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Global research trends in complex oral sensitivity disorder: A systematic bibliometric analysis of the structures of knowledge

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Abstract

Objectives: A systematic bibliometric analysis of the structure of knowledge was performed to investigate the co-word analysis, the co-citation analysis, and the social network analysis regarding complex oral sensitivity disorder (COSD).

Methods: Web of Science database from 1985 to 2018 was systematically searched to identify all relevant articles using the MeSH terms "complex oral sensitivity disorder" and all synonyms used in the literature. We included original articles, review articles, letters to the editor, and book chapters in the English language and in 27 different ISI categories of medical sciences. Several bibliometric indicators were used.

Results: The co-word analysis identified 741 KeyWords Plus (KWP) grouped into 4 different clusters. The terms "pain," "management," "prevalence," and "efficacy" reached the highest centrality, whereas the top 10 KWP had a frequency of 7%-29% in 443 articles. Over a period of 32 years, a complex thematic evolution occurred, going from 2 to 6 different themes, and the KWP migration rate from one cluster to another ranged from 11% to 100%. The co-citation network analysis based on the complete reference list (5932 references) of 443 articles identified only 2 clusters for journals, authors, and articles. The most prominent co-cited journal was "Oral Surgery Oral Medicine Oral Pathology" (centrality: 171.75), the most co-cited author was "Grushka M" (centrality: 330.95), and the most co-cited article was "Grushka M. *Oral Surg Oral Med Oral Pathol* (1987) **63**:30-36" (centrality: 269.79). On the other hand, the direct citation network revealed that "Scala A et al, 2003" reached both the highest global citation score (GCS = 231) and local citation score (LCS = 161). Lastly, the social network analysis revealed an isolated collaboration among groups of authors, or countries or institutions. The worldwide collaboration analysis indicated that United States-Israel and United Kingdom-Italy were the most collaborative countries.

Conclusions: The structure of knowledge of publications on COSD revealed that research in this field has been dominated by few core topics and a limited collaboration among authors and institutions from different countries. More multicenter studies on COSD are warranted in the near future when launching new projects.

KEYWORDS
bibliometric, BMS, complex oral sensitivity disorder, COSD, intellectual structure, systematic

1 | INTRODUCTION

Complex oral sensitivity disorder (COSD) is an idiopathic and chronic medical condition, characterized by discomfort in the oral cavity, and by the absence of any local and/or systemic diseases, alterations in blood tests, and/or any significant findings in radiologic imaging.[1]

With the rapid development and application of new technologies, such as functional magnetic resonance, and new clinical studies on systemic implications and therapeutic strategies, there has been a dramatic increase in the science of COSD mainly in terms of pathophysiology[2,3] and management.[4,5] Therefore, it is imperative that clinicians continue to enhance and upgrade their knowledge by following emerging research on this topic. However, despite this important scientific progress, until recently, not much was known regarding the global trend of scientific research on COSD.

A new scientific approach was recently employed to answer several crucial questions regarding research related to COSD, such as what are the most productive countries, institutions, and departments? What are the highest contributing journals and authors? What are the most cited keywords and articles? What was the publications' research trend on COSD over the past 30 years? And finally, how can all these data be interpreted in a quantitative manner and be visually displayed in a single article?[6]

To address all these questions, a systematic bibliometric analysis approach was performed. This analysis consisted of a set of mathematical and statistical methods that utilize specific indicators to obtain information regarding the output of research activity from written publications,[7] thereby playing a pivotal role in evaluating research trends and keeping scientists up to date with a specific scientific field.[8]

Although bibliometrics is mainly known for quantifying the scientific production and measuring its quality and impact, it is also useful for displaying and analyzing the intellectual, conceptual, and social structures of research as well as their evolution and dynamic aspects.[9-11]

In this way, bibliometrics aims to describe how specific disciplines, scientific domains, or research fields are structured and how they evolve over time. In other words, bibliometric methods help to map the science (so-called science mapping) and are very useful in the case of research synthesis, especially for systematic ones.[12,13]

The results of the bibliometric analysis of the framework on COSD have demonstrated that Italy has been the leading country in COSD research, whereas the University of Turku in Finland and the Journal of Oral Pathology and Medicine were the most productive institution and source, respectively.[6] Despite the annual increasing trend of publications on COSD, the collaboration among different countries was still poorly represented.[6]

However, other questions are yet to be elucidated on COSD, such as what is the keyword co-occurrence network? What are the motor themes (thematic map) in existing studies? What are the developing topic trends (thematic evolution) currently? What is the co-citation network for authors, articles, and journals? What is the collaboration network among authors, institutions, and countries worldwide? We have tried to answer these questions by employing a new quantitative and visual analysis which has included the map of knowledge on COSD.

The aim of this study was to analyze the three structures of knowledge (K-structures): intellectual, conceptual, and social, in order to create a research map and highlight the processes, which have led to the generation of new knowledge in the field of COSD. This new analysis may offer new prospectives for oral medicine specialists and orofacial pain practitioners when planning future research projects.

2 | MATERIALS AND METHODS

2.1 | Selection strategy

Our investigation followed the Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidelines,[14] and consisted of a computerized bibliometric analysis from January 1985 to December 2018 regarding articles on COSD retrieved from the Web of Science (WoS) database as previously illustrated.[6]

Searches focused on one main topic: complex oral sensitivity disorder (COSD), also commonly known as burning mouth syndrome (BMS). To identify all publications related to COSD, we included all synonymous MeSH terms used over the past decades to describe the same entity, by including the Boolean separator "OR," as previously described.[6] In addition, all included articles were examined manually (by authors GF and MDM) to identify articles that were not relevant to the quantitative analyses because these were either unrelated to the main topic or the nature of the disease was not considered idiopathic.[6]

2.2 | Selection criteria

After eliminating non-pertinent journals and non-relevant articles, two investigators (GF and MA) independently verified data entry and collection, and then reformulated the dataset for bibliometric analyses. Any disagreement was discussed and resolved by consensus between the two investigators, and if not reached, a third investigator (AP) was consulted to resolve the issue.

The systematic review included the following: (a) original articles, review articles, letters to the editor, and book chapters only; (b) articles in the English language only; (c) articles in 27 different ISI categories of medical science; (d) articles reporting any aspect of COSD; and (e) articles whose title included at least one of the above-mentioned MeSH terms.[6]

Selection strategy and criteria, as well as results of the pertinent included studies, were depicted in Figure 1 of our previous investigation.[6]

2.3 | Data extraction

Studies that met the inclusion criteria were independently reviewed by two investigators (GF and MA) who extracted and analyzed the following relevant bibliometric indicators: keyword co-occurrence and related clusters, co-citation network including the historical direct citation network, and the collaboration network in the world. All these indicators represent the foundation of the structure of knowledge.

The standard competition ranking (SCR) is used for ranking purposes, and only the first top twenty ranked data for each analyzed bibliometric indicator were taken into consideration. If the measurements of bibliometric analysis had the same ranking number, then a gap was left in the following ranking numbers. The publication was assigned the country, the institution, and the department of the corresponding author.

