RESEARCH

Current Trends in Cleft Lip and Palate Publications During the Last 10 Years: A Bibliometric Analysis

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ABSTRACT

Current Trends in Cleft Lip and Palate Publications During the Last 10 Years: A Bibliometric Analysis

Background: The purpose of this study is to investigate the apparent trends in cleft lip and palate (CLP) studies published over the last 10 years, and assesses the effectiveness of the studies.

Methods: The SCImago Journal Rank (SJR) data were utilized to select the journals with a high SJR indicator in each of the orthodontics, pedodontics, general dentistry, speech therapy, clinical genetics, pediatrics, plastic esthetic and reconstructive surgery and oral surgery areas. CLP-related studies were identified in the databases accessed through Web of Science owned by Clarivate Analytics. The articles were assessed in terms of year of publication, journal title, specialty, article subject matter, affiliations of the authors, citation relationships and countries/regions of origin. A total of 2,696 CLP-related articles published over the last 10 years were identified based on our search criteria.

Results: The analysis revealed that the most prominent keywords were "palatoplasty", "alveolar bone grafting", "distraction osteogenesis" and "orthognathic surgery" among the treatment procedures. The most common WoS categories among the articles were dentistry, oral surgery & medicine, and surgery. The most cited publications over the last decade have included such terms as "environmental risk factor", "GRHL3", "FGFR2", "loci", "candidate gene" and "BMP".

Conclusion: Recent CLP articles in the literature have focused mainly on treatment procedures, with the most-cited articles generally containing evaluations of the relationship between CLP and genetics. More recent methods have been discussed in only a limited number of studies.

KEYWORDS

Cleft lip. Cleft palate. Trends. Bibliometrics.

ÖZ

Son 10 Yılda Yarık Dudak ve Damak Yayınlarında Güncel Eğilimler: Bibliyometrik Analiz

Amaç: Bu çalışmanın amacı, son 10 yılda yayınlanan yarık dudak damak (YDD) araştırmalarındaki belirgin eğilimleri araştırmak ve çalışmaların etkinliğini değerlendirmektir.

Gereç ve Yöntemler: SCImago Journal Rank (SJR) verileri, ortodonti, pedodonti, genel diş hekimliği, konuşma terapisi, klinik genetik, pediatri, plastik estetik ve rekonstrüktif cerrahi ve ağız cerrahisi alanlarının her birinde yüksek SJR göstergesi olan dergileri seçmek için kullanılmıştır. Clarivate Analytics'e ait Web of Science üzerinden erişilen veri tabanlarında YDD ile ilgili çalışmalar tespit edildi. Makaleler, yayın yılı, dergi başlığı, uzmanlık alanı, makale konusu, yazarların bağlantıları, atıf ilişkileri ve menşe ülke/bölge açısından değerlendirildi. Son 10 yılda YDD ile ilgili yayınlanmış toplam 2.696 makale, arama kriterlerimize göre belirlendi.

Bulgular: Analiz, tedavi prosedürleri arasında en öne çıkan anahtar kelimelerin "palatoplasti", "alveolar kemik grefti", "distraksiyon osteogenezi" ve "ortognatik cerrahi" olduğunu ortaya koydu. Makaleler arasında en yaygın WoS kategorileri diş hekimliği, ağız cerrahisi & tıp ve cerrahi idi. Son on yılda en çok atıf alan yayınlar arasında "çevresel risk faktörü", "GRHL3", "FGFR2", "loci", "aday gen" ve "BMP" gibi terimler yer almıştır.

Sonuç: Literatürdeki son YDD makaleleri esas olarak tedavi prosedürlerine odaklanmıştır ve en çok alıntı yapılan makaleler genellikle YDD ve genetik arasındaki ilişkinin değerlendirmelerini içerir. Daha yeni yöntemler sadece sınırlı sayıda çalışmada tartışılmıştır.

