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Research Article

Evolution of Thoracic Outlet Syndrome Research: A Detailed Bibliometric Study

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Abstract

Aim: Thoracic outlet syndrome (TOS) is a complex clinical condition caused by the compression of neurovascular structures in the thoracic outlet. Over the past few decades, research on TOS has expanded significantly. This study aims to analyze the literature on TOS using bibliometric methods to identify critical trends, influential authors, prominent themes, and emerging topics in the field.
Material and Method: A comprehensive literature search was conducted using the Web of Science (WoS) database for articles on TOS published between 1980 and 2023, employing the keyword 'thoracic outlet syndrome.' The collected data were analyzed using bibliometric methods. VOSviewer software was utilized to visualize bibliometric networks and map critical findings. Citation analysis was performed to identify influential journals and significant articles in the field. Additionally, keyword clustering and trend analyses were conducted to explore the thematic landscape of TOS research.
Results: In a literature search on TOS, 2248 publications were found, with 1509 journal articles (67.13%) included in the bibliometric summary. The most common research area was surgery (633 articles, 41.94%), and the United States had the highest publication productivity (684 articles, 45.32%). The University of California System had the highest number of publications among institutions (62 articles, 4.1%). These 1509 articles received 7831 citations (6596 excluding self-citations), averaging 13.1 citations per article and with an h-index of 57. 'Annals of Vascular Surgery' published the most articles (71, 4.7%), while 'Journal of Vascular Surgery' had the highest number of cited articles (65, 4.3%), accumulating 2563 citations.
Conclusion: This bibliometric analysis provides valuable insights into the evolution and trends of research on TOS. The keyword analysis offers a roadmap for researchers to design new studies. Additionally, the study highlights the influence of economic size and development levels on academic productivity in TOS, underscoring the importance of promoting multidisciplinary studies, especially in less developed countries.

Keywords: Thoracic outlet syndrome, bibliometric analysis, citation analysis, Web of Science, VOSviewer

INTRODUCTION

Thoracic Outlet Syndrome (TOS) is characterized by conditions where bony, muscular, or fibrous structures compress the neurovascular bundle in the thoracic outlet, leading to various symptoms (1). Peet et al. introduced the term "thoracic outlet syndrome" in 1956, and it has since become well-established (2).

TOS is divided into three subcategories based on the compressed anatomical structure and clinical symptoms. These categories include neurogenic TOS, which involves brachial plexus compression; arterial TOS, characterized by subclavian artery compression; and venous TOS, involving subclavian vein compression (3). It is worth noting that neurogenic TOS is the most prevalent, accounting for approximately 90% of cases (4).

Compression in TOS typically occurs in the interscalene triangle, costoclavicular space, or coracopectoral tunnel (5,6). The interscalene triangle, a narrow area between the anterior and middle scalene muscles and the first rib, is particularly prone to compression due to fibrous and muscular abnormalities (6-8). Contributing factors include trauma, tumors, clavicle fractures, fibromuscular bands, and rib anomalies (9).

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The exact number of cases of TOS is uncertain, but it is estimated to range from 2.5 to 4.0 cases per 100,000 people annually in the United States of America (USA) (10,11). Diagnosing TOS is challenging because there are no definitive neurodiagnostic tests or objective criteria (4). While treating arterial and venous TOS is straightforward, treating neurogenic TOS remains controversial, with no consensus on surgical timing or approach (12-16).

TOS is addressed by various surgical specialties, such as thoracic, vascular, orthopedic, neurosurgery, and plastic surgery (17). Misdiagnosis and complications are frequent, potentially leading to severe outcomes (18). A multidisciplinary approach is often beneficial for patients (17).

Given the ongoing debates and complexities surrounding TOS, conducting a bibliometric analysis can offer valuable insights into research trends and developments in this field. Our study aims to provide a quantitative description of the current status and trends in TOS research, identify influential publications, journals, researchers, and countries, and offer a reliable overview of the state of TOS research to guide future investigations.

MATERIAL AND METHOD

Data Sources

The literature review utilized the Web of Science (WoS) database to gather relevant studies. WoS database is a comprehensive indexing resource provided by Clarivate Analytics in Philadelphia, Pennsylvania. This database includes the Science Citation Index Expanded (SCI-E), Social Sciences Citation Index (SSCI), Arts & Humanities Citation Index (A&HCI), Conference Proceedings Citation Index (CPCI), and other relevant sources. WoS is recognized globally as a multidisciplinary resource, providing access to numerous influential, high-quality academic journals. Leveraging WoS's built-in functions, we could delve into publication trends, geographical distribution, research areas, active organizations, journals, authors, trending topics, and citation counts (19). These robust features make the WoS database an ideal resource for conducting comprehensive bibliometric analyses of research related to TOS. Ethical approval was not required as the search utilized publicly available databases. Data collection was completed on May 17, 2024, ensuring the inclusion of all relevant publications up to that date.