2.4 | Network analysis

We constructed a co-citation network (sociogram) consisting in a graphic representation of the most significant and intense relationships between members of a network that are represented by "vertices" or "nodes." Vertices represent high-frequency items (eg, cited articles, or cited sources, or cited authors), and their size shows the occurrence frequency. The smaller size about a node, the lower occurrence the frequency is. On the other hand, "lines" or "edges" represent the connection relationship or interaction between vertices, which exists in the same article, and their thickness reflects the degree or intensity of co-citation between vertices. The thicker the edges between two vertices, the stronger the connection is.[15,16]

In this network analysis performed on the bibliographic data from scientific publications on COSD, it is possible to distinguish isolated nodes that have no connection with other nodes whatsoever, and nodes or groups of nodes that are interconnected directly or indirectly via intermediaries. Some groups of nodes (each with a different color) may show a degree of interconnection with other groups.[16] Although different algorithms (clustering methods) exist to identify these subcomponents, this study used Louvain community detection algorithm.[17]

Lastly, the indices of centrality help to identify the most important nodes in a network and the propensity of two vertices that are connected to be both connected to a third vertex.[13]

2.5 | The conceptual structure of the COSD field: co-word analysis

The co-word analysis is based on the co-occurrence frequency of keywords of interest in an entire collection. Its aim is to map the conceptual structure of a framework using the keyword co-occurrences

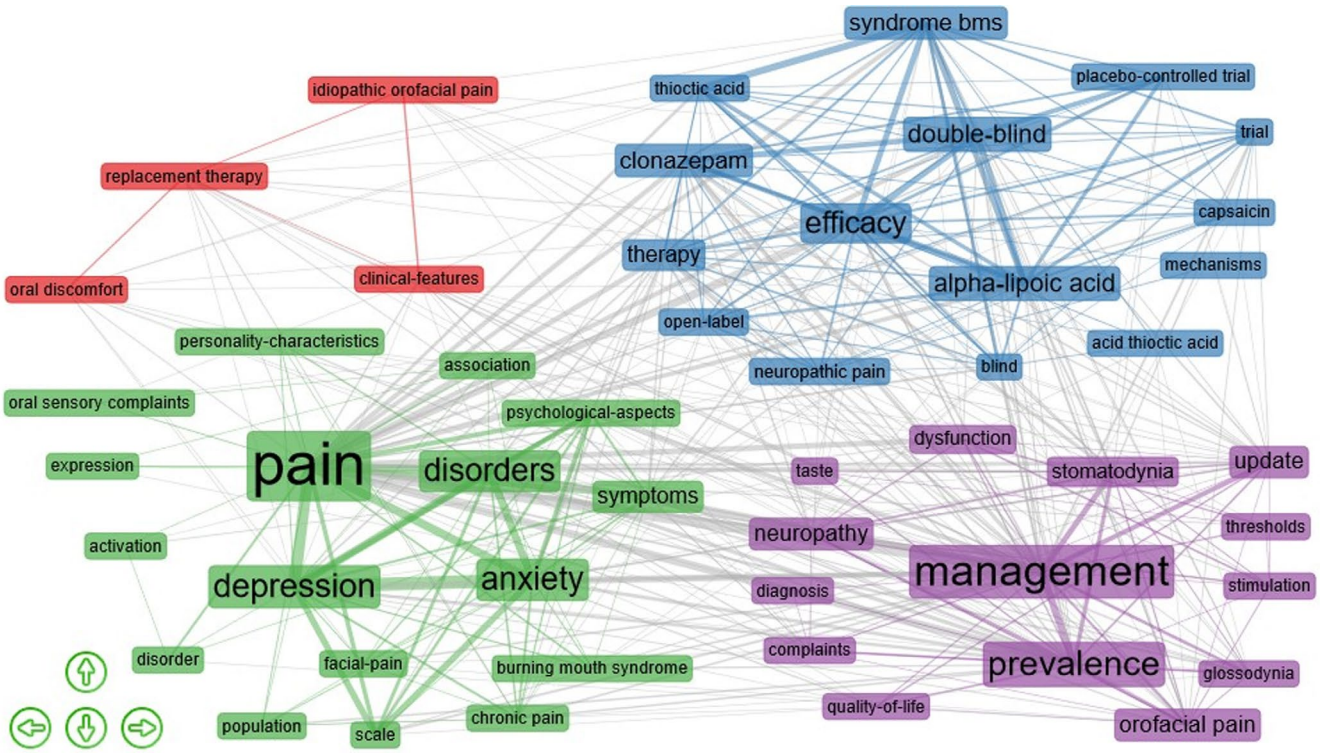


FIGURE 1 Network visualization of KeyWords Plus (KWP) co-occurrence. The thickness of the connecting line between 2 keywords represents the strength of co-occurrence. The size of the KWP represents the index of their centrality

in a bibliographic collection. In our collection of articles on COSD, the higher the co-occurrence frequency of two keywords, the closer their relationship is. We mapped a co-word network by analyzing the co-occurrence frequency of keywords in our entire collection and investigated specific research areas of interest on COSD.[8]

The analysis can be performed through dimensionality reduction techniques[18] or co-occurrence network analysis.[10]

Here, we used co-occurrence network to draw a conceptual structure of the field and hierarchical clustering to identify clusters of documents which express common concepts. Results are plotted on a thematic map and evolution, by employing the top 500 KeyWords Plus (KWP) (which are generated independently of the title and author keywords and include the traditional keywords plus additional terms to describe the article's contents with greater details and variety), a minimum cluster frequency of 5, and minimum weight index of 0.1, divided into different clusters.

In co-occurrence network, we used association as normalization parameter and Louvain method as clustering algorithm, and 50 as the number of nodes without removing isolated nodes and having each node with a minimum of 2 edges (which represent how often two items are linked together). In the thematic evolution, we set three "time-slicing" periods, based on overall time distribution of publications: 1986-1997; 1998-2007; and 2008-2017, in order to identify thematic areas, defined as a group of evolved themes across different subperiods. Based on the interconnections among them, one theme could belong to a different thematic area, or could not come from any.[19]

We also fabricated a thematic map, in which we can find four kinds of themes according to the quadrant in which they are placed[19]: (a) Themes in the upper-right quadrant are both well-developed and important for the structuring of a research field. They are known as the motor themes of the specialty and are conceptually closely related; (b) themes in the upper-left quadrant have well-developed internal ties but unimportant external ties and so are of only marginal importance for the field; (c) themes in the lower-left quadrant are both weakly developed and marginal; and (d) themes in the lower-right quadrant are important for a research field but are not developed.

2.6 | The intellectual structure of the COSD field: co-citation analysis

Scientific publications regularly contain references to other scientific works. This generates further networks, such as co-citation or coupling networks.[20] These networks are analyzed in order to capture meaningful properties of the underlying research system and, in particular, to determine the influence of bibliometric units such as scholars and journals.

Two articles are said to be bibliographically coupled if at least one cited source appears in the bibliographies or reference lists of both articles.[20]

We refer to co-citation of two articles when both are cited in a third article. Thus, co-citation can be seen as the counterpart of bibliographic coupling. The historiographic map is a graph to represent a chronological network map of most relevant direct citations resulting from a bibliographic collection.[21] The function generates a chronological direct citation network matrix which can be plotted against a number of nodes of 20 to better depict the relationship among the top 20 authors included in our collection.