ANAHTAR KELİMELER

Yarık Dudak, Yarık Damak, Trendler, Bibliometri

INTRODUCTION

The treatment of patients with cleft lip and palate (CLP) is based on a long-term multidisciplinary collaboration involving orthodontists, pediatricians, surgeons, language and speech therapists, genetics specialists and dentists. CLP is an area of interest of various specialties, can be examined in a multifaceted manner through inclusive research methods, such as systematic reviews and meta-analyses [1]. A bibliometric analysis is used as an auxiliary approach in such research studies, in that it allows the effectiveness of studies to be analyzed via publications or journals in the available literature, identifying relationships between authors, frequency of words and citations, removing the need to read the content of the publications [2,3]. In addition, the results of the analysis can be visualized using auxiliary software to gain a better understanding. During a bibliometric analysis, it is necessary to limit the sources used so as to represent the subject matter in a consistent and harmonic way [4]. Indicators (such as, SJR indicator, H index and Eigen Factor) that are used to identify the importance of journals can be used also to identify the journals making the greatest contributions to literature [5]. Several researchers have carried out bibliometric analyses of the most cited articles on CLP in literature. Christou et al.[6] examined the most cited articles published in dentistry, medicine

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and oral surgery journals, and concluded that their research could serve as a guide to determine the direction of future research on CLP. Mahon et al. [7] conducted the citation analysis of the most cited publications in 24 journals, and showed the historical development in this field. In the bibliometric analysis research conducted by Zhang et al., [8] in addition to the publication characteristics, research interests were determined by using keyword analysis.

One of main goals in bibliometric studies is to establish the current trends. The treatment approaches to CLP have changed in line with technological developments, as in other fields. For instance, it is now easier to diagnose, plan treatment and evaluate with the developments in imaging and software, leading to better results [9]. In our research, we aim to define the changing trends in this field more comprehensively, not only with the keywords in the most cited publications, but also with subject-specific terms, by using the software tool that provides an up-to-date visualization technique. In order to better determine the features of the publications, we conduct more detailed citation analysis of authors and journals from different disciplines that contribute to this field. In the light of the above information, the present study assesses the current trends in CLP literature spanning a 10-year period, as well as the effectiveness of studies considering the perspectives of various specialties.

METHODS

The study was granted ethics committee approval (blinded) by the ethics committee with decision number 2020-127. The SCImago Journal Rank (SJR) data were utilized to identify the journals to be used in the study, and five journals with a high SJR indicator were selected in each of the orthodontics, pedodontics, general dentistry, speech or communication disorders, clinical genetics, pediatrics, plastic esthetic and reconstructive surgery and oral surgery areas, based on journal titles and SJR subject categories to have CLP-related journals. Subject categories used in journal selection were orthodontics, oral surgery, dentistry (miscellaneous), speech and hearing, pediatrics perinatology and child health, genetics (clinical) and surgery. All of the 40 journals (Table 1) selected were registered on the databases accessed through Web of Science (WoS) owned by Clarivate Analytics, and so the articles were retrieved from the journals using All Databases and the Web of Science Core Collection Database through WoS.

The search string TOPIC= ("cleft lip" OR "cleft palate" OR "oral cleft$") was used to identify CLP-related articles in the selected journals, allowing for the identification of articles in which the search terms were included in the title, abstract, author keywords and keywords plus. The study included articles published between January 2010 and March 2020. No other limiting filters were used. Finally, the selected articles were examined by the researchers to avoid duplicates.

Table 1.

40 journals included in the study for searching CLP-related publications in the fields of orthodontics, pedodontics, general dentistry, speech or communication disorders, clinical genetics, pediatrics, plastic aesthetics and reconstructive surgery, and oral surgery.

	JOURNALS		
1	Angle Orthodontist	21	JAMA Pediatrics
2	American Journal of Orthodontics and Dentofacial Orthopedics	22	Journal of Child Psychology and Psychiatry
3	European Journal of Orthodontics	23	Developmental Review
4	Korean Journal of Orthodontics	24	Child Development
5	Orthodontics and Craniofacial Research	25	Pediatrics
6	Journal of Dental Research	26	IEEE/ACM Transactions on Audio Speech and Language Processing
7	Dental Materials	27	Language and Speech
8	Journal of Dentistry	28	International Journal of Language & Communication Disorders
9	International Journal of Oral Science	29	American Journal of Speech Language Pathology
10	Community Dentistry and Oral Epidemiology	30	Journal of Communication Disorders
11	Nature Reviews Genetics	31	Pediatric Dentistry
12	Genome Research	32	International Journal of Paediatric Dentistry
13	American Journal of Human Genetics	33	European Journal of Paediatric Dentistry
14	Genetics in Medicine	34	Journal of Clinical Pediatric Dentistry
15	Genome Medicine	35	European Archives Of Paediatric Dentistry
16	Plastic and Reconstructive Surgery	36	Journal Of Cranio Maxillofacial Surgery
17	Aesthetic Surgery Journal	37	International Journal of Oral and Maksillofacial Surgery
18	Aesthetic Plastic Surgery	38	Journal of Oral and Maxillofacial Surgery
19	Journal of Plastic Reconstructive and Aesthetic Surgery	39	Cleft Palate Craniofacial Journal
20	Clinics in Plastic Surgery	40	Oral Surgery Oral Medicine Oral Pathology Oral Radiology

