, and countries. (A) Annual scientific publications throughout the entire publication period, (B) Top ten of most productive authors (C) Top ten of most productive countries, and (D) Top ten of most productive affiliations.**

The data for this bibliometric analysis were sourced from the SCOPUS database, a comprehensive repository of peer-reviewed literature. A search was conducted on May 22, 2024, using keywords related to "cleft lip and palate surgery." The search was restricted to articles published between 1974 and 2024, encompassing a 50-year span. To ensure the inclusion of only relevant and high-quality studies, the search was further refined to include only documents classified as articles, in the final stage of publication, and written in English. This initial search yielded a total of 10,692 documents. After applying these limitations, the dataset was narrowed down to 7,283 documents.

### b. Data Analysis

The bibliometric analysis was conducted using VOSviewer. VOSviewer is a software tool designed for constructing and visualizing bibliometric networks. It was used to create co-authorship, co-occurrence, and citation networks, providing a visual representation of the relationships and collaborations within the field. Through this tool, we systematically analyzed the selected documents to uncover significant trends, milestones, and shifts in the surgical management of cleft lip and palate over the past fifty years.

## III. RESULTS AND DISCUSSION

### a. Annual Publications

The analysis of annual publications in cleft lip and palate surgery reveals significant growth over the past fifty years. Starting with 75 publications in 1974, the number remained relatively steady until the early 1990s. A notable increase began in the 2000s, with a substantial rise in the 2010s. Recent years show a peak with 426 publications in 2021. The trend demonstrates a growing research interest and advancements in the field, culminating in 387 publications in 2022 and 399 in 2023. This upward trajectory highlights the field's expanding knowledge base and the ongoing focus on improving surgical outcomes for cleft lip and palate.

### b. Top Authors, Countries, and Affiliations

The bibliometric analysis identified the leading authors in cleft lip and palate surgery over the past fifty years. Lo, L.J. tops the list with 67 publications, followed by Kuijpers-Jagtman, A.M. with 60, and Semb, G. with 57. Friede, H. and Heliövaara, A. each have 43 publications, while Shi, B. has 41. Mulliken, J.B. contributed 39 articles, Chen, P.K.T. 38, Flores, R.L. 37, and Denadai, R. 34. These prolific authors have greatly influenced the field through their extensive research.

Research in cleft lip and palate surgery has been led by the United States with 2,284 publications, followed by

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significant contributions from the United Kingdom (631), Japan (565), India (460), and China (352). Germany, Brazil, the Netherlands, Sweden, and Canada have also made notable contributions with 335, 296, 235, 233, and 232 publications respectively, collectively advancing surgical techniques, public health approaches, and comprehensive care in this field.

Leading affiliations in cleft lip and palate surgery research include Universidade de São Paulo (170 publications), Chang Gung Memorial Hospital (138), and Harvard Medical School (104). Boston Children's Hospital (95), Chang Gung University (90), and The Hospital for Sick Children (89) also feature prominently. Radboud University Medical Center (84), The Children's Hospital of Philadelphia (83), and Göteborgs Universitet (80) round out the top contributors, showcasing their significant impact on the field.

### c. Top 10 Most Cited Papers

**Table 1. Top 10 Most Cited Papers in Cleft Lip and Palate Surgery**

Ran k	Title	Journal	Year	Citations
1	Aneurysm syndromes caused by mutations in the TGF- $\beta$ receptor	New England Journal of Medicine	2006	1,350
2	Anatomical classification of facial, cranio-facial and latero-facial clefts	Journal of Maxillofacial Surgery	1976	668
3	Obstructive sleep apnea in infants and children	The Journal of Pediatrics	1982	612
4	Treatment variables affecting facial growth in complete unilateral cleft lip and palate. Part 1: Treatment affecting growth	Cleft Palate Journal	1987	461
5	Clinical practice guideline: Tympanostomy tubes in children	Otolaryngology - Head and Neck Surgery (United States)	2013	374
6	A technique for cleft palate repair	Plastic and Reconstructive Surgery	2003	356

