

Scientometric Research Analysis of Cleft Lip and Palate Literature: Hot Spots, Most Influential Countries / Journals, History and Future

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ABSTRACT

Objective: Identifying and mapping the hotspots of the Cleft lip and palate (CLP) literature from 1980 to 2022, in addition it is aimed to present the results in a simplification. This article aims to offer an overview of pertinent research, examine prevailing research patterns, and stimulate appropriate areas for future investigation.

Methods: In this scientometric research analysis, CLP publications between 1980 and 2022 were obtained from the Web of Science Core Collection database. The keywords “cleft lip and palate”, “cleft lip”, “cleft palate” and “oral cleft” were used in the search. CiteSpace, R-Studio (biblioshiny program) and Excel 2016 software were used for visualizations and analysis.

Results: A total of 26,319 articles and 473,074 references were analyzed. The top 3 countries with the most publications were the United States (9,368 publications, 35.59% of the total), the United Kingdom (2,062, 7.83%), and Japan (1,786, 6.78%). After the co-citation clustering analysis, it was seen that the CLP literature was divided under 13 headings. The most influential articles were found with the citation burst analysis.

Conclusion: Scientometric analysis study has been done in CLP’s 42-year publication history. The results obtained present the general trend of the CLP literature, the topics on which the literature is divided, and the hot spots. With similar scientometric analysis studies to be carried out in the future, the progress of the literature can be followed and the future can be predicted.

Keywords: Cleft lip, Cleft palate, Scientometric.

INTRODUCTION

Cleft lip and palate (CLP), which is the most common human craniomaxillofacial birth defects, is a complex disease caused by the interaction of environmental and genetic factors [1]. For the treatment of CLP, which has many effects on the affected individual in terms of nutrition, speech, breathing, psychological and social aspects, many treatment types such as surgical intervention, as well as orthodontic treatments, speech therapies, psychological treatments are applied as multi-disciplinary [2].

Due to many reasons such as developing surgical technologies, decreasing costs, increasing experience in this field, and facilitating access to treatment, the number of access and treatment options for CLP patients is increasing day by day. As the number of treatments in this field increases, the number of scientific publications in this field, which develops in accordance with the nature of the development of the scientific process, also increases. Scientific fields have employed diverse statistical analysis methods, including scientometric analysis, to assess the efficacy of journals and articles [3–7]. Furthermore, these

analyses enable the anticipation of the evolutionary trajectory and potential advancements within specific subject domains [8, 9].

There were previous bibliometric analysis studies on CLP [1, 10] journal, authors, first authors, keywords, and citations were extracted and quantitatively analyzed using Bibliographic Item Co-Occurrence Matrix Builder software. The word matrix and co-occurrence matrix were established, and the co-citation analysis, keyword clustering, and social network analysis (SNA). Nevertheless, previous studies lacked a comprehensive assessment through reference and mapping analysis to gauge the extent of contributions made by the analyzed publications to the existing literature. Science mapping and visualization, on the other hand, facilitate the exploration of scientific knowledge [11–14] and give varying results. In this study we compare the accuracies of cluster solutions of a large corpus of 2,153,769 recent articles from the biomedical literature (2004–2008). Specifically, document co-citation analysis enables the identification of pertinent literature, academic communities, and societal influences that might be overlooked in conventional literature review approaches [11].

In this study, identifying and mapping the hotspots of the CLP literature from 1980 to 2022, in addition it is aimed to present the results in a simplification. This article aims to offer an overview of pertinent research, examine prevailing research patterns, and stimulate appropriate areas for future investigation.

MATERIALS AND METHODS

Search Strategies

The search was performed on 04.09.2022 on the Web of Science Core Collection (WoSCC) database to avoid bias due to daily database updates. Search terms (TOPIC = cleft lip and palate) or (TOPIC = cleft lip) or (TOPIC = cleft palate) or (TOPIC = oral cleft) search terms and time 1980-2022 search settings are used,

Main Points;

- The United States was the leading country with the highest number of publications and citations.
- CLP literature was divided into 13 clusters.
- ‘Cleft palate’, ‘palate’, ‘cleft lip and palate’, ‘cleft lip’, ‘lip’, ‘children’, ‘orofacial cleft’ were the most used keywords.

including all document types. Only articles written in English were included in the analysis.

