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Bibliometric and visual analysis of research on nutcracker syndrome from 1974 to 2021

A systematic review

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Abstract

Background: At present, researchers have obtained fruitful results in the study of nutcracker syndrome (NCS), but there is still a lack of systematic research on the overall status of this disease. This article aims to describe the past and current status of research into NCS, and predict future research trends and popular research topics.

Methods: Using bibliometric and visualization methods, 552 articles related to NCS collected from the Scopus database from 1974 to 2021 were analyzed from multiple perspectives.

Results: Overall, the amount of literature related to NCS is on the rise every year, and the number of citations is the turning point in 2006. The United States has the largest number of publications and has the most extensive cooperation with other countries. The main contents of the co-authored study focused on the symptoms, surgical procedures, and concomitant diseases of NCS. Keywords such as peak velocity, ultrasonography, orthostatic proteinuria, etc appeared earlier, whereas diagnosis, chronic pelvic pain, endovascular stents, etc appeared later.

Conclusions: The literature utilization rate of NCS is relatively insufficient. The pathogenesis and pathological mechanisms need to be further studied, and the diagnostic criteria and surgical methods will continue to be favored by clinicians.

Abbreviations: LRV = left renal vein, NCS = nutcracker syndrome, SMA = superior mesenteric artery.

Keywords: analysis, bibliometrics, nutcracker phenomenon, nutcracker syndrome, visualization

1. Introduction

Nutcracker syndrome (NCS) is a condition that usually affects the left renal vein (LRV) between the aorta and superior mesenteric artery (SMA) or between the aorta and spinal column. NCS is accompanied by hematuria,[1,2] abdominal pain on the left side,[3] proteinuria,[4,5] gonadal varices,[6,7] chronic fatigue syndrome,[8,9] and other signs and symptoms of rare diseases.[10] The position between the aorta and the SMA is termed "anterior NCS",[11] whereas if found between the aorta and the spinal column is termed "posterior NCS."[12] In the early literature, the terms "nutcracker phenomenon" and "NCS" are often mixed,[13] although "nutcracker phenomenon" is only used to describe anatomical features.[14] In 1937, anatomist Grant[15] first described the associated anatomical structure as "the LRV as it lies between the aorta and the superior mesenteric artery resembles a nut between the jaws of a nutcracker." Thirteen years later, El Sadr and Mina published the first clinical report on NCS,[16] whereas the term "nutcracker phenomenon" was first coined in 1972 by de Schepper.[17] In the intervening 49 years, extensive research has been conducted on the NCS. Compression of the LRV by adjacent blood vessels,[18–20] tissues,[21] and organs[22] is considered the main cause of NCS. Different researchers have put forward different views on the pathological mechanisms of NCS: Lopatkin et al[23] proposed that hematuria is caused by microscopic ruptures of parenchymal veins due to an increasing pressure in the LRV that result in blood flow into the renal collecting system or calyceal dome; Buschi et al[24] believed that hematuria is caused by the formation of a fistula between a dilated venous sinus and adjacent calyces; and Ozçakar et al summarized possible mechanisms of NCS that may lead to proteinuria.[25] At present, the diagnosis of NCS is based on exclusion criteria, and there is still a lack of unified diagnostic criteria. Radiological examination is of great significance in the diagnosis of NCS. Diagnostic techniques include renal venography,[26] computed tomography,[27] magnetic resonance angiography,[28] renal Doppler ultrasound,[29] and multislice helical computed tomography angiography.[30] Of these techniques, renal venography is considered the gold standard for

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The dataset generated for this study will be provided by the author to any qualified researcher.

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diagnosing NCS.[31] Conservative and surgical treatments are the 2 main approaches used for the treatment of NCS. The former is mostly employed in young patients and those with mild symptoms,[32] whereas the latter is mostly employed in elderly patients[33,34] and those with obvious symptoms and complex conditions (**Table 1**). **Figure 1** shows the evolution of surgical treatment for NCS over time. As mentioned above, although the research on NCS has achieved a wide range of results, there is still a lack of systematic research on the overall status of this disease. Therefore, bibliometrics and visualization methods were used to analyze 552 articles related to NCS retrieved from the Scopus database and dating from 1974 to 2021 in terms of literature type, publication, citation, journal, article, country, author, and author keywords. Relationships in terms of international cooperation, co-authorship, co-citation, and co-occurrence of author keywords were presented in the form of charts and visual maps. We describe the past and current status of research in NCS and predict future research trends and popular research topics in order to provide a valuable reference for clinicians engaged in NCS-related research.

2. Method

2.1. Source database

We conducted a literature search using Scopus (https://www.scopus.com/home.uri). The Scopus database is the largest database of abstracts and citations of peer-reviewed literature (scientific journals, books, and conference records).[35] As the only source database for this research, it contains various types of literature required for this research.

2.2. Search design

We plan to search for all types of documents related to NCS by title, abstract, and keywords. To obtain results that met our requirements as much as possible, after fully reading the literature, we formulated the search string as (TITLE-ABS-KEY [“nutcracker syndrome”] OR TITLE-ABS-KEY [“posterior nutcracker syndrome”] OR TITLE- ABS-KEY [“nutcracker phenomenon”] OR TITLE-ABS-KEY [“left renal vein compression syndrome”] OR TITLE-ABS-KEY [“left renal venous hypertension”] OR TITLE-ABS-KEY [“left renal vein obstruction”] OR TITLE-ABS-KEY [“The syndrome of the incarcerated left renal vein”] OR TITLE-ABS-KEY [“aortomesenteric left renal vein compression”] OR TITLE-ABS-KEY [“left renal vein entrapment syndrome”]) AND (LIMIT-TO [LANGUAGE, “English”]).

2.3. Data collection

All results are obtained in the Scopus database using the above search formula and export as much relevant information as possible in the comma-separated values format. The search was carried out and completed on September 10, 2021, to avoid data discrepancies caused by database updates.

2.4. Data screening

First, we limited the language type to English in the search style to derive documents published in English; then, we browsed the title, abstract, keywords, and full text of each document to determine whether the document is obviously related to NCS to screen. In the derived 688 results, after excluding duplicates, small relevance, and irrelevant documents, a total of 552 results were determined to be valid (**Fig. 2A**). As shown in **Figure 2B**, among the 552 results, the total number and proportion of articles ranked first (409 and 74.09%), followed by letters (64, accounting for 11.59%) and reviews (39, accounting for 7.07%). The number and proportion of erratum and short surveys were the smallest, at both 5% and 0.91%. The number and proportion of the remaining literature types were as follows: Note, 15 (2.72%); Conference Paper, 9 (1.63%); Book Chapter, 6 (1.09%).