Data Extraction

A total of 2,696 articles in 37 of the 40 journals were recorded, including all details and cited references. There are no publications containing the search keywords in three journals. therefore, these three journals were not included in the analysis. The articles were assessed in terms of year of publication, journal title, WoS categories, affiliations of authors and countries/regions of origin. For an advanced bibliometric analysis, the data were uploaded into VOSviewer v.1.6.10 (Leiden, Netherlands) software.

The terms included in the abstract of the articles were analyzed and the results were visualized using density differences. For this purpose, a threshold of term occurrence was set at a minimum of ten. To create a more understandable image, similar words and phrases that did not provide a better information about the content of the subject were excluded. A map of the 449 identified terms was created using the softwares default parameters and full counting method. In the analysis of the 3,819 author keywords used in the articles, the threshold of keyword occurrence was set to a minimum of five. A map of the 255 keywords that had a frequency above the threshold and that were connected to each other (co-occurrence link) was created. The maps for the terms and the keywords

were created using density visualization. In such maps, the larger the number and the greater the frequency of terms/keywords, the closer the color is to yellow, while the smaller the number, the closer the color is to blue. The larger the font, the greater is the significance of the terms/keywords.

The authors of the articles were also identified, and a map was created to show the citation links. The threshold set for this purpose was a minimum of nine publications by an author. An overlay visualization was used to create the map of the citations of the 125 authors with a frequency above the threshold value. Every node on the map indicates an author, and the node size becomes either larger or smaller in proportion to the number of citations received by the author. Authorship links indicate that the authors have cited each other, but do not specify the direction. The thicker the link, the stronger the citation relationship between the authors. Since different scientific fields may have different citation practices, normalization was applied by the default parameters of the software in the evaluation of all citations ([10]).

Possible correlations between the total number of articles, the total number of citations and the mean number of citations per article were tested. A Spearman's correlation test was performed using the IBM SPSS Statistics for Windows, version 22.0 (IBM Corp., Armonk, N.Y., USA) software package. Test results with a P<0.01 were considered statistically significant.

RESULTS

Regarding the different categories, 58.98% of the total articles were related to oral surgery, 20.91% were related to plastic esthetic and reconstructive surgery and 10.01% were related to orthodontics. For the time period of the search, the articles were most frequently published in 2018 (n=314) and 2019 (n=312). The authors were affiliated with 2,216 organizations in 88 countries/regions. The countries/regions that contributed most were the United States, England and China (Figure 1, A). The 10 most productive organizations are presented in Figure 1, B. The most common WoS categories of the articles were dentistry, oral surgery & medicine and surgery. The 10 journals with the highest number of articles are presented in Table 2.
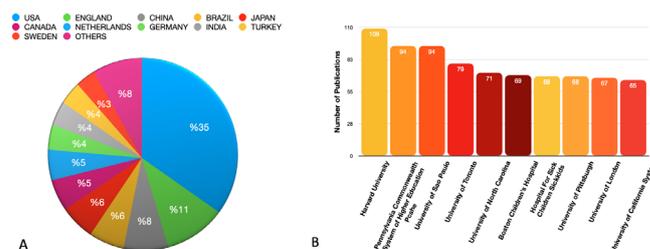

Figure 1

(A) Countries/regions contributing at least 3% of the total number of articles. (B) Graph of 10 most productive organizations.

Table 2.

The 10 journals with the highest number of articles. A Spearman's correlation test revealed a statistically significant, strong positive correlation between the total number of articles and the total number of citations in all journals (r=0.813, P=0.00); no significant association was identified between the total number of articles and the mean number of citations per article.