Programs and Methods Used in Data Analysis

Data were retrieved from the WoSCC database in “Tab Delimited File” and “Plain Text” formats. The data were processed in Excel 2016 software in order to standardize the writing style of author names, countries, institutes and to make a file ready for analysis in scientometric analysis programs. CiteSpace, R-Studio and Excel 2016 software were used for visualizations and analysis.

Maps are made up of nodes and links. Nodes represented the analyzed item, such as article, author, country, and keyword, depending on the analysis type. Node size in the visualization represents the frequency of citations, whereas the color spectrum ranges from purple to yellow (with increasing yellowness closer to the present), and the outermost pink ring indicates the centrality value of each node. The connecting lines between nodes depict co-occurrence or co-citation relationships, with line thickness denoting the strength of the relationship.

Parameters used in the analysis with Citespace software: time slicing (1980–2022), year per slice (5), term source (entire selection), node type (common-citation/country/keyword/journal), selection criteria (top 50), pruning (Pathfinder) and visualization (Cluster View - Static, Show Merged Network).

Citespace Analysis

Co-citation refers to the frequency at which two documents are cited together by other documents [3, 4]. If two common documents are mentioned in at least one other document, it is said that these documents are co-cited [3, 8].

Cluster analysis is a statistical method used to classify data based on the level of similarity, with the objective of uncovering the distribution of research content pertaining to specific topics [3,4]. In this analysis, in order to determine the cluster titles, the frequency of the words in the titles and abstracts of the articles were taken into consideration and the most frequently used words were assigned as the cluster title. Modularity (Q-score) and Silhouette (S-score) are measures used to assess cluster mapping. A Q-score greater than 0.3 indicates significant constrained associations within the clusters, while an S-score above 0.5 suggests that the cluster is reasonable. An S-score exceeding 0.7 indicates that the cluster is both efficient and persuasive [3, 8].

A burst refers to the frequency fluctuation of a particular type of event. By identifying the references with bursts of citations, important articles in the literature can be identified and an idea of the future path of the literature can be obtained [3, 8, 15].

By analyzing the keywords used in the articles, it was aimed to examine the connection between the publications that make up the literature and to determine the hot spots by performing an burst analysis [3,4].

RESULTS

General Data and Cross-Country Cooperation

A total of 26,319 articles and 473,074 references were included in the analysis. The annual growth rate of the number of publications was 4.32%. The United States (9,368 publications, 35,59% of the total) ranked first in the number of publications, followed by the United Kingdom (2,062, 7.83%), Japan (1,786, 6.78%) and People’s Republic of China (1,734, 6.58%). The cooperation map of the countries contributing to the CLP literature is shown in Figure 1 (The purple circles around the nodes and the values written in purple text are the centrality value of that country). A high level of cooperation has been demonstrated with 566 cooperation links between 81 countries.

Journal Analysis

In the journal analysis, the biblioshiny program was used over the R Studio software. In order to determine the most effective journals, the total number of publications, the number of citations and the zone in which they are according to Bradford’s

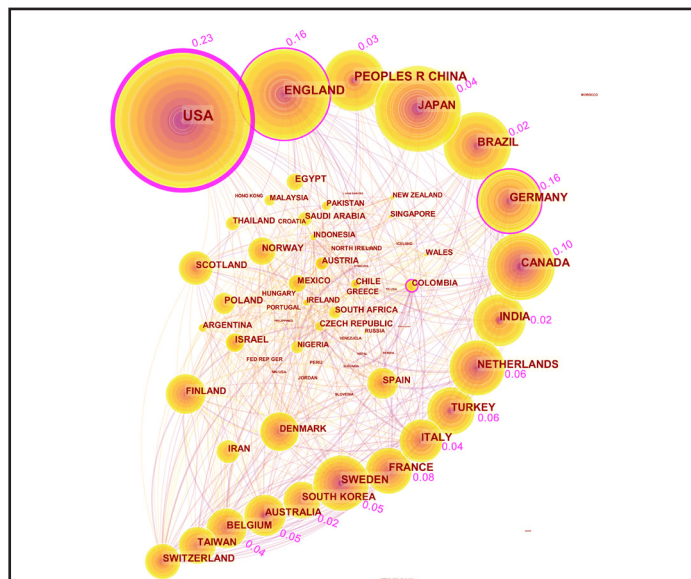


Figure 1. Cooperation network of countries. The outermost pink ring and text represent the centrality value.