Search Strategy

The search strategy was carefully crafted to explore the "Thoracic Outlet Syndrome". Only original articles were included in the search, while other document types such as reviews, meeting abstracts, proceedings papers, editorial materials, and letters were deliberately excluded. The search period was defined to span from 1980 to 2023 to capture a comprehensive range of literature. Raw data from the WoS database was meticulously extracted in plain text format to facilitate in-depth bibliometric analysis.

Data Extraction and Collection

We carefully retrieved relevant data from the WoS, manually excluding unrelated literature. Two authors independently browsed and extracted data from the selected articles. The information obtained from the articles included titles, authors, affiliations, publication year, contributing nations, journals, keywords, references, citation frequency, average citations per item, and h-index. We used Microsoft Office Excel 2017 for data entry and management. We conducted a thorough and meticulous process to gather relevant data from the WoS, carefully filtering out any unrelated literature by hand.

Bibliometric and Visualized Analysis

The world map was generated using GunnMap2 (http://lert.co.nz/map/), and bibliometric network and density visualizations were created with VOSviewer software (version 1.6.13, Van Eck and Waltman, Leiden University, The Netherlands) (20). In the network visualization map, larger circles indicated more significant contributions, and thick, closely positioned lines signified strong relationships. The density visualization map used a color scale from blue to red, with red representing areas with more items and greater weight of neighboring items.

Statistical Analysis

Statistical analyses were conducted using SPSS (Version 21.0, IBM Corp., Armonk, NY). The Kolmogorov-Smirnov test assessed data distribution normality, while Spearman's correlation analyzed the relationship between publications per country and GDP per capita (United et al., 2022) and HDI (United Nations, 2022). Linear regression was applied to predict future publication trends (2024-2034), with statistical significance set at $p<0.05$.

RESULTS

Quantity of Global Publications

The literature search resulted in 2248 publications. Of these, 1509 (67.13%) were journal articles, 227 (10.10%) were review articles, 205 (9.12%) were meeting abstracts, 135 (6.01%) were editorial materials, 113 (5.03%) were letters, 29 (1.29%) were proceedings papers, 23 (1.02%) were notes, 5 (0.21%) were corrections, 1 (0.04%) was early access, and 1 (0.04%) was a reprint. In total, 1509 articles were analyzed bibliometrically in this study. The most common languages of publication were English (n=1365; 91%), German (n=55), French (n=53), Spanish (n=13), Turkish (n=8), Korean (n=6), Japanese (n=3), Portuguese (n=3), Polish (n=2), and Czech (n=1). These 1509 articles received 7831 citations (6596 excluding self-citations), with a mean of 13.1 citations per article. The total h-index for all included journal articles was 57.

Active Research Areas

The research landscape is diverse, with surgery being the most common focus area, comprising 41.94% of published articles. Following closely behind are peripheral vascular disease (20.41%), orthopedics (13.18%), and clinical neurology (11.99%).

The research articles span a wide range of fields, with general internal medicine (165), radiology, nuclear medicine, and medical imaging (115), sports sciences (92), and rehabilitation (91) being the most prominent. Additionally, significant contributions were made in the areas of cardiac and cardiovascular systems (88), neurosciences (72), and the respiratory system (57). The research also extended to pediatrics (40), anatomy and morphology (37), anesthesiology (27), and rheumatology (22), among others. It is important to note that some articles were labeled in more than one field, reflecting the interdisciplinary nature of research.

Annual Publication Production

The graphical representation in Figure 1 illustrates the distribution of the number of articles by year. Notably, there was a substantial surge in the number of articles focusing on TOS in 2021 compared to previous years. Additionally, Figure 1 visually represents the projected number of publications for the next ten years using regression analysis, accompanied by 95% confidence intervals (CI). The regression analysis forecasts that 70 journal articles [95% CI: 51-89] are anticipated to be published in 2024, increasing to 84 [95% CI: 65-103] by 2033.