Table 1

Comorbidities of nutcracker syndrome.

Complications	Year	Authors
Renal vein thrombosis	1995	Hiekata T, et al
IgA nephropathy		Ozono Y, et al
Renin-dependent hypertension	2003	Hosotani Y, et al
Pelvic congestion syndrome	2004	Maes M, et al
Intussusception	2005	Shin JI, et al
Allergic purpura		
Idiopathic hypercalciuria and urolithiasis	2006	
IgA nephropathy		
HSP nephritis/post–streptococcal glomerulonephritis	2007	
Superior mesenteric artery syndrome	2008	Barsoum MK, et al
Right posterior vena cava ureter		Rao J, et al
Familial Mediterranean fever	2009	Ozcan A, et al
Familial Mediterranean fever	2010	Deger SM, et al
Urolithiasis		Şemsa Altugan F, et al
Congenital abnormality of the inferior vena cava	2012	Luo XL, et al
Secondary hypertension		Mazarakis A, et al
Arteriovenous malformations of the right kidney		Qin J, et al
Renal vein duplication malformation		Preza Fernandes J
Acute aortic dissection	2013	Kodama K, et al
Glomerulonephritis		Ma Z, et al
Superior mesenteric artery compression syndrome		Vulliamy P, et al
Glomerulonephritis		Zhong J, et al
Superior mesenteric artery syndrome	2014	Alenezy A, et al
Right occlusion of the ureter–pelvic junction		Chen W, et al
IgA nephropathy		Imai N, et al
Pelvic congestion syndrome	2015	Atkinson TH, et al
Down syndrome		Koh ES, et al
Congenital portosystemic shunt		Lee SH, et al
Disabling pelvic congestion syndrome		Thaveau F, et al
Hypertension	2016	Deser SB, et al
Familial Mediterranean fever		Dogru A, et al
Nephrotic syndrome		Wang Y, et al
Pancreatic body cancer		Yadav P, et al
Sickle cell trait	2017	Ahmad A, et al
Inferior vena cava hypoplasia		Deşer SB, et al
Hypertension		Gokosmanoglu F, et al
Superior mesenteric artery syndrome		Michael PG, et al
Superior mesenteric artery syndrome		Oh MJ, et al
Superior mesenteric artery syndrome	2019	Al-Zoubi NA, et al
Thin basement membrane disease		Hirakawa Y, et al
Urolithiasis		Kanai H, et al
Arcuate ligament syndrome		Moreno-Márquez C, et al
Intestinal malrotation		Nishio Y, et al
Superior mesenteric artery syndrome		Shi Y, et al
Horseshoe kidney and superior mesenteric artery syndrome	2020	Diab S, et al
Superior mesenteric artery syndrome		Farina R, et al
Hypertension		Hadei SK, et al
Acute pulmonary thromboembolism		Ito T, et al
Double inferior vena cava		Waśniewska A, et al
Double inferior vena cava		Wu WW, et al
May–Thurner syndrome	2021	Aghdasi S, et al
Median arcuate ligament syndromeSuperior mesenteric artery syndrome		Farina R, et al
Superior mesenteric artery syndrome		Hurtado FB, et al
Marfan syndrome		Ichihara Y, et al
IgA nephropathy		Wang C, et al
Hypertension		Wang RF, et al

HSP = Henoch-Schönlein purpura, IgA = immunoglobulin A.

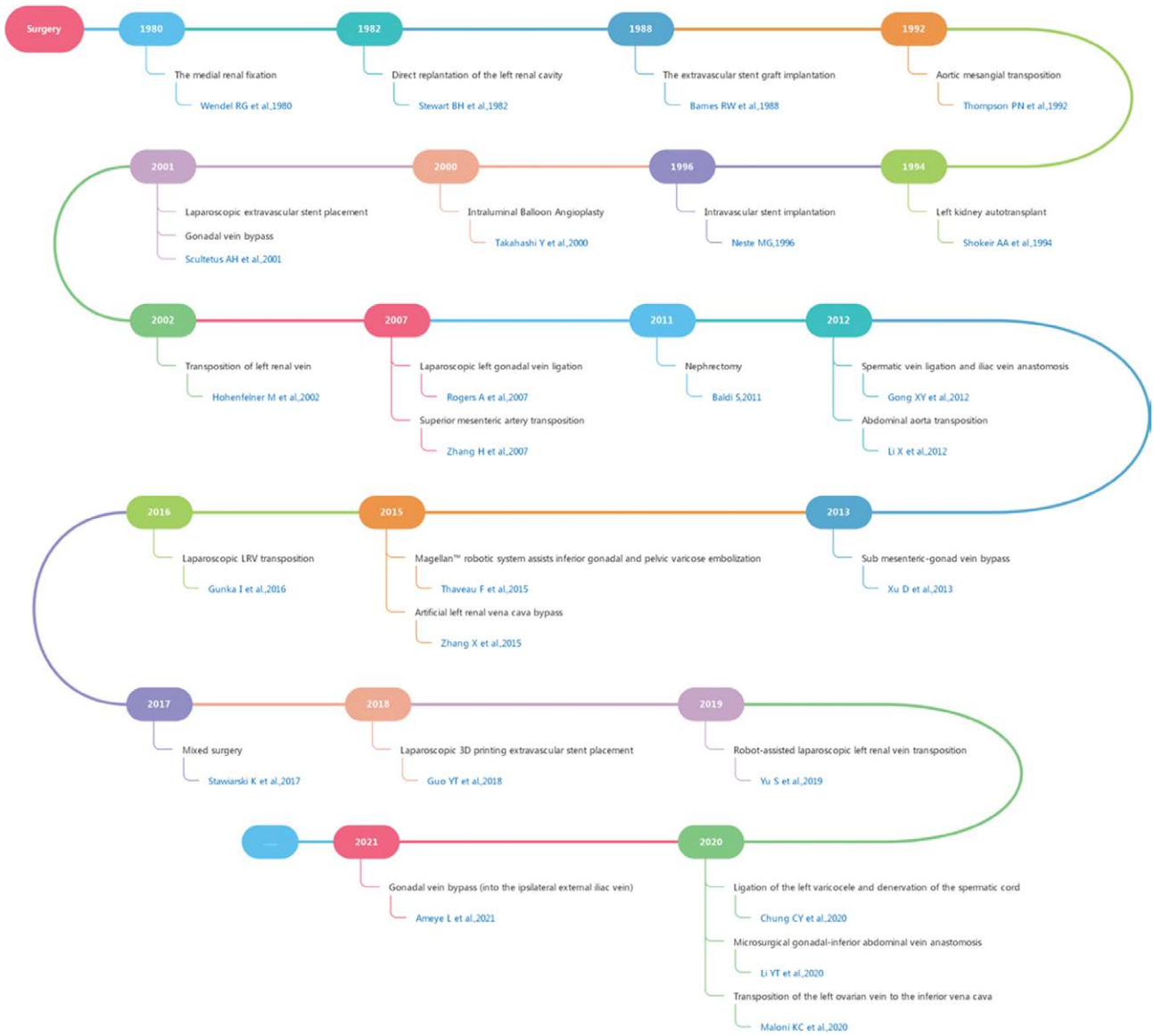

Figure 1. Development chart of operation mode with time. LRV = left renal vein.