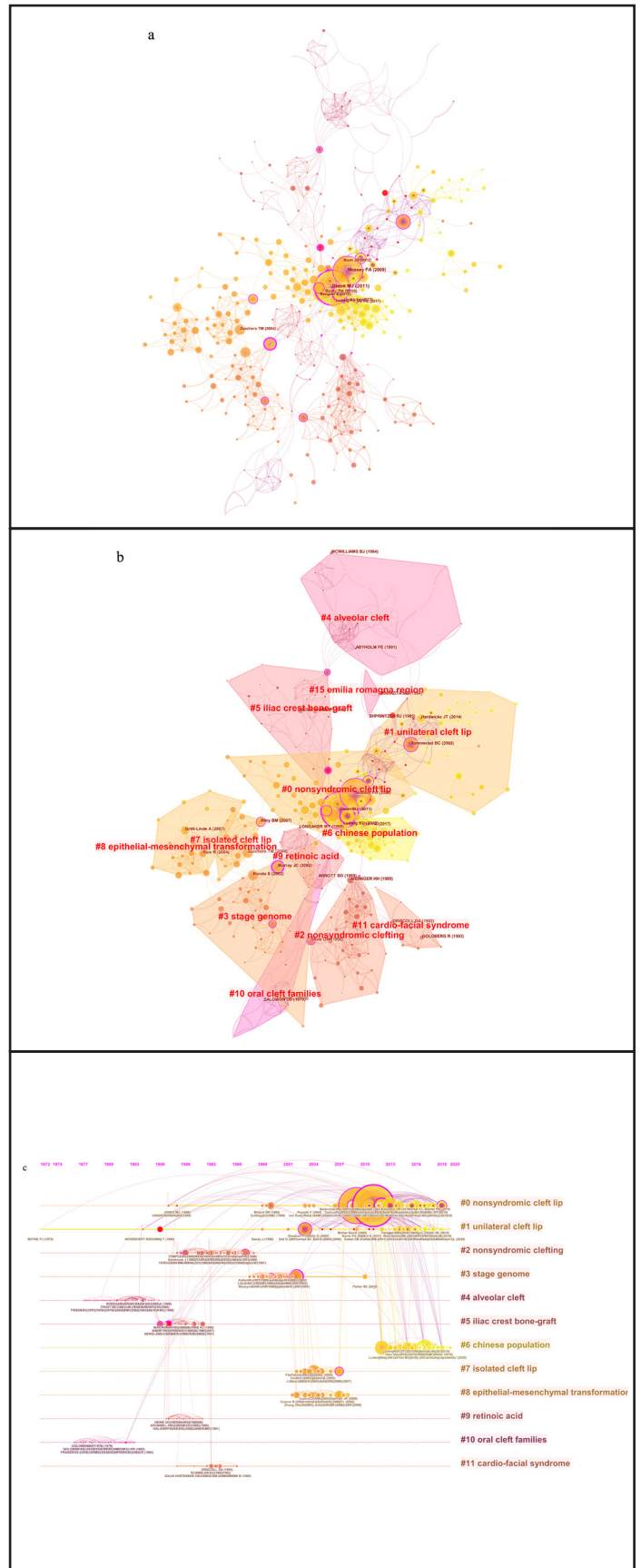


Figure 2. Co-citation analysis. **a.** View before clustering analysis. **b.** View after cluster analysis. **c.** Timeline view

Law of Scattering were determined. According to the results of the journal analysis, Cleft Palate-Craniofacial Journal, Journal of Craniofacial Surgery, and Plastic and Reconstructive Surgery were the top 3 journals with the highest number of publications, while the 20 most influential journals in the field of CLP were summarized in Table 1.

Co-Citation and Cluster Analysis

In the co-citation analysis, the period between 1980 and 2022 was divided into 5-year periods and the 50 most cited articles of the 5-year period were found and analyzed. As a result of the analysis, 439 nodes (reference) and 1,563 connections were revealed (Figure 2a).

The Q-score of the CLP literature was 0.7992 and the S-score was 0.9516. It was seen that the CLP literature was divided into a total of 13 clusters. Information including the metric

properties of the clusters in which the CLP literature is divided and the characteristics of the article, which is the main common-reference source, are summarized in Table 2. Figure 2b shows the boundaries of the clusters made on the co-citation analysis map. 3 clusters (Cluster #4-11-15) were separated by clustering in isolation from the others.