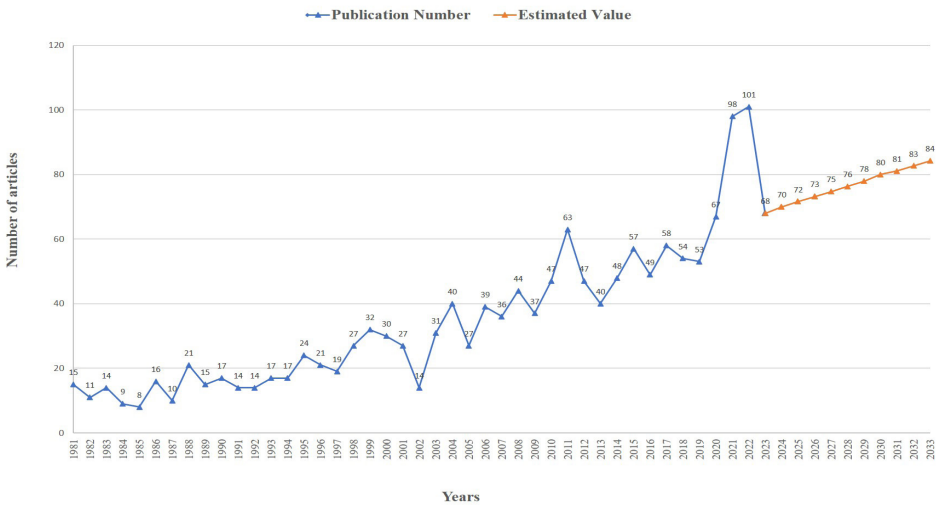

Figure 1. Trends in the number of annual publications by year

Active Countries

The productivity of countries in terms of published articles is as follows: The USA leads with 684 publications (45.32%), followed by France with 117, England with 73, Germany with 68, Türkiye with 67, Italy with 56, India with 48, Japan with 44, Canada with 37, Netherlands with 36, South Korea with 34, China with 31, Switzerland with 26, Belgium with 22, Finland with 21, Australia with 20, Spain with 19, Austria with 18, Brazil with 16, Sweden with 14, Poland with 11, and other countries contributing 152 publications (Figure 2). The corresponding authors indicate that the USA is the most productive country in the field related to TOS, as shown in Figure 3, with most publications originating from the USA.

Thirty-three countries contributed articles to the published articles. Figure 4 illustrates the map displaying the international collaboration networks among countries that have jointly published at least five articles.

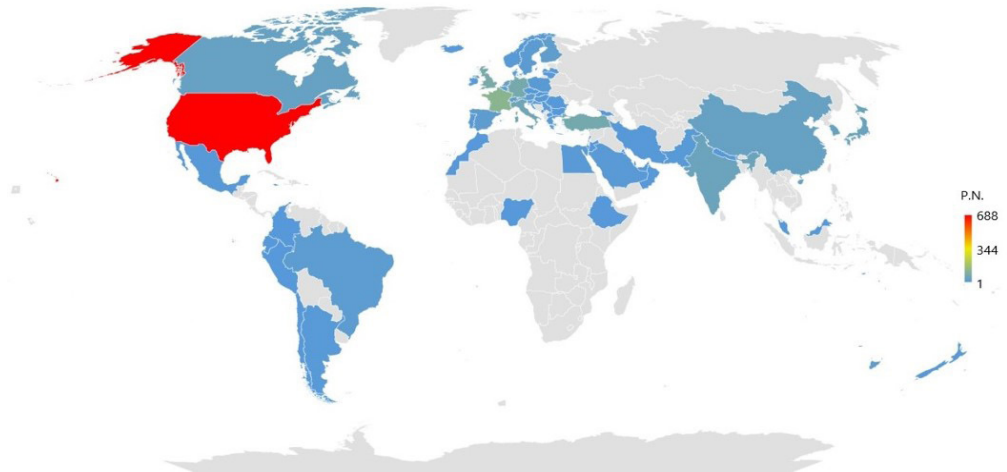

Figure 2. World map depicting publication productivity in the field of TOS by country (P.N.: Publication Number); Productivity is color-coded on a scale from blue (low) to orange (high)