In the co-citation network for articles, authors, and sources, Louvain method was used as a clustering algorithm, a number of nodes of 50, and a minimum edge strength of 20 (approximately 5% of the entire collection of articles on COSD).

2.7 | The social structure of the COSD field: collaboration network analysis

Social network analysis, also known as network mapping, is a method to study network centralization by analyzing nodes and links.[8] Scientific collaboration network is a network where nodes are represented by articles, or sources, or authors, or institutions, or countries and links are co-authorships, as the latter is one of the most well-documented forms of scientific collaboration.[22] Therefore, the links represent the collaboration of these nodes. The size and location of nodes depend on the total occurrence frequency of the items (eg, authors or institutions or countries). The thickness of the links between the nodes indicates the collaboration frequency of nodes.[8,13]

In the collaboration network among authors, or institutions or countries, Louvain method was used as a clustering algorithm, a number of nodes of 50, and minimum edges of 2 in order to avoid isolated and "one-time" collaboration. Isolated nodes were removed.

2.8 | Cluster-level analysis

At the end of the structure of knowledge analysis, we developed a set of clusters as conglomerates of different scientific aspects. The co-word analysis provides clusters of groups of textual information representing the conceptual base of different topics in the field of COSD, whereas the co-citation analysis provides clusters of groups representing the intellectual base of different subfields in COSD.[19]

2.9 | Statistical analysis

Data were collected and exported into open-source science mapping software called bibliometrix R package[13] for generating descriptive analyses, statistical graphs, and science maps.

TABLE 1 Top 10 KeyWords Plus co-occurrence in COSD field from 1986 to 2018

Cluster	Centrality	KeyWords Plus	Pain	Management	Prevalence	Efficacy	Update	Disorders	Depression	Syndrome BMS	Neuropathic pain	ALA
2	42.1	Efficacy	17	6	5	38	5	0	2	15	6	18
	20.2	Syndrome BMS	11	6	3	15	3	0	1	28	1	15
	16.7	Neuropathic Pain	0	3	0	6	3	1	0	1	21	3
	16.1	ALA	11	2	3	18	3	0	1	15	3	27
3	306.1	Pain	130	27	20	17	7	21	28	11	0	11
	25.3	Disorders	21	14	10	0	3	47	20	0	1	0
	25.0	Depression	28	13	8	2	2	20	49	1	0	1
4	119.0	Management	27	75	18	6	12	14	13	6	3	2
	80.4	Prevalence	20	18	55	5	5	10	8	3	0	3
	30.0	Update	7	12	5	5	34	3	2	3	3	3

Abbreviation: ALA, alpha-lipoic acid.

3 | RESULTS

3.1 | Conceptual structure

Our bibliometric analysis revealed the presence of 741 KWP used in our collection,[6] classified into 4 different clusters, in which these terms are based on the same topics (co-occurrence). The KWP with the highest centrality (pain: 306.1; management: 119.0; prevalence: 80.4; and efficacy: 42.1) is mainly localized in clusters 3, 4, and 2 respectively (Figure 1). They co-occurred with a frequency ranging from 0 to 130 times for pain, from 2 to 75 times for management, from 0 to 55 times for prevalence, and from 0 to 38 times for efficacy (Table 1).

Of all 741 KWP, only 485 occurred once. The top 10 KWP (pain, management, prevalence, depression, disorders, anxiety, efficacy, update, and therapy, and neuropathy) occupy a frequency in the entire collection of 443 articles ranging from 7% to 29% within only 4 out of 10 different clusters: "pain," "prevalence," "efficacy," and "neuropathy" (Table 2).

The period of 32 years considered for our collection of articles was split into three periods: 1986-1997; 1998-2007; and 2008-2018, as highlighted in Figure 2. In the first period 1986-1997, research identified only two thematic areas (Figure 2A): "management" as motor theme, and "pain" as emerging theme (Figure 2B), whereas in the second period 1998-2007, "pain" evolved as basic and transversal theme, and five new thematic areas appeared: "burning mouth syndrome," "symptoms," "neuropathic pain," "prevalence," and "glossodynia" (Figure 2C). The previous thematic area "management" split into two new thematic areas of "symptoms" and "pain" (Figure 2A). Finally, in the third period 2008-2018, three new thematic areas ("efficacy," "orofacial pain," and "disorders") developed from previous thematic areas "neuropathic pain" and "pain," whereas five new thematic areas ("acid," "expression," "disorder," "neuropathy," and "clinical features") developed de novo (Figure 2D). As described in Table 3, the percentage of KWP migrated from one cluster to another varied from 11% (therapy) to 100% (burning mouth syndrome).

3.2 | Intellectual structure

The co-citation network and clustering analyses were performed to identify the most influential journals, authors, and articles, each of the three being grouped in only two main clusters. In the journals' clusters, the top 5 co-cited journals with the highest centrality were "Oral Surgery Oral Medicine Oral Pathology" (171.75), "Pain" (168.48), and "Journal of the American Dental Association" (60.25) in cluster 1. The other 2 journals with the highest centrality were "Journal of Oral Pathology and Medicine" (159.51) and "Journal of Orofacial Pain" (57.11) grouped in cluster 2. The top 5 co-cited articles with the highest centrality were "Grushka M, 1987-2" (269.79) and "Grushka M, 1987-1" (87.79) in cluster 2; and "Bergdahl M, 1999" (177.82), "Scala A, 2003-1" (125.22), and "Lauria G, 2005-1" (113.75) in cluster 1.

TABLE 2 Description of the KeyWords Plus occurrence in COSD field from 1986 to 2018 ordered by cluster number