2.5. Data analysis and visualization

GraphPad Prism Version 8.0.2 was used to analyze the type, quantity, and citation of the literature and to draw the corresponding pie chart and histogram. Microsoft Office Excel 2007 was used to draw a 3-line graph of the complications of NCS, the top 20 journals in total literature volume, and the top 15 in total citation volume. This study mainly uses MindMaster Version 8.1.0, and VOSviewer Version 1.6.17 for visual analysis. MindMaster is a cross-platform mind mapping software used to map the evolutionary process of NCS surgery. VOSviewer is a scientific knowledge mapping software tool that uses "network data" (mainly document knowledge units) for relationship construction and visual analysis to perform scientific knowledge mapping and display structure, evolution, cooperation, and other relationships. After importing the CVS file containing 552 publications, the author, country, citation, and author keywords were analyzed using VOSviewer. The above projects are linked through co-authorship, international cooperation, author co-citation, and co-occurrence of author keywords and are finally presented in the form of a visual map.

2.6. Research ethics

The research was conducted as a bibliometric analysis; all data sources were available on the Internet, and no animal or human subjects were involved. Therefore, permission was not required from the ethics committee.

3. Results

3.1. Publication and citation analysis

Figure 3A shows the number of papers published annually. It can be seen that from 1974 to 2021, although the number of published papers declined temporarily in some years, there was a gradual upward trend overall. The number of published papers increased slowly from 1974 to 2001, with no papers published in 7 of these years and <10 published in each of the remaining years. Although the number of papers published each year from 2002 to 2021 fluctuated, the growth rate was significantly higher than in the first phase. Excluding incomplete statistics for 2021, the number of papers published in 2020 was 40, and 2020 was ranked first among all the years

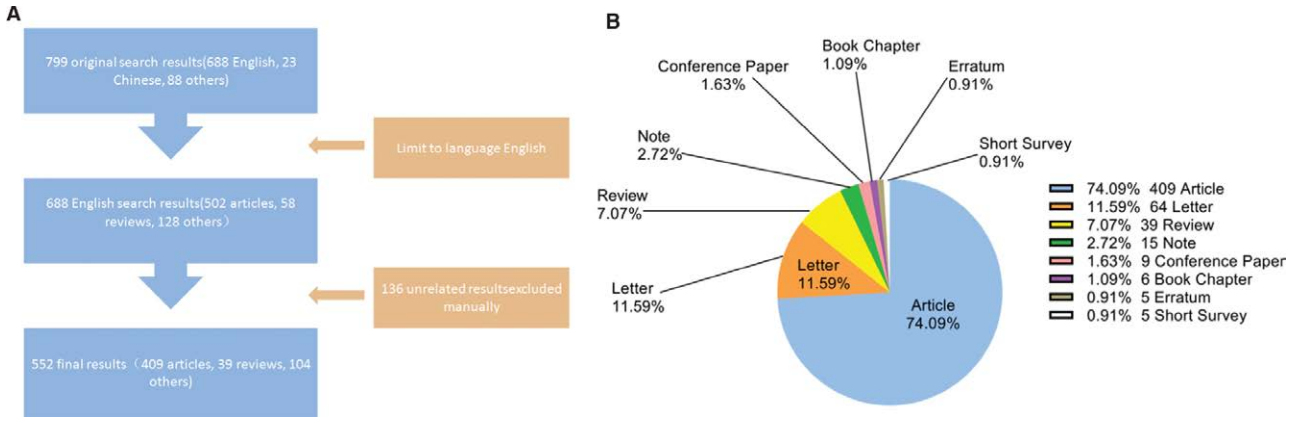

Figure 2. (A) Schematic diagram of literature screening. (B) Document type pie chart.