7	Nevoid basal cell carcinoma syndrome (Gorlin syndrome)	Orphanet Journal of Rare Diseases	2008	348
8	Neurodevelopmental outcome at 5 years of age of a national cohort of extremely low birth weight infants who were born in 1996-1997	Pediatrics	2005	304
9	Cleft palate fistulas: A multivariate statistical analysis of prevalence, etiology, and surgical management	Plastic and Reconstructive Surgery	1991	296
10	A review of tooth formation in children with cleft lip/palate	American Journal of Orthodontics and Dentofacial Orthopedics	1986	274

The top ten most cited papers in cleft lip and palate surgery highlight key advancements and influential research in the field. Leading the list is a 2006 study from the New England Journal of Medicine on TGF- $\beta$  receptor mutations, crucial for understanding genetic syndromes related to cleft conditions. Foundational work like the 1976 anatomical classification of facial clefts and the 1987 analysis of treatment impacts on facial growth have significantly shaped diagnostic and treatment strategies. Clinical practice guidelines, such as the 2013 recommendations for tympanostomy tubes, and innovative surgical techniques introduced in 2003, have improved patient management and outcomes.

Additionally, these top-cited studies cover a broad range of topics, including the management of complications like obstructive sleep apnea and cleft palate fistulas, the intersection of rare genetic syndromes with cleft conditions, and neurodevelopmental outcomes for affected children. The body of work underscores the importance of a multidisciplinary approach, combining genetic research, surgical innovation, and comprehensive clinical care to advance the treatment and understanding of cleft lip and palate.

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## d. Topic Hotspots

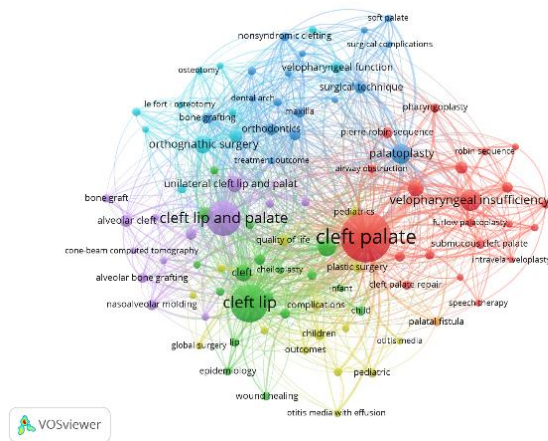


Figure 2. The visualization map of keyword occurrence.

Figure 2 shows a network visualization map depicting keyword co-occurrences related to cleft lip and palate surgery. Each color represents a different cluster of related terms. These clusters are groups of keywords that frequently appear together in the literature, indicating closely related topics or themes within the field of cleft lip and palate surgery. Red Cluster focuses on terms related to "cleft palate" and includes keywords like "velopharyngeal insufficiency," "palatoplasty," and "pharyngoplasty." This suggests a concentration on surgical techniques and complications associated with cleft palate repair. Green Cluster centers around "cleft lip" and includes terms such as "cheiloplasty," "wound healing," and "epidemiology." This cluster seems to focus on the surgical repair of cleft lip and related procedures. Purple Cluster Includes terms like "cleft lip and palate," "unilateral cleft lip and palate," and "alveolar cleft." This cluster appears to cover broader aspects of cleft conditions that involve both lip and palate. Blue Cluster contains keywords like "orthognathic surgery," "orthodontics," and "palatoplasty" indicating a focus on more complex surgical interventions that may be necessary for severe cases or secondary deformities.

The size of each node represents the frequency of that term's occurrence in the literature. Larger nodes indicate terms that are more frequently discussed. Keywrkds such as "cleft palate" "cleft lip" and "cleft lip and palate" are among the largest nodes, indicating they are central and highly researched topics in this field. The lines connecting the nodes represent co-occurrences of terms in the same articles. Thicker lines signify stronger connections or more frequent co-occurrences. For example, the strong connections between "cleft palate," "velopharyngeal insufficiency," and "palatoplasty" suggest these terms are often discussed together, highlighting their clinical and research interdependence.

This network visualization helps identify the main research themes, trends, and relationships in the surgical management of cleft lip and palate over the past 50 years. It shows the evolution and concentration of research efforts,

with some clusters focusing on surgical techniques, outcomes, and specific types of clefts, while others look at broader or ancillary issues such as quality of life and epidemiology.