Occurrences	(%)*	KeyWords Plus	Cluster	Cluster label	Occurrences	(%)*	KeyWords Plus	Cluster	Cluster label	Occurrences	(%)*	KeyWords Plus	Cluster	Cluster label
14	3	Clinical features	1	Clinical features	5	1	Contact hypersensitivity	2	Pain	5	1	Sensitivity	5	Disorder
14	3	Complaints	1	Clinical features	5	1	Women	2	Pain	7	2	Mechanisms	6	Mechanisms
13	3	Taste	1	Clinical features	5	1	Oral galvanism	2	Pain	7	2	Oral sensory complaints	7	Oral sensory complaints
9	2	Idiopathic orofacial pain	1	Clinical features	31	7	Neuropathy	3	Neuropathy	5	1	Pathophysiology	7	Oral sensory complaints
130	29	Pain	2	Pain	23	5	Dysfunction	3	Neuropathy	6	1	Acid	8	Acid
75	17	Management	2	Pain	15	3	Stimulation	3	Neuropathy	7	2	Expression	9	Expression
49	11	Depression	2	Pain	8	2	Thresholds	3	Neuropathy	6	1	Receptors	9	Expression
47	11	Disorders	2	Pain	6	1	Nerve	3	Neuropathy	5	1	Receptor	9	Expression
47	11	Anxiety	2	Pain	6	1	Hyperalgesia	3	Neuropathy	5	1	Rat	9	Expression
30	7	Symptoms	2	Pain	6	1	Perception	3	Neuropathy	38	9	Efficacy	10	Efficacy
23	5	Psychological aspects	2	Pain	6	1	Blink reflex	3	Neuropathy	32	7	Therapy	10	Efficacy
20	5	Scale	2	Pain	5	1	Hypersensitivity	3	Neuropathy	31	7	Double blind	10	Efficacy
19	4	Burning mouth syndrome	2	Pain	55	12	Prevalence	4	Prevalence	31	7	Clonazepam	10	Efficacy
17	4	Chronic pain	2	Pain	34	8	Update	4	Prevalence	28	6	Syndrome BMS	10	Efficacy
17	4	Glossodynia	2	Pain	27	6	Stomatodynia	4	Prevalence	27	6	Alpha-lipoic acid	10	Efficacy
15	3	Replacement therapy	2	Pain	27	6	Orofacial pain	4	Prevalence	21	5	Neuropathic pain	10	Efficacy
10	2	Facial pain	2	Pain	10	2	Population	4	Prevalence	17	4	Thioctic acid	10	Efficacy
10	2	Association	2	Pain	10	2	Quality of life	4	Prevalence	15	3	Trial	10	Efficacy
8	2	Profiles	2	Pain	9	2	Diagnosis	4	Prevalence	14	3	Blind	10	Efficacy
7	2	Salivary flow	2	Pain	9	2	Personality characteristics	4	Prevalence	13	3	Open label	10	Efficacy
7	2	Allergy	2	Pain	7	2	Saliva	4	Prevalence	12	3	Placebo controlled trial	10	Efficacy
6	1	Salivary flow rate	2	Pain	7	2	Placebo	4	Prevalence	10	2	Capsaicin	10	Efficacy
6	1	Stress	2	Pain	6	1	Menopause	4	Prevalence	6	1	Meta-analysis	10	Efficacy
6	1	Oral discomfort	2	Pain	6	1	Psychopathology	4	Prevalence	6	1	Clinical trial	10	Efficacy

(Continues)

TABLE 2 (Continued)

Occurrences	(%)*	KeyWords Plus	Cluster	Cluster label	Occurrences	(%)*	KeyWords Plus	Cluster	Cluster label	Occurrences	(%)*	KeyWords Plus	Cluster	Cluster label
5	1	Sleep disturbance	2	Pain	5	1	Profile	4	Prevalence	6	1	Gabapentin	10	Efficacy
5	1	Had scale	2	Pain	9	2	Disorder	5	Disorder	6	1	Acid thioctic acid	11	Acid thioctic acid
5	1	Interleukin 6	2	Pain	6	1	Activation	5	Disorder					
5	1	Questionnaire	2	Pain	5	1	Cortex	5	Disorder					

(%)*: percentage of KeyWords Plus occurrence in the collection of 443 articles.

On the other hand, the top 5 co-cited authors with the highest centrality were "Grushka M" (330.95) and "Lamey PJ" (116.86) in cluster 1, and "Bergdahl M" (53.23), "Scala A" (43.68), and " Jaaskelainen SK" in cluster 2 (32.28) (Table 4). These co-citation networks are depicted in Figure 3 based on the complete reference list (5932 references) in 443 articles on COSD, where the thickness is the interconnecting bar between two (or more) nodes and the height is the degree of interconnection(s) within these nodes.

The analysis of direct citation network revealed that "Scala A et al, 2003" reached both the highest global (GCS = 231) and local citation score (LCS = 161), followed by "Bergdahl M & Bergdahl J. 1999" with 198 GCS and 135 LCS and "Grushka M. 1987" with 208 GCS and 130 LCS (Table 5, Figure 4).

3.3 | Social structure

In the social structure of 443 included articles, 10 different clusters of authors were formed (Figure 5A). The most prolific authors per cluster were "Abe O." in cluster 5, "Toyofuku A." in cluster 4, and "Mignogna MD" in cluster 8, "Lopez-Jornet P." in cluster 9, "Sardella A." in cluster 2, "Boras VV" in cluster 3, and "Spadari F." in cluster 10. On the other hand, the most prominent institutions were "University of Turku" in cluster 3, "University of Milan" in cluster 6, and "University of Zagreb" in cluster 1 (Figure 5B). The countries reaching the highest centrality were United States (37.0) in cluster 5, United Kingdom (27.0) in cluster 1, Canada (19.09) in cluster 2, and Japan (10.0) in cluster 4 (Figure 5C).

Finally, the top three collaborations among countries present in our collection of 443 articles were United States-Israel: 5; United Kingdom-Italy: 4; United Kingdom-Ireland; and USA-Canada: 3 as depicted in Figure 6.

4 | DISCUSSION

The structure of knowledge analysis performed in this study led us to understand three fundamental structures in bibliometric analysis. We have identified the most influential documents and authors that have constituted the intellectual basis in the field of COSD. Similarly, the analysis of co-citation has highlighted the macrostructure behind the scientific knowledge in COSD, identifying areas of actual research and possible future developments.

In the conceptual structure, the co-word analysis shows the links between concepts through term co-occurrences and extracts research hotspots in a certain research field.[23] Our study showed that the top 10 out of 741 KWP occurred in our collection of articles in the range between 7% and 29%, with four of them (pain, management, prevalence, and efficacy) reaching the highest centrality and co-occurrence frequency. This means that over a period of 30 years, researchers have mainly covered these topics.

Our results support the scientific research trend mainly on possible etiopathogenetic factors and management. In our co-word