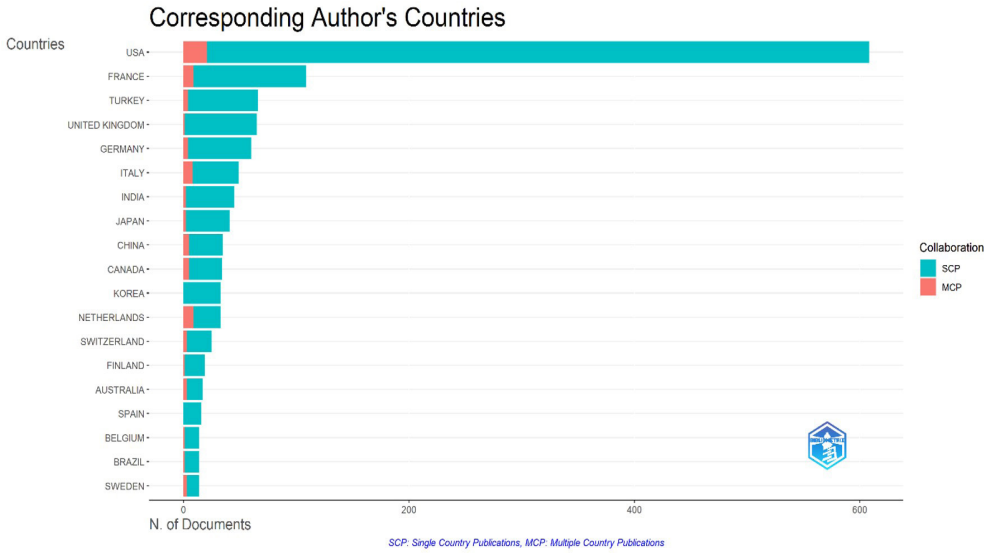

Figure 3. The most productive countries in the TOS-related field (MCP: multiple corresponding author publication; SCP: single corresponding author publication)

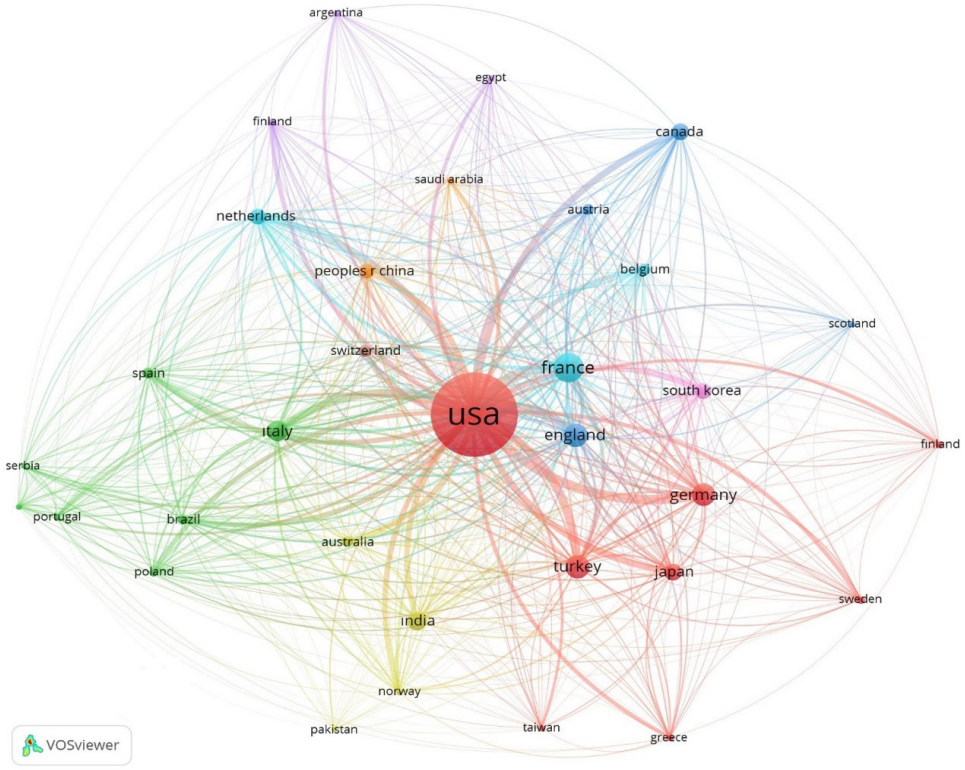

Figure 4. Network visualization map for international collaborations on TOS. (The sizes of the circles represent the number of publications, the colors indicate clusters of collaborations, and the thickness of the lines indicates the strength of collaborations)

Factors Associated with the Numbers of Publications

The analysis revealed a statistically significant correlation ($p<0.01$) between the number of publications on TOS and both GDP per capita ($r=0.36$) and HDI ($r=0.43$).

Active Journals

A comprehensive analysis revealed that 1509 articles were published across 558 distinct journals. Specific journals stood out for their exceptional frequency of publications and citations on TOS. These standout journals, which had each published a minimum of 10 articles, are detailed in Table 1. Annals of Vascular Surgery boasted the highest number of articles (71 articles, 4.7%), while the Journal of Vascular Surgery garnered the most citations (65 articles, 2,563 citations). The table also provides insights into the number of publications and citations per article. Furthermore, Figure 5 showcases a visually compelling citation network visualization map highlighting the most active journals.