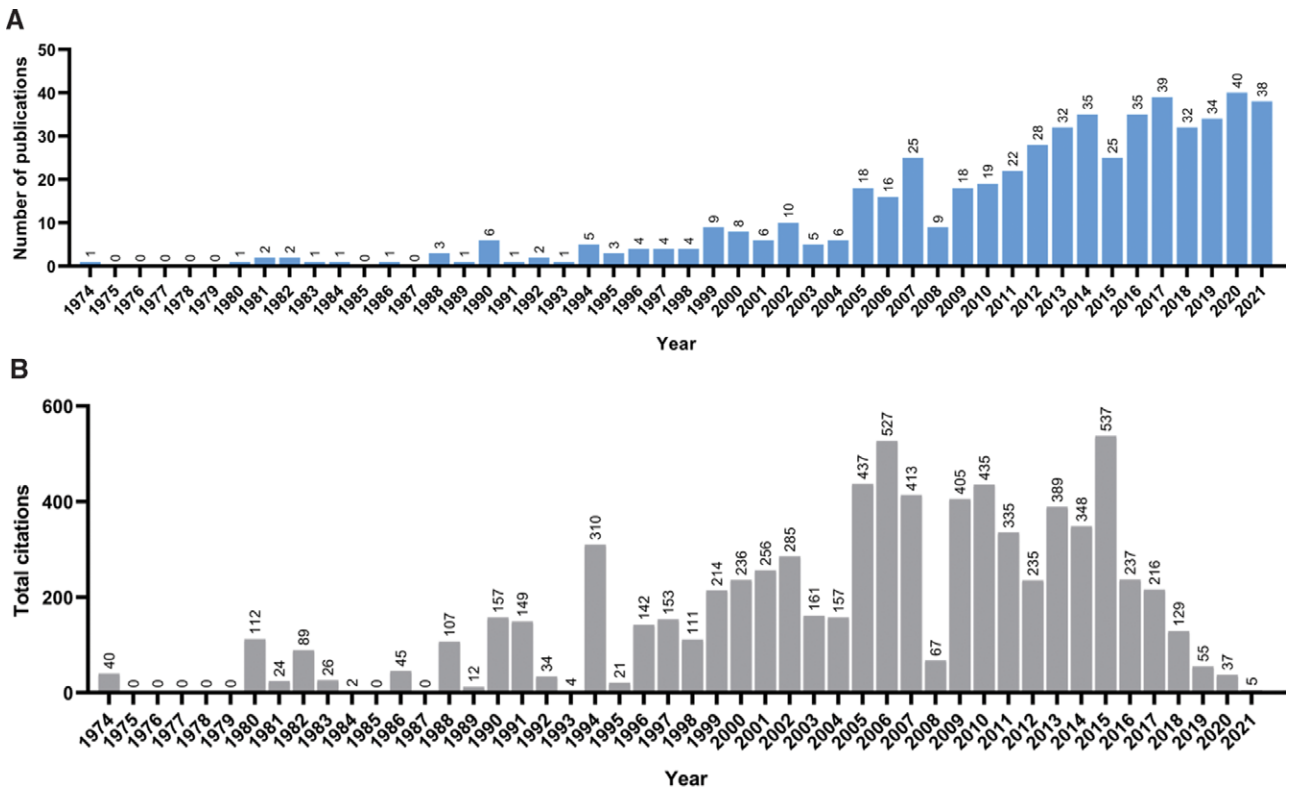

Figure 3. (A) Histogram of documents issued each year. (B) Histogram of total citations per year.

surveyed. The total number of citations of papers related to NCS per year is shown in **Figure 3B**. The period surveyed can clearly be divided into 2 phases: the first phase extends from 1974 to 2006, and the second phase extends from 2007 to 2021. Although there were fluctuations in both stages, the second stage contained the highest annual number of citations. Overall, 2006 marked a turning point from an upward trend to a downward trend. Comparing **Figure 3A** and **Figure 3B**, it is not difficult to see that the total number of citations of papers each year did not increase with an increase in the number of papers published each year. We speculate that this is due to the cumulative effect of citations of earlier documents over time. The citation range from 0 to >200 was equally divided into 21 intervals, and the number of papers contained in each interval is shown in **Figure 4**. The largest number of papers was cited 10

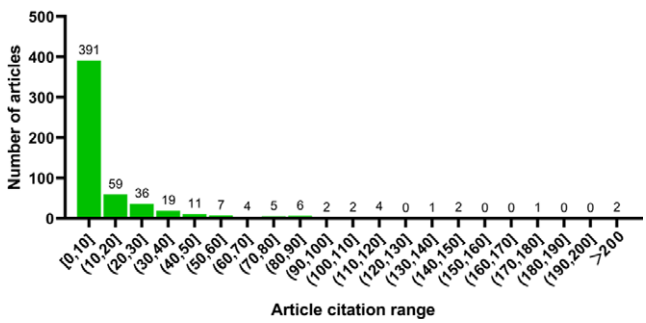

Figure 4. Histogram of literature number in different citation ranges.

Table 2

The top 15 articles cited in studies.

Rank*	Journal	Article title	Authors	CA	IF	Article type	Publication year
1	*European Journal of Vascular and Endovascular Surgery*	Editor's choice – Management of chronic venous disease: Clinical practice guidelines of the European Society for Vascular Surgery (ESVS)	Wittens C et al	412	7.069	Journal article	2015
2	*Mayo Clinic Proceedings*	Nutcracker phenomenon and nutcracker syndrome	Kurklinsky AK, Rooke TW	250	7.616	Journal article	2010
3	*European Journal of Vascular and Endovascular Surgery*	Current trends in the diagnosis and management of renal nutcracker syndrome: A review	Ahmed K et al	175	7.069	Journal article	2006
4	*Journal of Vascular Surgery*	The nutcracker syndrome: Its role in the pelvic venous disorders	Scultetus AH, et al	150	4.268	Journal article	2001
5	*Journal of Urology*	The nutcracker syndrome: New aspects of pathophysiology, diagnosis and treatment	Hohenfellner M, et al	149	7.450	Journal article	1991
6	*Radiology*	Nutcracker syndrome: Diagnosis with Doppler US	Kim SH, et al	139	11.105	Journal article	1996
7	*American Journal of Roentgenology*	Diagnosis of the nutcracker syndrome with color Doppler sonography: Correlation with flow patterns on retrograde left renal venography	Takebayashi S, et al	117	3.959	Journal article	1999
8	*Annals of Vascular Surgery*	Mesoaortic compression of the left renal vein (nutcracker syndrome): Case reports and review of the literature	Rudloff U, et al	112	1.466	Journal article	2006
9	Journal of Urology	The "nutcracker" phenomenon: An unusual cause for renal varicosities with hematuria	Wendel RG, et al	112	7.450	Journal article	1980
10	*British Journal of Urology*	The nutcracker syndrome: New methods of diagnosis and treatment	Shokeir AA, et al	111	1.690	Journal article	1994
11	*Journal of Vascular Surgery*	Left renal vein transposition for nutcracker syndrome	Reed NR, et al	105	4.268	Journal article	2009
12	*Journal of Vascular Surgery*	Endovascular stenting in the treatment of pelvic vein congestion caused by nutcracker syndrome: Lessons learned from the first 5 cases	Hartung O, et al	104	4.268	Journal article	2005
13	Urology	Transposition of the left renal vein for treatment of the nutcracker phenomenon: Long-term follow-up	Hohenfellner M, et al	92	2.649	Journal article	2002
14	*Journal of Urology*	A possible ontogenic etiology for idiopathic left varicocele	Braedel HU, et al	91	7.450	Journal article	1994
15	*European Journal of Vascular and Endovascular Surgery*	Nutcracker syndrome: An update on current diagnostic criteria and management guidelines	Ananthan K, et al	88	7.069	Journal article	2017

CA = citations per article, IF = 2020 impact factor, US = ultrasound

*Rank by citations per article.

Table 3

The top 20 journals in the number of nutcracker syndrome publications.