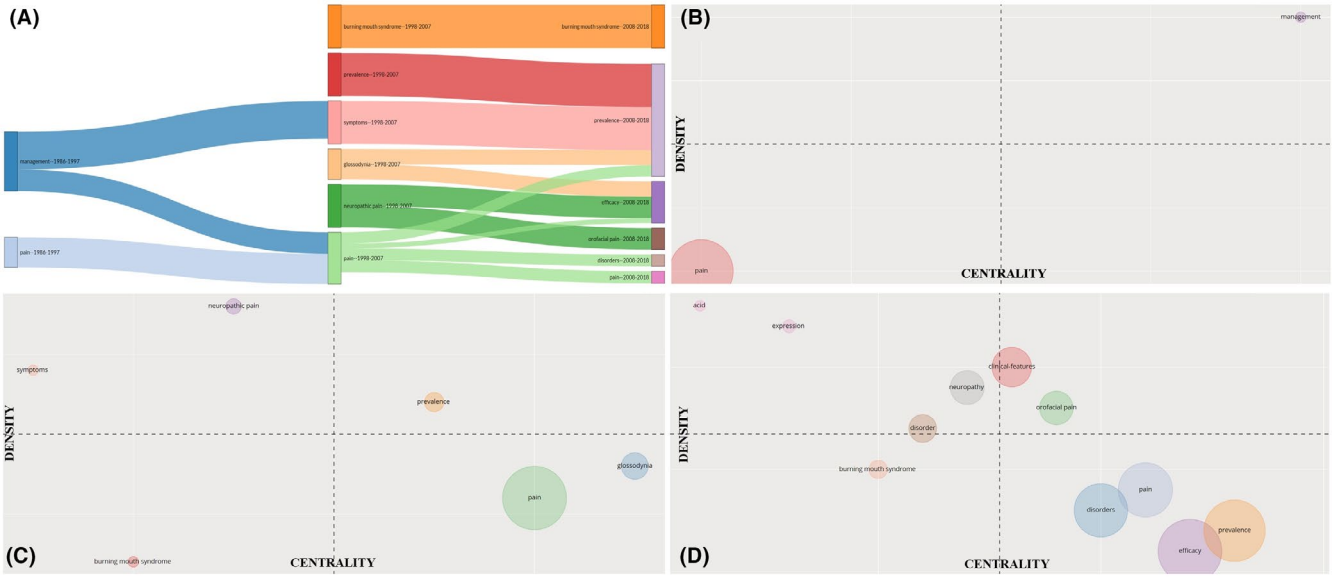


FIGURE 2 A, Thematic evolution of KeyWords Plus in field of research on COSD 1986-2018; B, time slice 1:1986-1997; C, time slice 2:1998-2007; and D, time slice 3:2008-2018. The size of the circle is proportional to the total frequency of the KeyWords Plus included in that specific cluster. The thickness of the bar is proportional to the weighted inclusion index

TABLE 3 Thematic evolution of KeyWords Plus

From	To	KeyWords Plus[a]	Weighted Inclusion Index[b] (%)
Burning mouth syndrome—1998-2007	Burning mouth syndrome—2008-2018	Burning mouth syndrome	100
Pain—1998-2007	Disorders—008-2018	Depression, disorders, anxiety	27
Glossodynia—1998-2007	Efficacy—2008-2018	Syndrome BMS	35
Neuropathic pain—1998-2007	Efficacy—2008-2018	Neuropathic pain	50
Pain—1998-2007	Efficacy—2008-2018	Therapy	11
Neuropathic pain—1998-2007	Orofacial pain—2008-2018	Orofacial pain	50
Management—1986-1997	Pain—1998-2007	Management	50
Pain—1986-1997	Pain—1998-2007	Pain, disorders, depression, Psychological aspects, anxiety	70
Pain—1998-2007	Pain—2008-2018	Pain	28
Glossodynia—1998-2007	Prevalence—2008-2018	Glossodynia	35
Pain—1998-2007	Prevalence—2008-2018	Management	26
Prevalence—1998-2007	Prevalence—2008-2018	Prevalence	100
Symptoms—1998-2007	Prevalence—2008-2018	Symptoms	100
Management—1986-1997	Symptoms—1998-2007	Symptoms	88

[a]KWP migrated from one cluster to another during the evolution period.

[b]Percentage of KWP migrated from one cluster to another based on the frequency of KWP present in the new cluster.

analysis, the most important clusters were cluster 2: "pain" and cluster 3: "neuropathy." Topics in both clusters have flourished over the past 15 years: The cluster "pain"—which includes terms such as anxiety, depression, psychological aspect, and management—is the most prominent, giving a large contribution not only in understanding the psychological factors[24-30] behind the possible etiopathogenesis of COSD but also in understanding the complexity of its management.[4,5,31,32]

On the other hand, the other major cluster "neuropathy"—which includes terms such as neuropathy, dysfunction, stimulation, threshold, nerve, and blink reflex—has explored different aspects of COSD etiopathogenesis mainly involving the peripheral and central nervous systems.[2,33-38]

When the whole time span was divided into different time slices, the complexity of research in the field of COSD was also more evident and supported by the thematic evolution analysis.

TABLE 4 Top twenty co-cited journals, authors, and articles

Rank by Centrality	Journals	Cluster	Centrality
1	Oral Surgery Oral Medicine Oral Pathology	1	171.75
2	Pain	1	168.48
3	Journal of Oral Pathology and Medicine	2	159.51
4	Journal of American Dental Association	1	60.25
5	Journal of Orofacial Pain	2	57.11
6	Oral Diseases	2	36.28
7	British Dental Journal	1	24.41
8	Critical Review Oral Biology & Medicine	2	22.56
9	British Medical Journal	1	13.46
10	Medicina Oral Patologia Oral y Cirugia Buccal	2	9.75
11	Journal of Dental Research	1	8.94
12	Journal of Oral Rehabilitation	2	2.28
13	Journal of Psychosomatic Research	2	1.90
14	Journal of Pain	2	1.69
15	Minerva Stomatologica	2	1.47
16	Cochrane database of systematic reviews	2	1.34
17	Clinical Journal of Pain	1	1.15
18	American Family Physician	2	0.96
19	Acta Odontologica Scandinavica	1	0.31
20	Archives of Oral Biology	2	0.26
	Articles[a]		
1	Grushka M, 1987-2; https://doi.org/10.1016/0030-4220(87)90336-7	2	267.79
2	Bergdahl M, 1999; https://doi.org/10.1111/j.1600-0714.1999.tb02052.x	1	177.82
3	Scala A, 2003-1; https://doi.org/10.1177/154411130301400405	1	125.22
4	Lauria G, 2005-1; https://doi.org/10.1016/j.pain.2005.03.028	1	113.75
5	Grushka M, 1987-1; https://doi.org/10.1016/0304-3959(87)90113-8	2	87.79
6	Lamey PJ, 1988-1; https://doi.org/10.1136/BMJ.296.6631.1243	2	59.43
7	Forssell H, 2002; https://doi.org/10.1016/S0304-3959(02)00052-0	1	44.20
8	Gremeau-Richard C, 2004; https://doi.org/10.1016/J.PAIN.2003.12.002	1	29.23
9	Gorsky M, 1991; https://doi.org/10.1016/0030-4220(91)90162-6	2	16.22
10	Basker RM, 1978-1; https://doi.org/10.1038/SJ.BDJ.4804107	2	7.61
11	Grushka M, 1998-1; https://doi.org/10.1016/S1079-2104(98)90345-6	1	5.40
12	Grushka M, 2002-1; Am Fam Physician. 2002;65:615-621	1	5.33
13	Browning S, 1987; https://doi.org/10.1016/0030-4220(87)90085-5	2	5.29
14	Jaaskelainen SK, 1997 https://doi.org/10.1016/s0304-3959(97)00140-1	1	3.67
15	Zakrzewska JM, 2005-1; https://doi.org/10.1002/14651858.CD002779.PUB2	1	2.41
16	Yilmaz Z, 2007; https://doi.org/10.1016/j.jocn.2006.09.002	1	2.11
17	Albuquerque RJ, 2006; https://doi.org/10.1016/J.PAIN.2006.01.020	1	1.92
18	Vanderploeg HM, 1987; https://doi.org/10.1016/0030-4220(87)90366-5	2	1.63
19	Femiano F, 2002-1; https://doi.org/10.1034/J.1600-0714.2002.310503.X	1	1.50
20	Grushka M, 1991-1; Dent Clin North Am. 1991;35:171-84	2	1.05
	Authors		
1	Grushka M	1	330.95
2	Lamey PJ	1	116.86
3	Bergdahl M	2	53.23
4	Scala A	2	43.68

(Continues)

TABLE 4 (Continued)

	Authors		
5	Jaaskelainen SK	2	32.28
6	Bergdahl J	2	27.95
7	Gremeau-Richard C	2	19.93
8	Sardella A	2	18.17
9	Zakrzewska JM	2	17.62
10	Lauria G	2	17.61
11	Forssell H	2	13.80
12	Gorsky M	1	10.00
13	Lopez-Jornet P	2	9.24
14	Basker RM	1	8.95
15	Femiano F	2	8.88
16	Woda A	2	5.21
17	Tammialasalonen T	2	4.02
18	Rojo L	1	2.18
19	Browning S	1	1.78
20	Eli I	1	1.75

[a]DOI number was available for all top 20 co-cited articles but 2. However, a full reference was provided for the two.