Journals	Total Publication Count (% of 2696)	Total Citation Count	Citation Per Manuscript
Cleft Palate Craniofacial Journal	1074 (39,84 %)	6708	6,25
Plastic and Reconstructive Surgery	369 (13,69 %)	2941	7,97
Journal of Cranio Maxillofacial Surgery	218 (8,09 %)	1683	7,72
Journal of Plastic Reconstructive and Aesthetic Surgery	152 (5,64 %)	1133	7,45
Journal of Oral and Maxillofacial Surgery	138 (5,12 %)	909	6,59
International Journal of Oral and Maxillofacial Surgery	120 (4,45 %)	1006	8,38
Journal of Dental Research	97 (3,60 %)	1268	13,07
American Journal of Orthodontics and Dentofacial Orthopedics	93 (3,45 %)	836	8,99
European Journal of Orthodontics	65 (2,41 %)	827	12,72
Orthodontics & Craniofacial Research	51 (1,89 %)	337	6,61

Figure 2 shows a visualized map of 449 of the 1,642 terms that scored above the threshold among the 40,245 terms in the abstracts of the articles. An analysis of these 1,642 terms revealed that the articles with a high number of citations discussed mostly the etiology and genetics of CLP: "environmental risk factor", "GRHL3", "loci", "FGFR2", "gene expression", "mutation", "candidate gene", "phenotype" and "BMP". The dentition-related terms used with a higher than average number of citations were "dental agenesis", "microdontia", "dental anomaly", "malocclusion" and "maxillary lateral incisor". The most common terms used in relation to CLP treatment procedures were "palatoplasty", "orthognathic surgery", "orthodontic treatment", "NAM", "rhinoplasty" and "maxillary advancement". The term "speech outcome" was associated with the terms "hypernasality", "pharyngeal flap", "velopharyngeal insufficiency" and "palatoplasty".
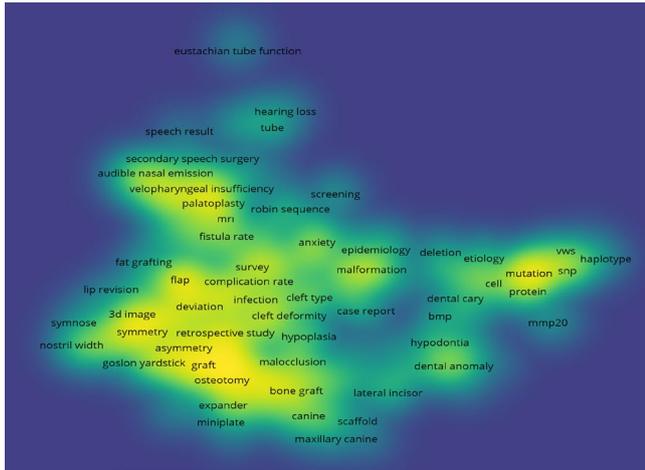
Figure 2

Map of 449 terms included in the abstracts of the articles.

An analysis of the 3,819 keywords revealed that the most frequently used keywords after the search keywords were "palatoplasty", "velopharyngeal insufficiency" and "speech" (Table 3).

Table 3.

The 20 most frequently used author keywords in articles.

Keywords	Occurence (% of 2696 publications)
Cleft Palate	439 (16.28%)
Cleft Lip and Palate	259 (9.60%)
Cleft Lip	248 (9.19%)
Cleft	80 (2.96%)
Unilateral Cleft Lip and Palate	60 (2.22%)
Palatoplasty	53 (1.96%)
Velopharyngeal Insufficiency	49 (1.81%)
Speech	38 (1.40%)
Alveolar Cleft	36 (1.33%)
Distraction Osteogenesis	32 (1.18%)
Alveolar Bone Grafting	32 (1.18%)
Pierre Robin Sequence	30 (1.11%)
Quality of Life	28 (1.03%)
Cephalometry	28 (1.03%)
Maxilla	27 (1.00%)
Orthognathic Surgery	27 (1.00%)
Children	27 (1.00%)
Genetics	25 (0.92%)
Orthodontics	24 (0.89%)
Craniofacial	24 (0.89%)

Figure 3 provides a visualization of the most frequent keywords above the threshold in the articles. A detailed assessment of the keywords revealed "hypodontia", "supernumerary teeth" and "tooth agenesis" to be the most frequently used keywords related to dental abnormalities. "Pierre Robin syndrome", "velocardiofacial syndrome" and "Van der Woude syndrome" were the most common syndrome types in