The time-line view was used to understand in which years the clusters were more active (Figure 2c). The accumulation of nodes in the Nonsyndromic Cleft Lip (Cluster #0), Unilateral Cleft Lip (Cluster #1) and Chinese Population (Cluster #6) clusters in recent years indicates that these clusters are younger. CLP terminology has varied over time. When Figure 2c is examined, while the term 'Nonsyndromic clefting' was active until the 1990s, it has evolved into the term 'Nonsyndromic cleft lip' since the late 1990s.

Table 1. Top 20 journals in the field of cleft lip and palate

Rank	Title	Number of Articles	Total Citation	Bradford's Law of Scattering	Impact Factor	Centrality	Quartile
1	Cleft Palate-Craniofacial Journal	3928	61887	Zone 1	1.915	0.77	Q2
2	Journal of Craniofacial Surgery	1390	11669	Zone 1	0.924	0.07	Q2
3	Plastic and Reconstructive Surgery	1245	27445	Zone 1	2.299	0.49	Q1
4	Cleft Palate Journal	641	12919	Zone 1	1.477	0.35	Q3
5	American Journal of Medical Genetics Part A	594	10523	Zone 1	2.249	0.13	Q2
6	Journal of Cranio-Maxillofacial Surgery	439	7316	Zone 1	3.289	0.09	Q1
7	American Journal of Medical Genetics	395	13334	Zone 1	2.090 (2004)	0.24	Q2
8	Journal of Oral and Maxillofacial Surgery	375	6413	Zone 1	1.699	0.09	Q1
9	International Journal of Pediatric Otorhinolaryngology	369	4730	Zone 2	1.641	0.10	Q2
10	Journal of Dental Research	365	3403	Zone 2	7.347	0.05	Q1
11	Annals of Plastic Surgery	343	3604	Zone 2	1.425	0.00	Q2
12	International Journal of Oral and Maxillofacial Surgery	274	4715	Zone 2	2.933	0.00	Q1
13	American Journal of Human Genetics	234	9282	Zone 2	9.318	0.14	Q1
14	British Journal of Oral & Maxillofacial Surgery	221	3005	Zone 2	1.597	0.00	Q2
15	Journal of Plastic Reconstructive and Aesthetic Surgery	220	2440	Zone 2	1.550	0.04	Q1
16	Teratology	202	5428	Zone 2	1.689 (2002)	0.32	-
17	American Journal of Orthodontics and Dentofacial Orthopedics	191	4019	Zone 2	1.546	0.13	Q1
18	Birth Defects Research Part A-Clinical and Molecular Teratology	187	6863	Zone 2	2.154 (2018)	0.58	Q2
19	Scandinavian Journal of Plastic and Reconstructive Surgery and Hand Surgery	175	3390	Zone 2	-	0.04	Q4
20	British Journal of Plastic Surgery	163	3199	Zone 2	-	0.00	-

Table 2. After the co-citation analysis, the topics and summary information on which the cleft lip and palate literature is divided are shown. (Cluster #15 was not included because it was too small and too specific)

Cluster	Size	Silhouette	Mean (Year)	Main References	Title	Main Conclusion	Quartiles
#0 nonsyndromic cleft lip	66	0.883	2009	Mossey PA, 2012, Front Oral Biol [23]	Epidemiology of oral clefts 2012: an international perspective	In this article, the epidemiological features of CLP have been evaluated.	Q1
#1 unilateral cleft lip	45	0.945	2010	Parvaz MA, 2009, J Plast Reconstr Aesthet Surg [24]	Width of cleft palate and postoperative palatal fistula-do they correlate?	Cleft palate width has been found to have an effect on postoperative palatal fistula formation, with a width of 15 mm or more having a statistically significant risk of fistula formation.	Q1
#2 nonsyndromic clefting	39	0.98	1993	Ardinger HH, 1989, Am J Hum Genet [25]	Association of genetic variation of the transforming growth factor-alpha gene with cleft lip and palate	A significant association was observed between two RFLPs at the transforming-growth-factor-alpha (TGFA) locus and the occurrence of clefting.	Q1
#3 stage genome	37	0.956	1999	Murray JC, 2002, Clin Genet [26]	Gene/environment causes of cleft lip and/or palate	An overview of the gene-environment contributions to nonsyndromic forms of clefting and their implications for developmental biology and clinical counseling is presented.	Q1
#4 alveolar cleft	37	0.965	1983	Abyholm FE, 1981, Scand J Plast Reconstr Surg [27]	Secondary bone grafting of alveolar clefts. A surgical/orthodontic treatment enabling a non-prosthetic rehabilitation in cleft lip and palate patients	The basic principles used in primary cleft palate grafting are explained. Stabilizing the mobile primary palate, closure of oronasal fistulas, and grafting before eruption of impacted canines on the cleft side have been suggested.	Q4
#5 iliac crest bone-graft	30	0.968	1988	Semb G, 1991, Cleft Palate Craniofac J [28]	A Study of Facial Growth in Patients with Bilateral Cleft Lip and Palate Treated by the Oslo CLP Team	Various treatment modalities used in CLP patients are described.	Q2
#6 chinese population	30	0.964	2016	Yu Y, 2017, Nat Commun [29]	Genome-wide analyses of non-syndromic cleft lip with palate identify 14 novel loci and genetic heterogeneity	For non-syndromic cleft palate, 41 SNPs were identified in 26 loci, 14 of which were novel (RAD54B, TMEM19, KRT18, WNT9B, GSC/DICER1, PTCHI, RPS26, OFCC1/TFAP2A, TAF1B, FGF10, MSX1, LINC00640, FGFR1 and SPRY1), providing genome-wide significance.	Q1
#7 isolated cleft lip	20	0.972	2004	Zuccherro, 2004, N Engl J Med [30]	Interferon regulatory factor 6 (IRF6) gene variants and the risk of isolated cleft lip or palate	DNA-sequence variants associated with interferon regulatory factor 6 (IRF6) are major contributors to cleft lip, with or without cleft palate. The contribution of variants in single genes to cleft lip or palate is an important consideration in genetic counseling.	Q1