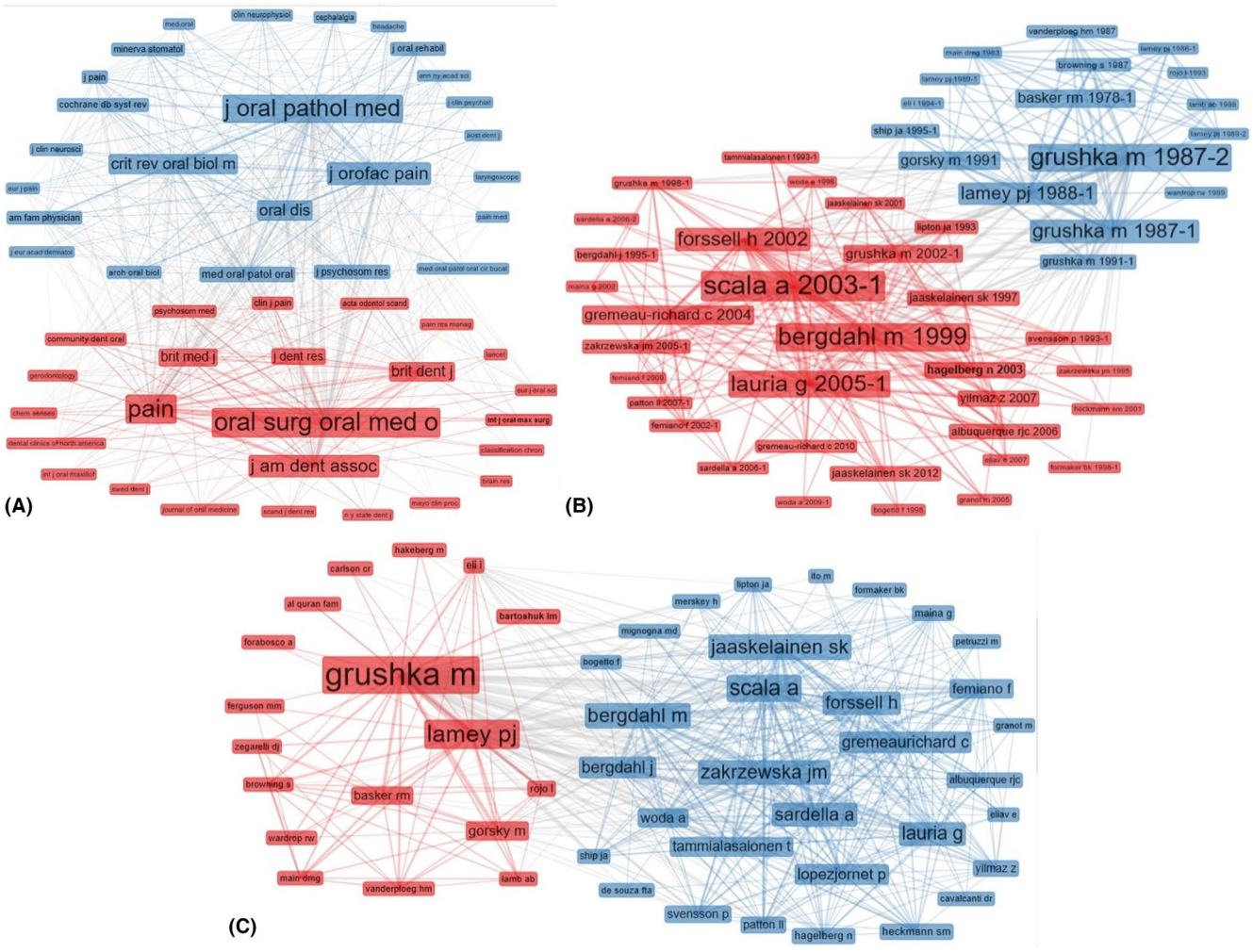


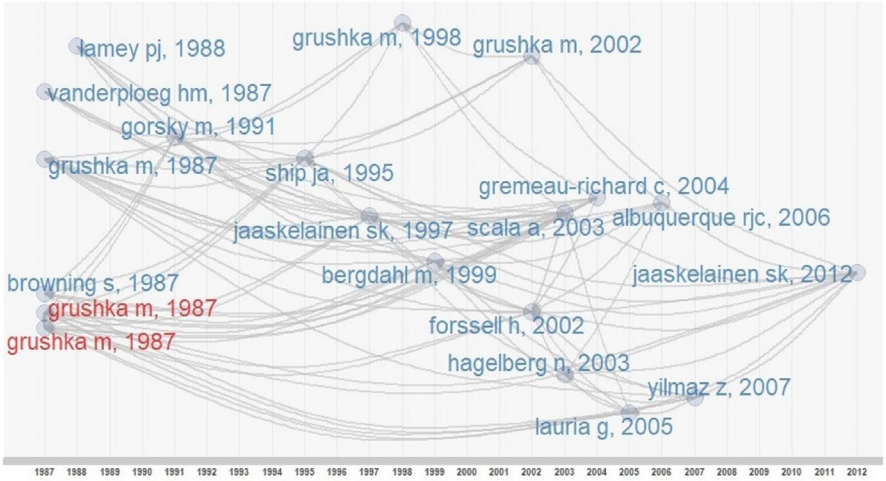
FIGURE 3 Co-citation network analysis with the identification of only 2 co-citation clusters for (A) journals, (B) authors, and (C) articles