the articles discussing syndromes. "IRF6", "single nucleotide polymorphism" and "gene expression" were the most frequently used keywords in the studies of CLP and genetics.
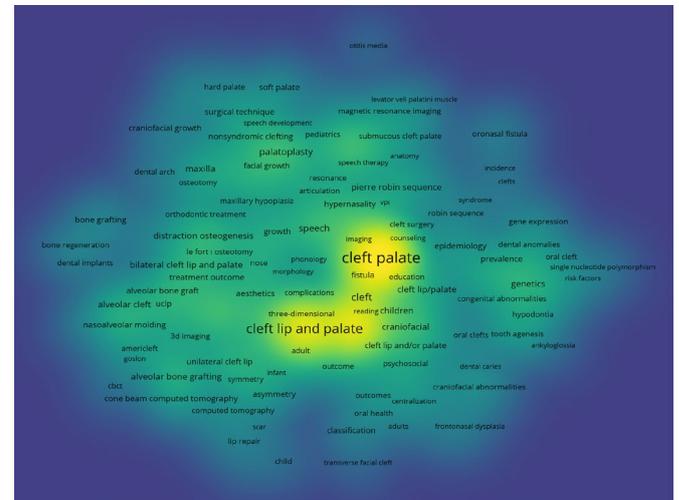

Figure 3

Map of the 255 most frequently used author keywords in the articles.

Besides cleft lip and palate, the keywords that had strong co-occurrence links with "3D" were "Le Fort I osteotomy", "presurgical orthopedics" and "orthognathic surgery". "Psychosocial adjustment" had co-occurrence links with the terms "speech perception" and "esthetics".

When the mean publication year of the keywords was examined, it was found that the keywords "nonsyndromic clefting", "orthopedic treatment", "palatal development", "surgical complications", "infant orthopedics" and "speech development" were identified more often in CLP articles from 2018 and 2019 than from other years.

In an analysis of the number of articles they produced, the most productive authors were Bing Shi, John B. Mulliken and Nicola M. Stock. Figure 4 presents the citation map of the 125 authors who were included in at least 0.3% of the articles. Regarding articles with multiple authors, 8,397 of the 8,440 authors were found to have collaborate with other researchers at various levels.
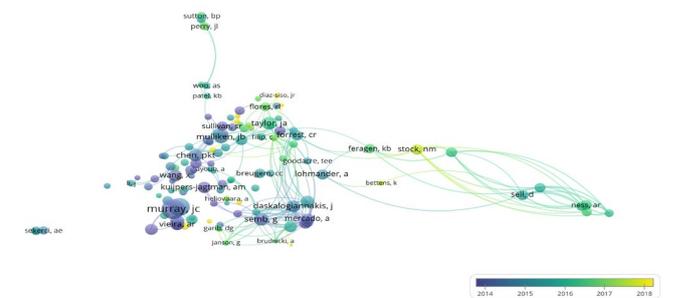

Figure 4

Map of the citation links of the 125 most prolific authors in the articles. The color scale below represents the mean years of the author's articles.

DISCUSSION

In the present study, an analysis is made of the subject trends in CLP-related articles, the citations of the authors and the contributions of various disciplines to literature, and the origins of publications. The CLP topic was chosen for the present study as an extensive subject that involves various disciplines.

Bibliometric studies require the establishment of limits for the subject among a great number of articles. To limit the sources, bibliometric tools such as Impact Factor, Eigenfactor score, CiteScore and H-index are used, in addition to the SJR indicator [11]. The preference for the SJR indicator in the present study is based on SJR's broad, open access database and its evaluation of citations in a different way, excluding the impacts of self-citations in journals [12].

An analysis of keywords revealed that recent articles have mostly discussed treatment procedures, the most common of which was "Palatoplasty", while treatment techniques such as "Alveolar Bone Grafting", "Distraction Osteogenesis" and "Orthognathic Surgery" were among the keywords most frequently encountered over the past decade. Similar keywords were also among the most common in the publications from between 2000 and 2011, indicating that the subject of treatment techniques has remained popular [8]. The term "Nasoalveolar Molding" (NAM) – a CLP treatment technique – recorded frequent use. There have been recent technological developments in this field in the last decade. Quan Yu et al. [13] introduced a novel NAM method based on 3D digital surface imaging for patients with a unilateral CLP. Recent studies have evaluated the effectiveness of presurgical nasoalveolar molding appliances manufactured using 3D printers [14], and have compared various CAD/CAM molding therapies [15]. Despite the above mentioned developments regarding the NAM technique, such subjects do not rank among the most discussed subject matters.

In addition to treatment techniques, as one of the most discussed subjects in publications, the publication themes investigating CLP and the genetics have been discussed frequently. New researches of genetics started to gain prominence in literature between 2000 and 2011, attracting increasing interest over time [8]. Researchers have identified new links, genetic loci and variations that may be involved in the etiology of orofacial cleft, and cleft lip and palate [16,17]. We believe that such developments in CLP and genetics might have led to an overall increase in the number of citations received by publications over the last decade.