#8 epithelial-mesenchymal transformation	20	0.967	2005	Gritti-Linde, 2006, Dev Biol [31]	Molecular control of secondary palate development	This review highlights the current understanding of the molecular and cellular mechanisms involved in normal and abnormal palate development with special respect to recent advances derived from studies of mouse models.	Q1
#9 retinoic acid	16	0.967	1988	Longaker, 1990, J Pediatr Surg [32]	Studies in fetal wound healing VI. Second and early third trimester fetal wounds demonstrate rapid collagen deposition without scar formation	It has been reported that collagen accumulates much more rapidly in fetal wounds than in adult wounds. Wound collagen deposition occurred in a normal dermal and mesenchymal pattern in second and early third trimester fetal lambs. These findings are consistent with the observation that the fetus heals rapidly and without scarring. In contrast, wounds in late pregnancy fetal lambs showed some evidence of scarring.	Q1
#10 oral cleft families	16	0.992	1979	Tyan, 1978, Proc Soc Exp Biol Med [33]	Genetic and environmental factors in cortisone induced cleft palate	susceptibility to cleft palate in the mouse appears to be under multigenic control. The evidence suggests that perhaps two factors determining the degree of susceptibility to cortisone induced cleft palate are associated with the major histocompatibility complex (H-2) and certain dietary factors can modulate this susceptibility via a gene(s) which tentatively maps in the region of H-2D.	Q1
#11 cardio-facial syndrome	9	0.998	1992	Goldberg, 1993, Am J Med Genet [34]	Velo-cardio-facial syndrome: a review of 120 patients	The purpose of this report is to update the findings associated with the velo-cardio-facial syndrome, including: deletions of DNA probes specific to the 22q11 chromosome region, hypocalcemia, psychiatric findings, vascular and cardiovascular anomalies, constitutional growth delay, and hypothyroidism.	Q2



Figure 3. Citation burst analysis. **a.** Top 25 references with the strongest citation bursts. **b.** Top 25 keywords with the strongest citation bursts.

Citation Burst Analysis

In Figure 3a, the 25 articles with the strongest citation explosion in the CLP literature were found and visualized with the Citespace software. In the visualization, the starting point of a blue line represents the publication date of an article. On the other hand, the beginning of a red segment signifies the commencement of a burst period, while the end of the red segment indicates the

conclusion of the burst period. Of the 25 references, 22 were on genetics, while 3 were on the epidemiology and etiology of CLP. The burst power of the article titled “*Cleft lip and palate: understanding genetic and environmental influences*” published by Dixon et al.[16] in 2011 was 1.5 times that of its closest competitor.

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Keyword Analysis

The network of keywords co-occurrence is shown in Figure 4. ‘Cleft palate’, ‘palate’, ‘cleft lip and palate’, ‘cleft lip’, ‘lip’, ‘children’, ‘orofacial cleft’ were the most used keywords. The citation burst analysis of the keywords is shown in Figure 3b. In this image, the idea of which sub-headings about CLP attracts the attention of the authors in certain time periods from 1980 to the present is reached.