TABLE 5 Historical direct citation network

Articles directly cited	LCS	GCS	ICR (%)
Scala A et al Crit Rev Oral Biol Med 2003;14:275-91.	161	231	69.70
Bergdahl M & Bergdahl J. J Oral Pathol Med 1999;28:350-4.	135	198	68.18
Grushka M. Oral Surg Oral Med Oral Pathol 1987;63:30-6.	130	208	62.50
Lauria G et al Pain 2005;115:332-7.	122	194	62.89
Lamey PJ & Lamb AB. Br Med J (Clin Res Ed) 1988;296:1243-6.	111	162	68.52
Grushka M et al Pain 1987;28:155-67.	107	107	100.0
Grushka M et al Pain 1987;28:169-84.	107	122	87.70
Forssell H et al Pain 2002;99:41-7.	100	162	61.73
Grushka M et al Am Fam Physician 2002;65:615-20.	87	112	77.68
Gremeau-Richard C et al Pain 2004;108:51-7.	85	132	64.39
Gorsky M et al Oral Surg Oral Med Oral Pathol 1991;72:192-5.	80	101	79.21
Ship JA et al J Am Dent Assoc 1995;126:842-53.	64	88	72.73
Jaaskelainen SK et al Pain. 1997;73:455-60	64	102	62.75
Jaaskelainen SK. Clin Neurophysiol. 2012;123:71-7	64	81	79.0
Browning S et al Oral Surg Oral Med Oral Pathol 1987;64:171-4.	63	80	78.75
Grushka M et al Oral Surg Oral Med Oral Pathol Oral Radiol Endod 1998;86:557-61.	61	87	70.11
Yilmaz Z et al J Clin Neurosci. 2007;14:864-71	60	102	58.82
Hagelberg N et al Pain 2003;101:149-54.	59	129	45.74
van der Ploeg HM et al Oral Surg Oral Med Oral Pathol 1987;63:664-8.	57	78	73.08
Albuquerque RJ et al Pain. 2006;122:223-34	56	92	60.87

Note: LCS is defined as how many times the author's articles included in this collection have been cited by other articles also in the collection.
GCS is defined as how many times the author's articles included in this collection have been cited, corresponding to the total citations (TC).
ICR represents the ratio between LCS and GCS.
Abbreviations: GCS, global citation score; ICR, intra-citation rate; LCS, local citation score.

FIGURE 4 Historical direct citation network



This demonstrated how themes, such as pain, evolved from a single and emerging theme to a basic and transversal theme, which in the last decade split in at least three other different themes, such as prevalence, efficacy, and disorders. While the thematic area "pain" remains stable over the period of 30 years, the thematic area "management" disappears after the first decade of research partly giving birth to another thematic area "symptoms" and partly merging into the thematic area "pain." This does not imply that scientists have stopped performing research on COSD management, but just that this single initial motor theme has evolved and expanded in different areas of research.

Interestingly, in the last decade, five new additional thematic areas appeared, two of which, such as "acid" and "expression," were highly specialized and isolated themes; they are well-developed internal ties but unimportant external ties and so are of only marginal importance for research on COSD. On the other hand, the new theme "clinical features"—along with "orofacial pain"—became

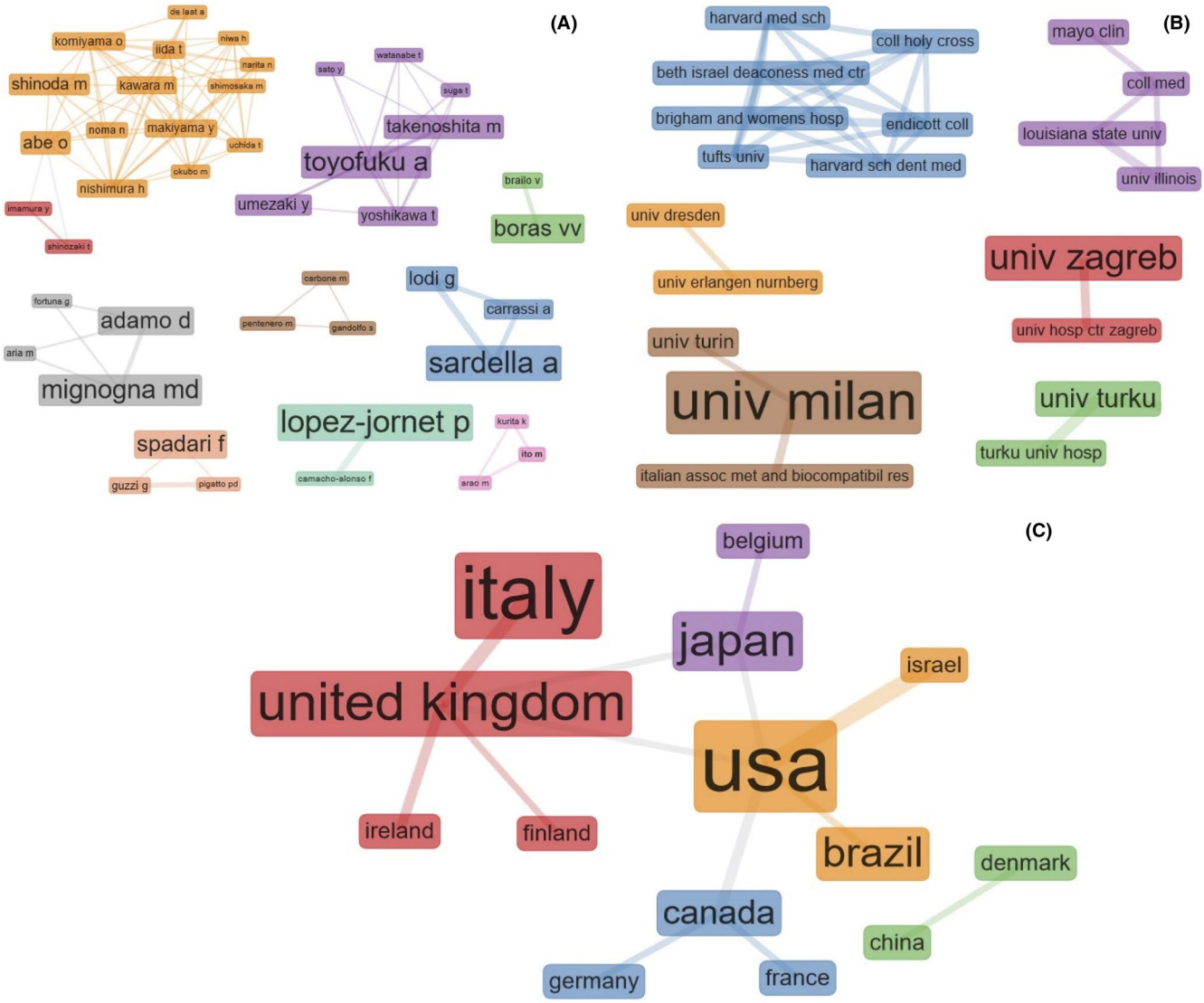


FIGURE 5 Collaborations network among (A) authors, (B) countries, and (C) institutions

a *motor theme*, that is, well-developed and important topics for the structuring of research in COSD. The thematic analysis demonstrated that research on COSD has highly progressed over the past 10 years, but still requires further development.

The intellectual structure analysis has revealed the knowledge base of initial research on COSD, in which co-citations express the relationship between documents. In our collection, 5 co-cited articles stand out from the rest in the co-citation network: "Grushka M, 1987-2" and "Grushka M, 1987-1" in cluster 2; and "Bergdahl M, 1999," "Scala A, 2003-1" (125.22), and "Lauria G, 2005-1" (113.75) in cluster 1.