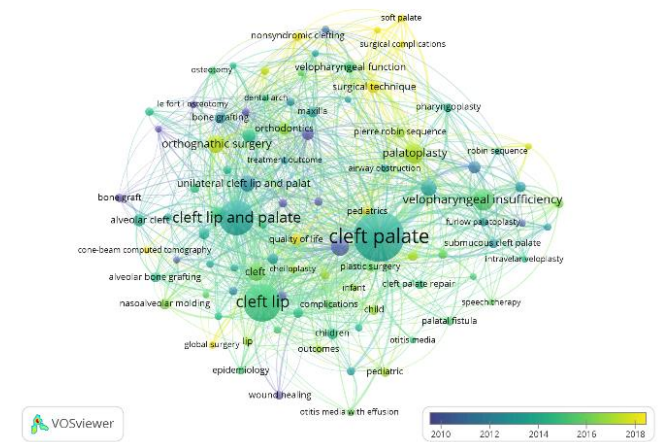


Figure 3. The visualization map of the keyword overlay by year

The colors illustrate different years with a gradient from purple to yellow. This allows us to see how certain topics have gained or lost prominence over time. Purple nodes represent terms that were more commonly discussed around 2010. Yellow nodes show terms that have become more prominent in recent years, around 2018.

Keywords such as "wound healing", "bone graft", and "le fort osteotomy" are in purple nodes, indicating that these areas have been longstanding focal points in cleft surgery research. Advances in these areas reflect a deeper understanding of the biological, material, and technical aspects of cleft repair and reconstruction, driving continuous improvements in patient care. The green nodes manifested by keywords such as "cleft lip and palate", "cleft lip", "cleft palate", "velopharyngeal insufficiency", "velopharyngeal function", "alveolar cleft", and "pharyngoplasty". The green nodes highlight central themes in cleft lip and palate research during the mid-2010s. These terms encompass critical areas of surgical intervention, functional outcomes, and interdisciplinary care necessary for the comprehensive treatment of patients with cleft conditions. The yellow nodes such as "quality of life", "cone-beam computed tomography", and "nonsyndromic clefing", represent topics that have been prominent around 2018 and more recently. This approach helps to pinpoint new areas of interest or focus within a particular field of study, providing valuable insights for researchers and scholars.

## IV. LITERATURE REVIEW

Cleft lip and palate (CLP) are among the most common congenital anomalies (1 in 600-800 live births) affecting the craniofacial region, with significant implications for speech, hearing, aesthetics, and psychosocial development(1,3)(4–6). Over the past fifty years, surgical techniques and



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multidisciplinary approaches to CLP have evolved substantially, driven by advances in medical technology, understanding of embryology and genetics, and a focus on patient-centered outcomes(7,8).

The treatment of cleft lip and palate dates back centuries, with early documentation of surgical attempts in ancient China and Rome(9). However, significant progress in CLP surgery began in the 20th century. In the early 1900s, surgeons like Victor Veau in France and Sir Harold Gillies in the UK made pioneering contributions to surgical techniques. The 1960s and 1970s marked a period of standardization and refinement of these techniques, laying the groundwork for the modern approaches discussed in this review(10).

## *Primary Repair Techniques*

### 1. Cleft Lip Repair

Millard Rotation-Advancement is introduced by Ralph Millard in the 1950s, this technique became the gold standard for unilateral cleft lip repair (11). It focuses on creating a natural-looking lip with minimal scarring. Tennison-Randall Technique is a method which involves a triangular flap, offers another approach to achieving symmetry and functional outcomes(12).

### 2. Cleft Palate Repair

Von Langenbeck Technique developed in the 19th century, remained popular through the 20th century(13). It involves simple bipedicle flaps to close the cleft(14). V-Y Pushback Technique is introduced to lengthen the palate and reduce the incidence of velopharyngeal insufficiency (VPI)(15). Furlow Double Opposing Z-Plasty developed in the 1980s, this technique aimed to improve speech outcomes by reconstructing the velopharyngeal musculature more effectively(16).