DISCUSSION

This scientometric research analysis study provides a key insight into the field, identifying trends and providing a visual analysis of publications in the CLP literature. As a result of the analysis of the data obtained from the WoS database, it was seen that the number of publications on CLP increased significantly on an annual basis. The increasing number of publications and the increase in the number of journals in which articles are published increase the area where the literature is distributed. Bradford’s Scattering Law can help researchers know about a particular literature and choose the journals to publish their research. Bradford’s Scattering Law “describes the scatter or distribution of literature on a particular subject” [3,4]. If you wish to compile a bibliography on a particular topic, a small core group will find that the journal always contains a significant (one-third) of the articles published in that topic or discipline. It is then seen that a group of second or more journals contains the other third of all articles, and a much larger group of journals the last third [3,4].

When the contributions of the countries to the literature were examined, the United States, the United Kingdom, Japan, People’s Republic of China and Germany were in the top 5 places. If we open a special title for the People’s Republic of

China here, although the People’s Republic of China is in the top 5 in the total number of publications, the fact that the centrality value is relatively low compared to other countries can be interpreted as low cooperation with other countries. However, when the subject headings in which the literature is divided are examined (Figure 2b-c), it is seen that the ‘Chinese Population’ (Cluster #6) cluster is such a large cluster that it can be in a separate place in the literature. Carefully following the study of the People’s Republic of China in this area and increasing international cooperation with this country will make a great contribution to the literature.

Although most articles emphasize the importance of speech therapy [17, 18] and psychological support [19, 20] among the treatment steps of CLP, there is a lack of studies on these issues in the literature. Neither the clustering analysis nor the citation burst analysis found any findings related to these two issues. There are differences in the literature regarding the benefits of early intervention regarding the timing of speech therapy. The systematic reviews of Bessell et al. [21] and Meinusch and Romonath [22] found little evidence for the benefits of early talk therapy. However, in the systematic review published by Lane et al., it is suggested that early speech therapy before the age of 3 may be beneficial. Conducting more multidisciplinary studies will help us to see the effects of these two basic disciplines on CLP more clearly.

By examining the keywords, which are accepted as the spirit of the articles, many summary information about the publications can be obtained [1, 3, 8]. When Figure 3 is examined carefully, it is seen that the keywords include 2 different disciplines as surgery and genetics and that the keywords connecting these two disciplines are ‘Cleft palate’, ‘palate’, ‘cleft lip and palate’, ‘cleft lip’, ‘lip’, ‘children’, It can be said that there are keywords such as ‘orofacial cleft’. The lack of direct linking paths from surgical keywords to genetic keywords can be interpreted as studies conducted in these two disciplines are independent of each other. The citation burst analysis of the keywords allowed us to identify the time range of topics of interest to the authors (Figure 2b). For example, while publications on ‘palatoplasty’ were very active between 1980 and 1990, ‘msx1’ was more active between 2001-2010 and ‘irf6’ was more active between 2008 and 2020, and more studies were conducted on these issues in these years. Today, Etiology and Single Nucleotide Polymorphisms are among the most interesting topics. According to the results of the analysis made in this study, it is estimated that these two

issues will increase their importance in the coming years and will find more place in the literature.

Limitations

The study had certain limitations. Firstly, the data analysis conducted was incomplete as it only included English studies. To broaden the scope of the study's data, analysis could have been extended to other databases such as Scopus or Embase. However, this approach was not chosen due to integration issues between the data from the Web of Science (WoS) database and other databases. Consequently, information from other databases was not considered. Nonetheless, given the regular updates to the Web of Science database, there were an ample number of publications available to complete the study.

CONCLUSION

As a result, the general trend in this field, the topics on which the literature is divided, the most influential countries and journals were determined by scientometric analysis of the international publications that make up the CLP literature. The most influential countries in terms of centrality value were the United States, the United Kingdom and Germany, while the People's Republic of China had a significant number of publications in this field, despite its low centrality value. Significant progress can be made in the field of CLP by increasing international cooperation with the People's Republic of China. While the CLP literature is examined under 13 titles in general, the subtitles of Etiology and Single Nucleotide Polymorphisms appear as hot spots that continue to have an impact today, and it is estimated that this situation will continue in the coming years. With similar scientometric analysis studies to be carried out in the future, the progress of the literature can be followed and the future can be predicted.

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