Except for the article from Scala et al,[39] which is a review article on COSD, the other 4 articles were original articles, exploring different aspects of COSD. The two papers from Grushka M. published in 1987 were focused on the clinical features and possible local and systemic factors involved in COSD[40] as well as the intensity of pain and personality changes occurring in COSD patients.[26] Later on, Bergdahl M. published in 1999, a study on COSD in a large population of patients in Sweden, confirmed that several different local, systemic, and psychological factors were associated with COSD.[41]

On the other hand, the article from Lauria G. published in 2005 shed light into the possible etiopathogenesis of COSD related to a trigeminal small-fiber neuropathy, characterized by a significant loss of epithelial and subpapillary nerve fibers. [33]

Considering the large number of articles published on COSD in the last 20 years that have discussed many aspects in COSD pathogenesis and management, these five papers have represented the knowledge carriers recognized by the scientific community, and constituted a starting point of new research to produce new knowledge in the field of COSD.

It is therefore not surprising to acknowledge "Grushka M" in cluster 1 being the most co-cited author. This author can be considered the most productive author if we take into account the

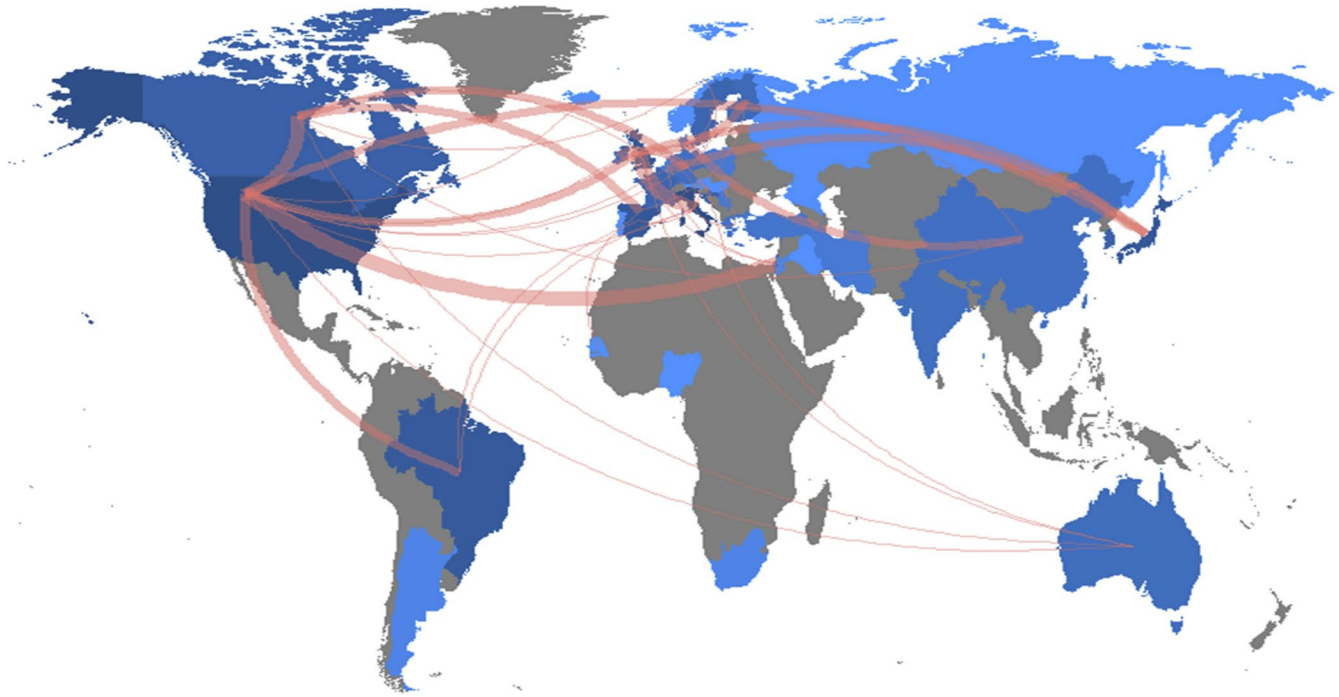


FIGURE 6 Worldwide collaborations among countries with at least 1 collaboration. List of the top 20 collaborating countries (times of collaboration): USA-Israel: 5; United Kingdom-Italy: 4; United Kingdom-Ireland and United States-Canada: 3; Denmark-China, France-Canada, Germany-Canada, Japan-Belgium, Japan-United Kingdom, Japan-USA, United Kingdom-Finland, United States-Brazil, and United States-United Kingdom: 2; Belgium-Israel, Canada-Australia, Canada-China, Canada-Norway, China-Sweden, Croatia-Australia, and Croatia-Austria: 1

entire period of 30 years, having published the first article in 1986 and the last in 2016 based on our collection. The author's three articles published in 1987 have represented a milestone in the field of COSD, being the three out of the top six most global and cited articles.

Lastly, only very few journals identified through the co-citation analyses occupied a prominent position in substantially contributing to the intellectual, conceptual, and methodological basis for COSD and in generating the most relevant findings in this field.

Our analysis highlighted that several research groups have contributed to the expansion of knowledge on COSD; however, these groups were isolated and performed research mainly within their group. These data were also supported by the analysis of the most predominant institutions which showed collaboration mainly within their own country. The relationship seen among different countries is explained by the fact that some authors within the same group of research might have different affiliations.

The lack of international collaboration could represent a true obstacle into future progress in better understanding the etiopathogenesis and management of COSD. It has been proposed that researchers from different areas of interest and different perspectives and background will work synergistically to improve this situation with constant support from academic journals, professional associations, and scientific meetings.[42] Hopefully, this will promote the intellectual exchange and new consensus on research terminology, and theoretical and methodological foundations.[16]

Our research on the structure of knowledge showed two main limitations: (a) Our analysis was based on KeyWords Plus, and as such, it did not analyze contents via abstracts or titles of the articles in our collection; and (b) it was not possible to evaluate the incidence of self-citations.

More generally, the co-citation analysis presents limitations due to its nature. It represents a quantitative measure of the research, but does not provide information about its quality, although we may hypothesize that the more citations an article receives, the greater impact that article may have on the scientific community. It is mainly a retrospective analysis; therefore, more recent articles are underrepresented, because they have had less time to be cited in comparison with older articles. Lastly, scientists may tend to cite more frequently from a group of research they have been in contact with or academic works coming from institutions with a highly respected reputation in the field.

5 | CONCLUSIONS

This study analyzes the knowledge structure of COSD over the past 30 years with a quantitative approach. Our results demonstrate that research on COSD is mainly driven by three different clusters that showed a moderate degree of interconnection, requiring a higher level of synergy to promote more translational research in this field. The mapping of thematic areas helped to identify main research interest and provided a clue into future research direction.

Unfortunately, the cooperation between different authors from different countries worldwide has been poor. In order for future advancement in this field of study, there is a need to change this stifling trend, and future global collaborative efforts should be encouraged, supported, and implemented.

AUTHOR CONTRIBUTION

Giulio Fortuna: Conceptualization; Data curation; Formal analysis; Investigation; Methodology; Writing-original draft; Writing-review & editing. **Massimo Aria:** Conceptualization; Data curation; Formal analysis; Methodology; Software; Supervision; Validation; Writing-original draft; Writing-review & editing. **Alfonso Piscitelli:** Data curation; Formal analysis. **Michele D. Mignogna**: CRediT contribution not specified. **Gary D. Klasser**: CRediT contribution not specified.

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