## *Secondary Procedures and Refinements*

Secondary surgeries have become integral to CLP management, addressing residual deformities and functional deficits. These procedures include alveolar bone grafting, typically performed around 7-9 years of age, using autologous bone (often from the iliac crest) to support tooth eruption and maxillary arch continuity(17). Rhinoplasty, or secondary nasal surgery, focuses on correcting nasal deformities associated with cleft lip and palate, enhancing both aesthetics and function(18). Additionally, orthognathic surgery, such as the Le Fort I osteotomy, is commonly performed on patients with significant maxillary hypoplasia to correct skeletal discrepancies and improve occlusion(19).

## *Multidisciplinary Approach*

The treatment of cleft lip and palate (CLP) is increasingly multidisciplinary, involving surgeons, orthodontists, speech therapists, audiologists, and psychologists(20). This team approach addresses the complex needs of CLP patients. Early and continuous orthodontic management aligns teeth and prepares for surgeries like alveolar bone grafting and orthognathic surgery(21).

Speech therapy is essential for treating articulation disorders and velopharyngeal insufficiency (VPI), with early and ongoing intervention improving speech and communication(22). Regular audiological evaluations and managing middle ear effusions are crucial for preventing conductive hearing loss(23).

Psychosocial support helps patients and families cope with social and emotional challenges. Counseling and support from psychologists improve self-esteem, social interactions, and overall well-being(24).

## *Technological Innovations*

### 1. 3D Imaging and Printing

Advances in imaging technologies, such as 3D CT and MRI, have revolutionized preoperative planning and intraoperative navigation. 3D printing has enabled the creation of patient-specific surgical models and guides, enhancing precision and outcomes (25).

### 2. Tissue Engineering and Regenerative Medicine

Emerging fields like tissue engineering also hold promise for CLP treatment. Research into the use of stem cells and growth factors for regenerating bone and soft tissue aims to reduce the need for autologous grafts and improve healing(26–28).

### 3. Minimally Invasive Techniques

The development of minimally invasive surgical techniques, including endoscopic approaches, has reduced morbidity and improved recovery times(29,30). These techniques are particularly useful in secondary procedures and for addressing complications such as fistulas(31).

## *Quality of Life and Global Disparities*

Longitudinal studies have shown sustained improvements in Quality of Life (QoL) for individuals undergoing modern CLP surgery. Patients report higher satisfaction with their appearance, better social interactions, and improved vocational achievements(32). Continuous advancements in less invasive techniques and more effective rehabilitative therapies continue to enhance these positive outcomes.

Despite significant advancements, disparities in access to quality CLP care persist globally, particularly in low-resource settings(33). Many regions lack the necessary infrastructure, trained professionals, and financial resources to provide comprehensive CLP treatment(34–36). This results in delayed or inadequate care, leading to poorer outcomes and lower QoL for affected individuals(34).

Various initiatives aim to bridge these gaps by standardizing care protocols and providing training and resources to local healthcare providers. International organizations and non-profits are instrumental in delivering surgical missions, establishing local treatment centers, and advocating for better health policies. These efforts are crucial in improving access to care and outcomes for CLP patients in underserved regions.

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Future research should focus on developing cost-effective, universally applicable surgical and rehabilitative interventions. Telemedicine and digital health platforms could play a significant role in providing remote support and education to healthcare providers in low-resource settings. Additionally, fostering partnerships between high-resource and low-resource regions can facilitate knowledge transfer and capacity building.

### CONCLUSION

Over the past fifty years, the field of cleft lip and palate surgery has witnessed remarkable advancements, driven by technological innovations, a multidisciplinary approach, and a focus on patient-centered care. The future of CLP care lies in continued research, innovation, and a commitment to comprehensive, individualized care for all patients.

Using bibliometric and visualization techniques, our study revealed three key findings: (1) There has been a significant rise in the number of publications related to cleft lip and palate surgery over the past fifty years. (2) Our analysis highlights the crucial importance of orthognathic surgery, palatoplasty, and addressing velopharyngeal insufficiency. It suggests that future research should focus on these areas, promote international collaboration, and increase funding to drive innovation in this field. (3) As this study was based solely on the Scopus database, we recommend that future research should include multiple databases such as PubMed, Web of Science, and Springer to capture a wider range of high-quality scientific contributions.

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