Table 1. Journals with the highest frequency of publications and citations on TOS

Journal	Publications (n)	Citations
Annals of Vascular Surgery	71	1078
Journal of Vascular Surgery	65	2563
Journal of Vascular Surgery: Venous and Lymphatic Disorders	16	132
Annals of Thoracic Surgery	15	384
Vascular and Endovascular Surgery	19	191
Diagnostics	13	114
Thoracic Surgery Clinics	11	53
Muscle & Nerve	21	720
Hand Clinics	24	501
European Journal of Vascular and Endovascular Surgery	10	174
Journal of Hand Surgery - American Volume	16	294
American Journal of Surgery	12	411
Vascular	13	115
American Surgeon	10	173
Clinical Orthopaedics and Related Research	12	165
Vasa - Journal of Vascular Diseases	14	61
Surgical and Radiologic Anatomy	10	73
Archives of Physical Medicine and Rehabilitation	11	189
Journal des Maladies Vasculaires	10	63
Cureus Journal of Medical Science	12	44

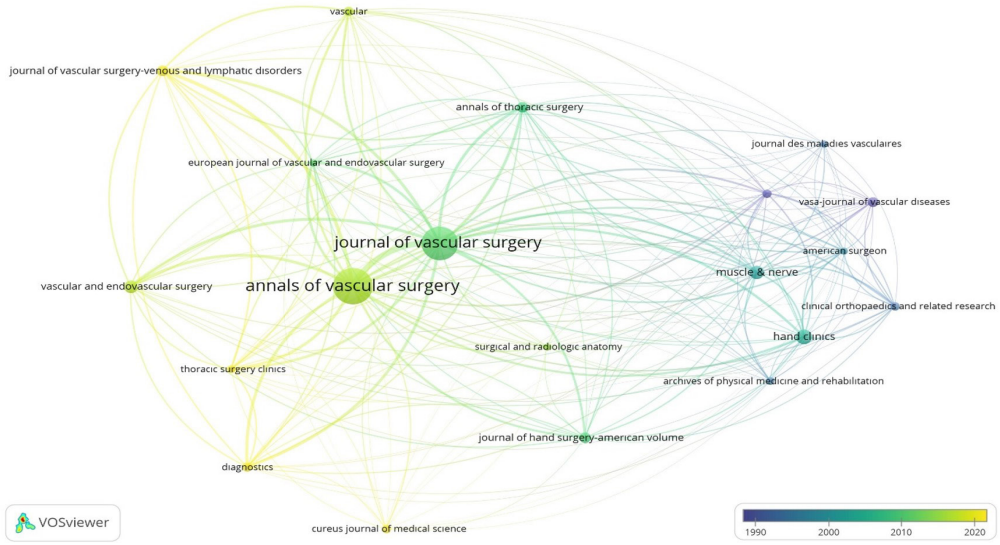

Figure 5. A network visualization map illustrating the citation analysis of the most active journals in the field of TOS. (The size of the circles indicates the frequency of each journal, while the thickness of the lines reflects the strength of the relationships)

Active Authors and Organizations

The following individuals were the most active authors in the field: Thompson RW (30 publications), Freischlag JA (29 publications), Gelabert HA (16 publications), Ozcakar L (15 publications), Teijink JAW (14 publications), Sanders R (14 publications), Abraham P (14 publications), Lum YW (12 publications), Henni S (12 publications), Pesser N (11 publications), Illig KA (11 publications), Hersant J (11 publications), and Donahue DM (11 publications). In addition, Figure 6 highlights the most active organizations in TOS research. The University of California System led 62 publications, followed by Harvard University and Johns Hopkins University with 53 publications.