Rank*	Journal	NA	NC	CA	IFI	Publisher
1	*Urology*	21	515	24.5	2.649	Elsevier Inc.
2	*Pediatric Nephrology*	17	386	22.7	3.714	Springer Science and Business Media Deutschland GmbH
3	*Journal of Vascular Surgery*	16	666	41.6	4.268	Mosby Inc.
4	*Annals of Vascular Surgery*	14	345	24.6	1.466	Elsevier Inc.
5	*Journal of Vascular Surgery: Venous and Lymphatic Disorders*	13	135	10.3	2.859	Elsevier Inc.
6	*BMJ Case Reports*	12	46	3.8	0	BMJ Publishing Group
7	*Journal of Urology*	11	595	54.0	7.450	Elsevier Inc.
8	*International Urology and Nephrology*	10	103	10.3	2.370	Kluwer Academic Publishers
9	*European Journal of Vascular and Endovascular Surgery*	9	163	84.7	7.069	W.B. Saunders Co., Ltd
10	*Journal of Ultrasound in Medicine*	9	222	24.6	2.153	John Wiley and Sons Inc.
11	*Clinical Nephrology*	9	222	24.6	0.975	Dustri-Verlag Dr. Karl Feistle
12	*Nephrology Dialysis Transplantation*	9	197	21.8	5.992	Oxford University Press
13	*Journal of Vascular Surgery Cases and Innovative Techniques*	9	13	1.4	0	Society for Vascular Surgery
14	*Vascular and Endovascular Surgery*	8	52	6.5	1.089	SAGE Publications Inc.
15	*Journal of Vascular and Interventional Radiology*	6	102	17	3.464	Lippincott Williams and Wilkins
16	*European Journal of Pediatrics*	6	102	17	3.183	Springer Verlag
17	*Kidney International*	6	28	4.6	10.612	Nature Publishing Group
18	*European Journal of Radiology*	5	94	18.8	3.528	Elsevier BV
19	*American Journal of Kidney Diseases*	5	30	6	8.860	W.B. Saunders
20	*Pediatrics International*	5	18	3.6	1.524	Blackwell Publishing

CA = citations per article, IF = 2020 impact factor, NA = number of articles, NC = number of citations.

*Ranked by article number. Journals with the same number of articles are ranked according to the number of citations. Journals with the same number of citations are ranked according to the impact factors.

times or fewer, whereas the number of papers that were cited 60 times or more was <10, and only 2 papers had >200 citations. The top 15 papers in terms of total number of citations are listed in **Table 2**. All the papers were published in only 9 journals. Among these, 5 journals rank among the top 20 journals (**Table 3**) in terms of the number of papers published. These journals account for 36.23% of the total number of papers. The urology journal, published by Elsevier, contained 21 papers and was ranked first.

3.2. Analysis of national and international cooperation

With regard to the top 10 countries in terms of the total number of papers published (**Fig. 5**), the United States had the largest number of published papers, followed by China, South Korea, Japan, Turkey, Italy, the United Kingdom, Spain, France, and India. Although China ranked second only to the United States, the number of published papers from China was only 62.50% of the total for the United States. After excluding documents that were produced with the participation of authors from >25 countries and setting the minimum publication number and minimum citation number for each country to 1, 50 countries met the requirements. Using VOSviewer 1.6.17, to conduct an analysis of international cooperation among the above-mentioned 50 countries, we selected the largest groups of cluster links, as shown in **Figure 6**. **Figure 6A** shows a visual map of the international cooperation network, in which different colors represent different clusters, the size of each circle represents the number of publications from the corresponding country, and the thickness and length of the line between the 2 countries indicate the strength of the cooperative relationship between these countries. As the United States had the largest number of papers, the intensity of cooperation with other countries was the highest, with a total intensity value of 10.

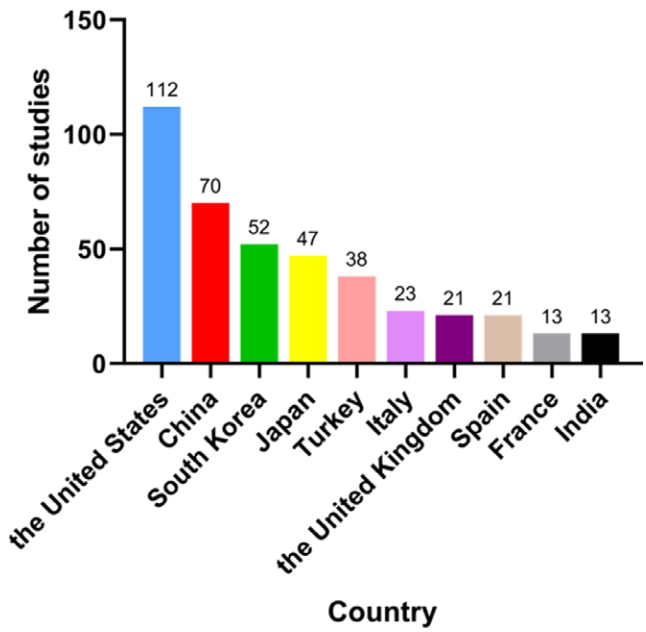

Figure 5. Histogram of the top 10 countries in the number of documents.

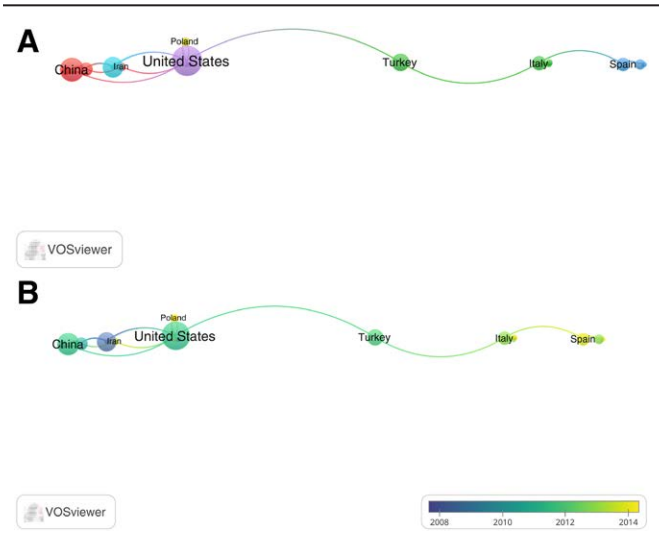

Figure 6. (A) National cooperation network visualization map. (B) National cooperation overlay visualization map.

Of the 10 countries involved, Pakistan and Israel have closer cooperation with the United States than the other 8. In contrast to **Figure 6A**, **Figure 6B** shows an overlay visualization map of international cooperation. Colors in the map correspond to the average publication years. The later (earlier) the average year of publication, the more yellow (blue) the respective circle appears. Germany, the country with the earliest average publication year (2004), is shown in dark blue. The country with the most recent average publication year was Pakistan, which had an average publication year of 2017. With regard to the number of publications, this does not mean that Pakistan had the largest number of published papers in 2017 but that its research in this field started late and is currently active.