Researchers have long been interested in the effects of CLP on dental abnormalities, malformations and occlusion, and this topic has become one of the most discussed and cited in literature, particularly over the last decade [6,8]. In addition to abnormalities, there have been recent studies relating to the treatment of dental cavities

and dental plaque, and oral microbiology and oral hygiene among cleft patients [18–20]. Besides studies on dental anomalies and NAM, orthodontics-related articles on CLP among the 100 most cited articles included in the present study evaluated facial asymmetry and variations with 3D approaches [21,22]. Meyer-Marcotty et al. [22] evaluated the degree of the facial asymmetry, the localization of the asymmetric regions and visual impact of asymmetry between patients with CLP and control groups using the 3D landmark-independent method. Bugaighis et al.[21] determined facial variations between children with unilateral cleft lip and palate, bilateral cleft lip and palate, unilateral cleft lip and alveolus, isolated cleft and palate and control groups using stereophotogrammetry and geometric morphometry.

Unlike in the previous bibliometric analyses in this field, the present study included an equal number of journals from disciplines that differed in terms of their subject matters, and thus compared the contributions of different disciplines to the literature. Although authors from different disciplines may submit studies to journals in several fields, the journals were classified into specific subject matter categories by the database and by the authors. Accordingly, it is seen that Oral Surgery; Plastic, Esthetic and Reconstructive Surgery; and Orthodontics sources, in which focus on primarily on treatment procedures, have apparently contributed more to CLP literature than other disciplines. This is because the Cleft Palate Craniofacial Journal, which is specific to the subject, and the Plastic and Reconstructive Surgery and the Journal of Cranio-Maxillofacial Surgery, in which the focus is on treatment procedures, are among journals with the highest number of articles specialized in these areas.

The present study found that most of the authors identified collaborated with other researchers at various levels. Researchers specialized either in the same or different disciplines have worked together on subjects requiring multidisciplinary approaches, as in the studies investigating the cleft etiology [23], in those evaluating innovative treatment approaches [14], and those assessing language development in CLP patients in literature [24]. In a similar vein to the use of promising artificial intelligence applications in healthcare following the developments in computer science [25] and the manufacture of novel biomaterials through basic science researchers and clinicians [26], we believe that the development of technologies and prevalent use of novel methods may lead researchers from different disciplines to work together in stronger interdisciplinary collaborations in the future.

As is the case with many of the studies conducted in different fields the United States was the most productive country in CLP literature [27,28]. The scientific productivity of developing countries in which

there are few publications may be affected by the several challenges faced by researchers [29]. The number of studies and the productivity of countries varies according to the number of research centers, the gross national product per capita (GNP) and the R&D expenditures [29,30].

There are a number of limitations to the present study. First, a set number of journals was selected to ensure the inclusion of an equal number of sources from all disciplines. Although we assessed the bibliometric analysis results of the publications with high SJR indicators in their categories, the analysis results may be more diversified if publications from a larger number of journals are included. Publications from the identified journals were registered in the WoS database in the period in which the search was made using the existing or former names of the journals, although it should be noted that names may be subject to change. Secondly, We searched the search keywords in the "TOPIC" section to obtain comprehensive results. Thus, we needed to include publications where the search terms were in keyword plus in addition to title, abstract, author keywords. Although keyword plus is a good option developed by Web of Science based on the power of cited-reference, it is not a convenient function for bibliographic studies with a large dataset as in our research. The results presented based on keyword plus may not always give the specifics directly related to the subject. In studies with a small data set, it is possible to examine each of the included publications. However, it may not be possible to examine each of the publications in the large data sets searched over a wide period. We believe that software specific to bibliometric research and enabling data treatment can fill this gap by providing more detailed and accurate results. Also, publications that do not contain search keywords in their titles, abstracts, author keywords and keywords plus but are related to CLP could not be evaluated in our research. The results obtained may differ depending on the time and search method and can not be generalized. It should be considered that reproducibility may be difficult in the selected time interval due to differences in publication times, as in early access articles. Furthermore, a detailed assessment may be challenging due to the overlapping of keywords on the visualized maps created by the software.

CONCLUSION

When analyzed the trends in numerical terms, it was found that CLP-related articles have mainly focused on treatment procedures. Among the terms that provided a more detailed opinion (specific to subject), those with a higher number of citations were mainly related to the CLP and genetics relationship. The current approaches, 3D printing, digital imaging methods and CAD/CAM applications have been discussed in only a limited number of studies in literature.

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