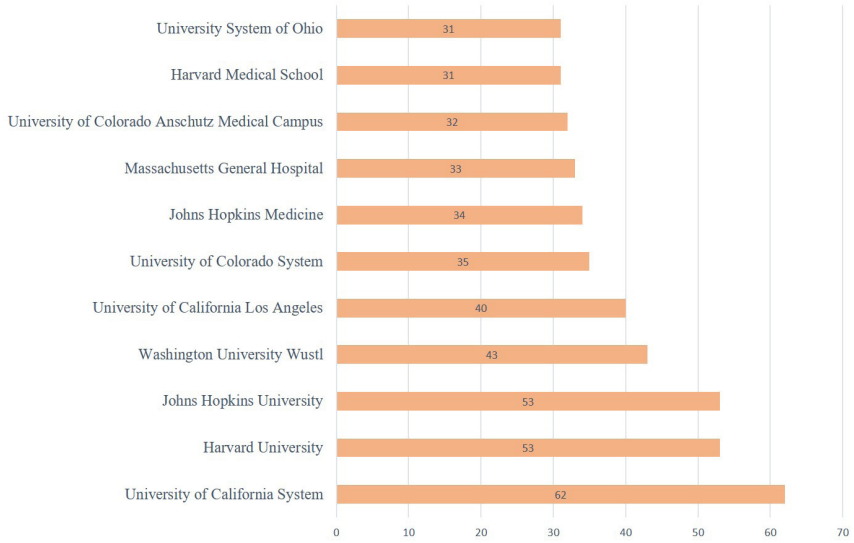

Figure 6. The most productive organizations are in the TOS-related field

Citation Analysis

Table 2 provides a comprehensive overview of the ten most extensively cited articles, including their total citation counts and average citations per year. Notably, all of these articles have amassed more than 100 citations each. The article by Sanders RJ et al. stands out with the highest number of citations, totaling 252, and was published in the Journal of Vascular Surgery in 2007 (21).

Table 2. The 10 most cited manuscripts on TOS

No	Article	Author (s)	Journal	PY	AC	TC
1	Diagnosis of thoracic outlet syndrome	Sanders RJ, Hammond SL and Rao NM	Journal of Vascular Surgery	2007	14	252
2	Ectopic sensory discharges and paresthesia in patients with disorders of peripheral-nerves, dorsal roots and dorsal columns	Nordin M, Nystrom B, Wallin U and Hagbarth KE	Pain	1984	5.41	222
3	Surveillance case definitions for work related upper limb pain syndromes	Harrington JM, Carter JT, Birrell L and Gompertz D	Occupational and Environmental Medicine	1998	7.22	195
4	Guidance for the treatment of deep vein thrombosis and pulmonary embolism	Streiff MB and et al.	Journal of Thrombosis and Thrombolysis	2016	21.56	194
5	Nonunion of the clavicle - associated complications and surgical- management	Jupiter JB and Leffert RD	Journal of Bone and Joint Surgery-American Volume	1987	4.68	178
6	Outcomes of surgery in 1019 brachial plexus lesions treated at Louisiana State University Health Sciences Center	Kim DH, Cho YJ, Tiel RL and Kline DG	Journal of Neurosurgery	2003	7.36	162
7	Reporting standards of the Society for Vascular Surgery for thoracic outlet syndrome	Illig KA and et al.	Journal of Vascular Surgery	2016	15.78	142
8	Imaging assessment of thoracic outlet syndrome	Demondion, X and et al.	Radiographics	2006	6.74	128
9	The place for scalenectomy and 1st-rib resection in thoracic outlet syndrome	Roos DB	Surgery	1982	2.93	126
10	The treatment of thoracic outlet syndrome - a comparison of different operations	Sanders RJ and Pearce WH	Surgery	1989	3.47	125

PY: publication year, AC: average citations per year, TC: total citation

Keyword Analysis

In scholarly publications, keywords are crucial as they provide essential information about the main themes and ongoing research trends. The frequency of specific keywords used during a particular period can offer valuable insights into the primary focus areas and critical research trends related to TOS. Figure 7A provides a detailed breakdown of the most frequently used keywords in TOS-related literature. 'Thoracic outlet syndrome' is the most commonly used keyword, appearing 250 times, followed by 'management' (216), 'diagnosis' (203), 'surgery' (120), and 'compression' (112).

Furthermore, analyzing the co-occurrence of keywords provides a comprehensive understanding of the research hotspots within the field. This analysis reveals the frequency with which a group of keywords appears in the same publications, forming a co-occurrence network that highlights their connections. To conduct this analysis on publications spanning from 1980 to 2023, we utilized VOSviewer. Figure 7B illustrates the change in the frequency of keywords over time, with "thoracic outlet syndrome" remaining the most prominent keyword, appearing 250 times. Figures 7C and 7D present detailed visual representations of the co-occurrence network of keywords, illustrating the relationships between frequently used terms in the analyzed literature. Each node in Figure 7C represents a keyword, with the node's size corresponding to the frequency of co-occurrence. Keywords of the same color belong to the same cluster. Notably, in Figure 7C, we have categorized all the keywords into twelve clusters, with the largest cluster containing 20 keywords, including "brachial plexus". This categorization provides a detailed insight into the interconnectedness of the keywords within the TOS-related literature.