3.3. Author and co-authorship analysis

Table 4 lists the top 15 authors in terms of the number of articles published. The numbers of articles in this table were calculated by VOSviewer 1.6.17 taking into account all articles that each author had participated in coauthoring, and auxiliary information was used to distinguish different authors. Shin JI ranked first in terms of the total number of publications, but the total number of citations was not as high as that for Hartung O, who ranked 10th. As mentioned above, the increase in the number of papers over time was closely associated with the active participation of an increasing number of researchers in this field. After setting the minimum number of published papers and the minimum number of citations to 1 and excluding small and unconnected clusters of authors, we selected a closely connected cluster link containing 130 authors. As shown in **Figure 7**, these 130 authors were divided into 13 clusters, which were marked with different colors. The size of each circle indicates the number of papers published, and the thickness and length of the line between 2 authors indicate the degree of closeness of the cooperative relationship between these authors. By reading and analyzing all the articles in each cluster, we summarized the main research contents of the clusters and manually added them. As shown in **Figure 7A**, these contents mainly comprised hematuria, varicocele, intravascular or external stent placement, and other vascular compression syndromes. The overlay visualization map of co-authorship shown in **Figure 7B** differs from that in **Figure 7A** in that the colors of the circles represent the average year of publication, and the earlier (later) was the average year of publication, the closer the color is to blue (yellow). The authors in the yellow cluster in the figure were later than those in the blue cluster in terms of their research in this field. When 2 authors appeared in the author lists of works cited in an article by a third author, they were deemed to have a co-citation relationship. Using the criterion that the total number of citations for each author was at least 20, a total of 260 authors were selected for author co-citation analysis. The results are shown in **Figure 8**, in which all the authors are labeled in red, green, blue, yellow, or purple. Authors with the same color had similar research interests and directions in this field. The size of each circle depends on the total number of citations of works by the corresponding author, and the length and thickness of the line between 2 circles indicate the degree of closeness of the academic relationship between the 2 authors. This picture visually shows the influential authors in the field of NCS-related research and the popular research topics in which they were involved.

3.4. Author keyword co-occurrence analysis

Keywords are substantive words used to summarize the main content and central concepts of a paper. VOSviewer 1.6.17 provides 3 keyword combinations: all keywords, author keywords, and keywords plus. Of these combinations, we selected author

Table 4

Top 15 authors with the most NCS-related articles.

Rank*	Author	No of articles	Total citations†	Citations per article	Affiliation
1	Jae Il Shin	24	338	14.0	Department of Pediatrics, Yonsei University College of Medicine, Seoul, Republic of Korea
2	Jae Seung Lee	18	313	17.3	Department of Paediatrics, The Institute of Kidney Disease, Yonsei University College of Medicine, Severance Children's Hospital
3	Myung Joon Kim	11	225	20.4	Diagnostic Radiology, Yonsei University College of Medicine, Severance Children's Hospital, Seoul, South Korea
4	H. Zhang	10	384	38.4	Department of Vascular Surgery, The First Affiliated Hospital of Medical School of Zhejiang University, Hangzhou, People's Republic of China
5	Jee Min Park	8	215	26.8	The Institute of Kidney Disease, Department of Pediatrics, Yonsei University College of Medicine, Severance Children's Hospital, Seoul, South Korea
6	Antonio Basile	8	49	6.1	Department of Diagnostic and Interventional Radiology, Ospedale Ferrarotto, University Hospital of Catania, Catania, Italy
7	Ming Li	7	305	43.5	Department of Vascular Surgery, First Affiliated Hospital of Medical College, Zhejiang University, Hangzhou, China
8	S.H. Kim	7	268	38.2	Department of Radiology, Seoul National University Hospital, Seoul, Republic of Korea
9	Se-Jun Park	7	58	8.2	Department of Cardiology, Ajou University School of Medicine, Suwon, Korea
10	Olivier Hartung	6	569	94.8	Department of Vascular and Endovascular Surgery, University Hospital of Marseille-Nord, Marseille-Nord, France
11	Lu Tian	6	227	37.8	Department of Vascular Surgery, The First Affiliated Hospital, School of Medicine, Zhejiang University, Hangzhou, China
12	Shanwen Chen	6	204	34.0	Department of Urology, the First Affiliated Hospital of Medical College, Zhejiang University
13	Wei Jin	5	270	54.0	Department of Vascular Surgery, the First Affiliated Hospital of Medical College, Zhejiang University, Hangzhou, China
14	Peter Gloviczki	5	202	40.4	Mayo Clinic, Rochester, United States
15	Sano A.	5	173	34.6	MR Center, Tenri Hospital, Nara, Japan

NCS = nutcracker syndrome.

*Sorted by the number of articles, and authors with the same number of articles are ranked according to the total number of citations.

†Total citations refers to the total number of citations of articles published by each author.

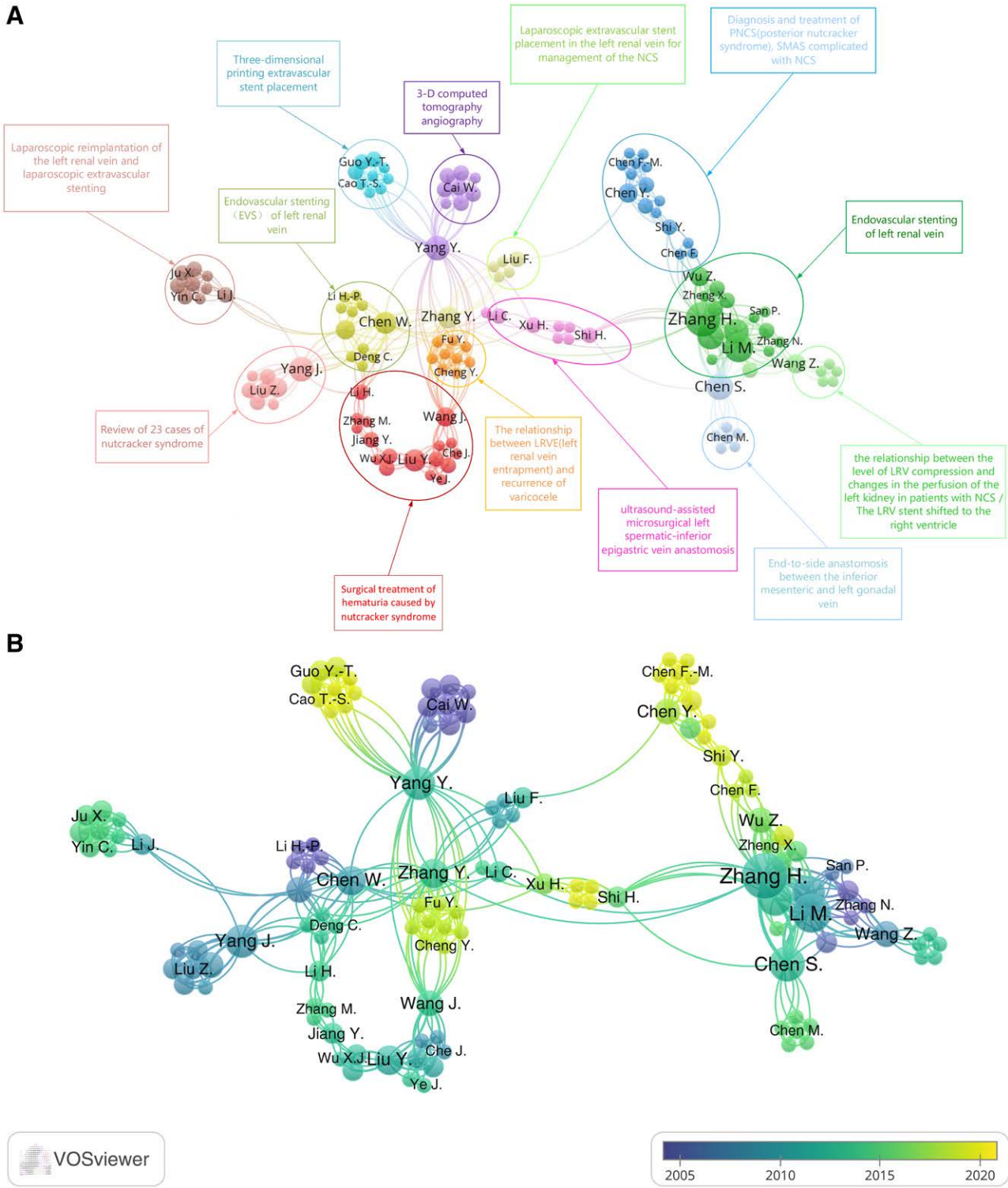

Figure 7. (A) Author cooperation network visualization map. (B) Author cooperation overlay visualization map. LRV = left renal vein, LRVE = left renal vein entrapment, NCS = nutcracker syndrome, PNCS = posterior nutcracker syndrome, SMAS = superior mesenteric artery syndrome.

keywords for the keyword co-occurrence analysis. By compiling synonym sets, different forms of the same keyword, including plurals, abbreviations, different parts of speech, and synonyms, were merged or replaced, and keywords with broad meanings, but no representative meanings, were deleted. Finally, the corresponding overlay visualization map of author keywords was created, as shown in **Figure 9**, where the size of each circle represents the number of times the corresponding keyword appeared and the color of the circle represents the average publication year. The bar in the lower right corner of **Figure 9** shows the relationship between the color and the publication year: the earlier (later) the average publication year, the closer the color is to blue (yellow). The keywords earlier peak velocity, ultrasonography, surgery, orthostatic proteinuria, and LRV hypertension appeared light blue or dark blue. With the passage of time, chronic pelvic pain, endovascular stents, SMA syndrome,

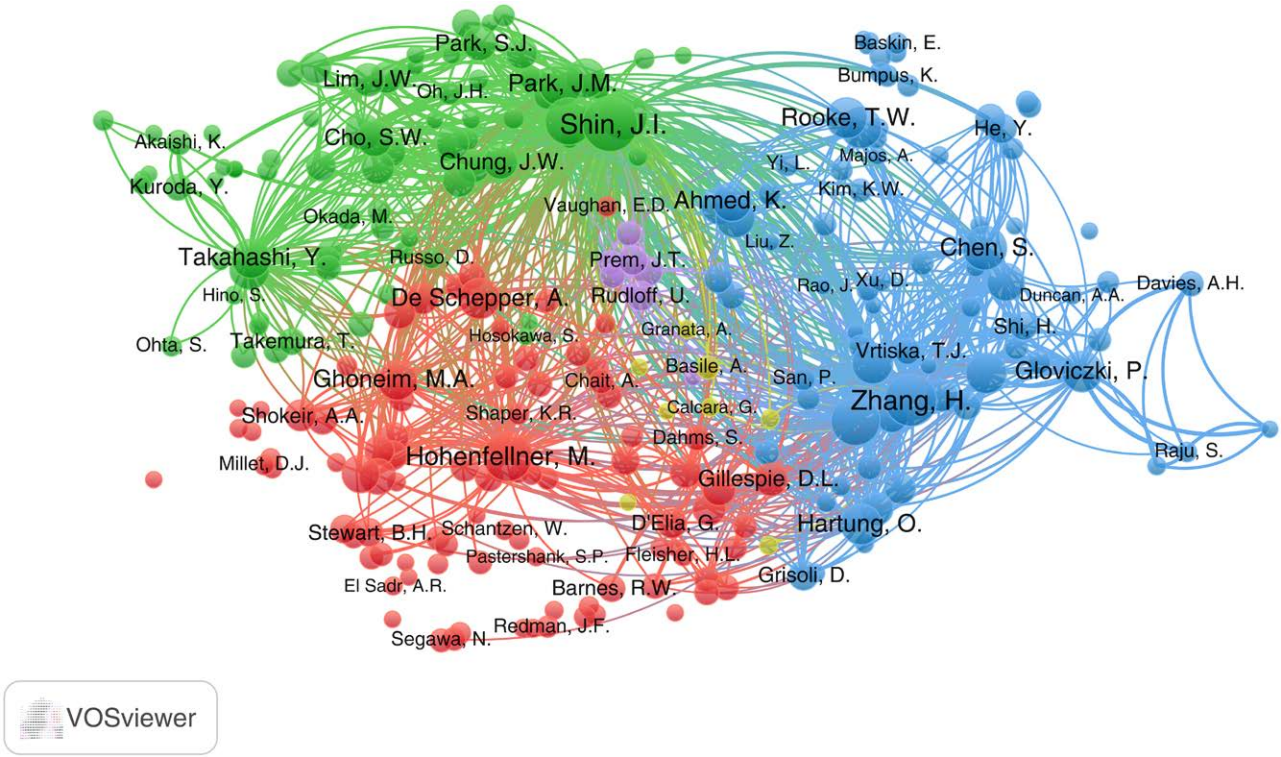

Figure 8. Author co-citation network visualization map.