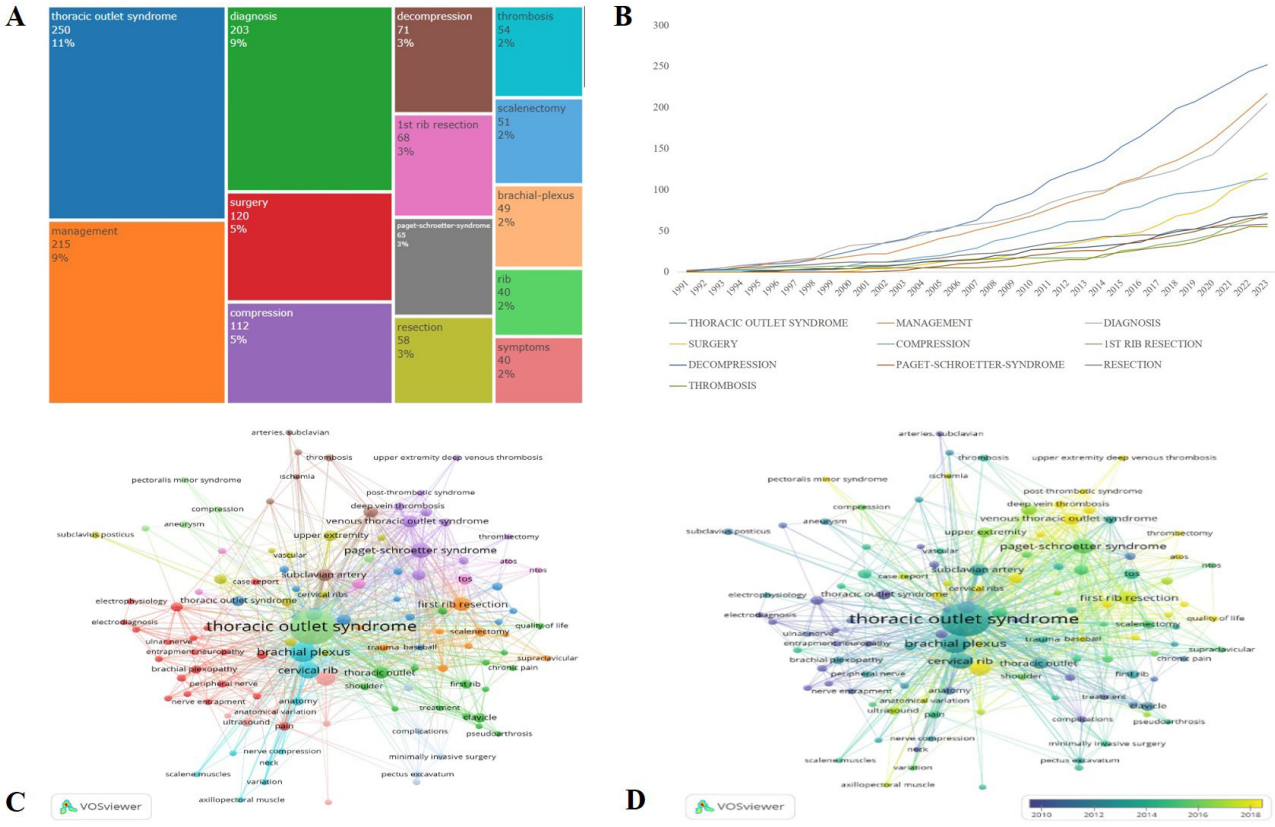

Figure 7. Keyword analysis of the TOS-related publications; **A.** Tree Map of the top 117 high-frequency keywords used, **B.** Keyword frequency over time, **C.** Co-occurring network of keywords, **D.** Co-occurring network of keywords in overlay visualization

DISCUSSION

This comprehensive study presents a detailed overview of the current status and global trends in TOS research. Over the years, there has been a consistent rise in the annual number of publications in this field, indicating a growing interest and focus on TOS. Our analysis has identified vital researchers, institutions, and countries involved in TOS research and their collaborative networks, shedding light on influential and highly cited publications. We have provided valuable insights into the landscape of TOS research through extensive bibliometric analyses, including keyword analysis and citation analysis of articles and journals, as well as an examination of international collaborations. This study is the first to conduct in-depth analyses, covering many articles on TOS and offering significant contributions to the knowledge base for researchers and practitioners in this field.

Growth of TOS Research

Research on TOS has grown significantly over the past four decades. The number of publications on TOS surged after

2021, which could be attributed to increased awareness, advancements in diagnostic techniques, and the findings of other studies (e.g., Illig et al., 2021). This increase in research activities may also signify the growing recognition of TOS as a significant clinical issue that demands more focused investigation and intervention.

Academic Influence and Key Contributors

The USA has a global academic influence, which is evident in its many publications and citations. These statistics support previous research highlighting the USA's prominent role in medical research, attributed to its substantial investment in healthcare and research infrastructure (Martín-Martín et al., 2021). The University of California System is the most active academic institution, fostering collaborative relationships with research organizations worldwide. This institution's prominence can be attributed to its extensive network of medical centers and its dedicated focus on multidisciplinary research.

Impact of Economic and Developmental Factors

Previous research has demonstrated a strong connection between a nation's economic strength, development indices, and publication productivity (22,23). Our study further substantiates this link by identifying specific correlations between certain economic and developmental indicators. It is evident that countries with higher wealth and more advanced healthcare systems are better positioned to allocate resources for research and produce high-impact studies, as evidenced by the research of Peet et al. in 1956 and Hooper et al. in 2010. These findings emphasize the critical role of resource allocation in driving scientific advancements and the importance of economic and developmental factors in shaping publication productivity.

Leading Journals in TOS Research

Two prominent journals greatly support the field of TOS research. The Annals of Vascular Surgery stands out with the most publications in TOS-related research and is the second most cited journal in this specialized area. On the other hand, the Journal of Vascular Surgery takes the lead regarding citations and holds the second position in the number of publications. These journals are renowned for their extensive readership and primary focus on vascular conditions, which naturally encompasses TOS. This underscores the multidisciplinary approach to TOS management, with a particular emphasis on the role of vascular surgeons. Researchers interested in TOS are strongly encouraged to prioritize these journals to stay updated on the latest developments and findings in the field.

Citation Trends and Influential Articles

Most of the top 10 most cited articles were published before 2010, suggesting that older studies have more time to accumulate citations than recent publications. These earlier works mainly focused on the diagnosis and treatment of TOS. The most frequently cited report, written by Sanders RJ in 2007, explores the diagnostic process for TOS, emphasizing the lasting impact of foundational studies on current understanding and practices.

Keyword Analysis and Research Hotspots

Keywords play a critical role in scholarly publications by offering valuable insights into the central themes and prevailing research trends in TOS. In TOS-related literature, some of the most frequently used keywords include "thoracic outlet syndrome", "management", "diagnosis", "surgery", and "compression". These keywords reveal interconnected research hotspots through co-occurrence analysis, forming networks that illustrate their associations. An analysis using VOSviewer to study publications from 1980 to 2023 highlighted "thoracic outlet syndrome" as the most prominent keyword. Additionally, the analysis showcased the grouping of keywords into clusters, with the largest cluster encompassing significant terms such as "brachial plexus". These clusters indicate concentrated research endeavors and evolving interests within the TOS field.

Limitations

While a bibliometric analysis can provide valuable insights into the publication data within a specific scientific field, it is essential to address the limitations of our study:

- Our selection was limited to the WoSCC database, meaning that other significant databases, such as PubMed and Scopus, were not included in our analysis.
- We only focused on English-language publications, potentially overlooking relevant studies published in other languages. It is crucial to note that the frequency of citations does not necessarily indicate the quality of a publication, which means that some high-quality studies with fewer citations might have been excluded from our analysis.
- Our study only included publications with sufficient data, which might have excluded more recent studies.

It is essential to recognize that research findings are subject to change over time. Lastly, our conclusions are based on database records, which may only partially represent the real-world scenario. This limitation is standard in all bibliometric studies.

Strengths and Innovations

Our research has significant strengths, notably its comprehensive coverage of the topic and the utilization of advanced bibliometric methods. No other study has conducted such extensive analyses within the field of TOS research, making our work a valuable and unprecedented resource for guiding future investigations in this area.

Future Research Directions

Future research could expand upon these findings by incorporating additional databases, such as PubMed and Scopus, offering a more comprehensive understanding of the subject. Including non-English publications may also reveal significant studies that should have been included

in this analysis. Additionally, longitudinal studies could be valuable in tracking changing trends in TOS research and its impact on clinical practices.

CONCLUSION

In this study, we used bibliometric and visualized analyses to identify current trends in research and collaborative relationships among countries, authors, and institutions. We also reviewed key scientific works to provide researchers with a comprehensive overview of the academic landscape in TOS research. Over the past forty years, there has been significant growth in TOS research, with the USA emerging as the leading contributor. The University of California System has been particularly active in this field. The Annals of Vascular Surgery and the Journal of Vascular Surgery have been identified as the most influential journals in this study area.

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Ethical approval: *As this study did not examine human subjects, it was exempt from institutional review board approval.*

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