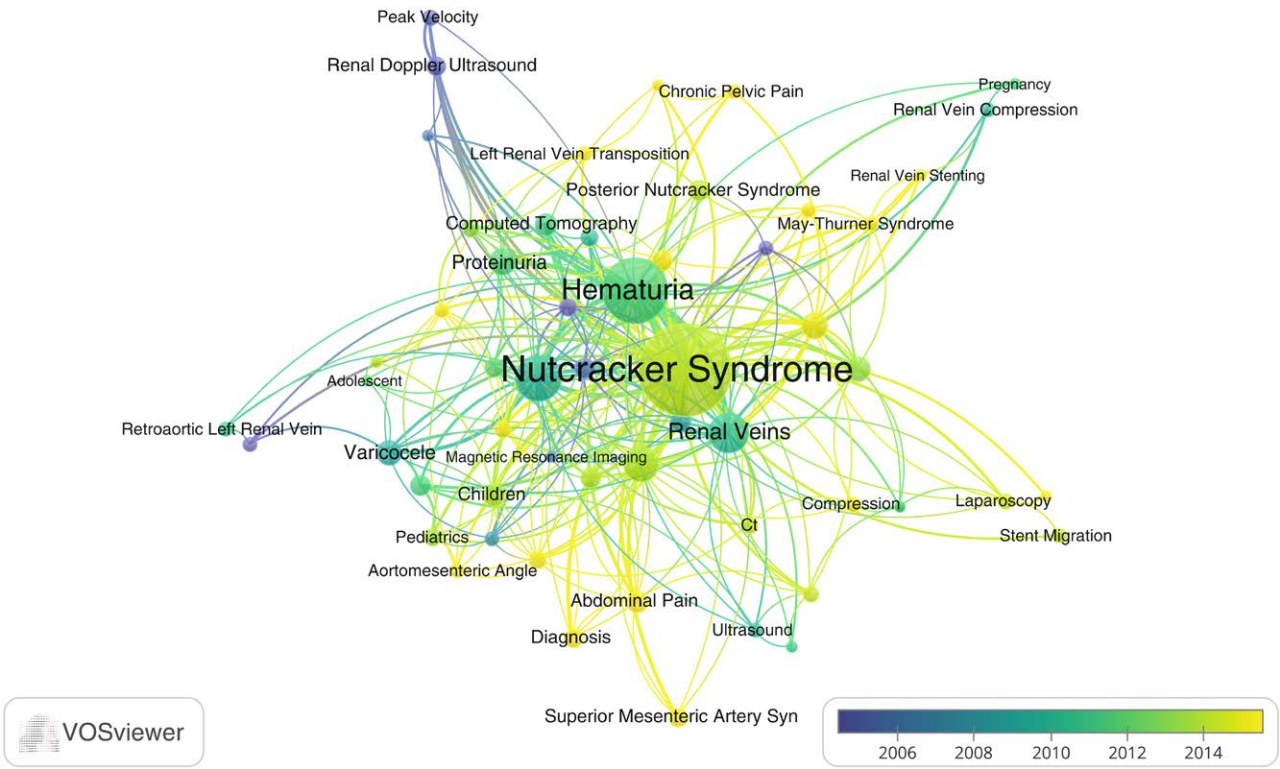

Figure 9. Author keywords co-occurrence overlay visualization map.

abdominal pain, extravascular stent, diagnosis, and other keywords (shown in yellow) gradually emerged. It should be noted that, according to the contents of the literature, we should not simply regard these results as indicating that the early focus of research was abandoned in subsequent research, but should understand them in terms of a gradual shift or deepening of the research focus from anatomical structures, symptoms, and signs to diagnostic criteria and treatment methods.

4. Discussion

It has been 84 years since the NCS was first described, and researchers have since made abundant achievements in this field. For the first time, we used bibliometrics and visualization methods to comprehensively and systematically analyze the global literature on NCS from 1974 to 2021. The latest results for progress in NCS-related research, popular research topics, and research trends are presented in the form of text, statistical charts, and visual maps of knowledge structure.

Of the 552 articles included in this study, the earliest one was published in 1974.[36] We found from relevant references that the earliest research on NCS can be traced back to 1937.[15] However, during the above-mentioned period, only a few articles described the anatomical structures,[16] basic meaning,[37] and specific terms[17] associated with the NCS. As shown in **Figure 3A**, although progress in research was slow from 1974 to 1980, the content in terms of pathological mechanisms has increased.[23] The average number of articles published per year increased from 1981 to 2001, including both the first use of certain diagnostic and surgical methods[38–42] and questions about previous research results.[43] The number of published papers displayed an overall upward trend after 2002, in which the growth rate was the highest and the growth trend was the most obvious from 2008 to 2014. Unlike the number of publications, the overall trend in the number of citations reached a turning point in 2006 (Fig. 3B). Specifically, 2006 and 2015 had the greatest number of citations in the literature. The first review of NCS appeared in 2006,[44] whereas the Clinical Practice Guidelines of the European Society for Vascular Surgery appeared in 2015.[45] As shown in Figure 4, documents with <10 citations accounted for 70.83% of the total number of documents, indicating that many documents had not been fully utilized. The highest number of citations was 412 for an article published by Wittens et al in 2015. Twelve articles had >100 citations and were published in *European Journal of Vascular and Endovascular Surgery, Mayo Clinic Proceedings*, *Journal of Vascular Surgery, Journal of Urology, Radiology, American Journal of Roentgenology, Annals of Vascular Surgery, and British Journal of Urology* (**Table 2**).

Using VOSviewer 1.6.17, we conducted an analysis of international cooperation and co-authorship on the above-mentioned 552 documents and created visual maps of international cooperation (**Fig. 6**) and co-authorship (**Fig. 7**). The former map shows that the countries that participated the most in research on NCS were the United States, the United Kingdom, and Italy. The United States also had the largest number of published papers. In recent years, Pakistan, Singapore, Poland, Belgium, and other countries have conducted research on NCS and published a certain amount of literature that presents the corresponding results. The co-authorship map shows that there was a wide range of collaborations among researchers involved in the study of the NCS. These authors were ranked according to the number of published papers, and relevant information on the top 15 authors was obtained. The results are presented in **Table 4**. Six of these authors were from South Korea, 5 were from China, and 4 were from the United States, France, Italy, and Japan. It is worth noting that, with the exception of Kim SH and Sano A, whose research on NCS started earlier, these authors only started to conduct research on this disease in 2003 and later. From 2005 to 2018, Shin JI from South Korea published 24 papers related to NCS that focused on the application value of renal Doppler ultrasound in the initial screening and diagnosis of the disease.[46–48] Like Shin JI, Hartung O from the Department of Vascular and Endovascular Surgery at the University Hospital of Marseille-Nord in France has been studying NCS since 2005. Although he has published only 6 papers, he has been cited 569 times.

The collection of documents published by researchers in different periods shows popular research topics in this field of study. These topics were not difficult to identify by reading the above-mentioned 552 articles and related literature in combination with the overlay visualization map of author keyword co-occurrence (**Fig. 9**). Early literature records related to NCS have remained at the level of simple descriptions of anatomical structures. With the formal use of terms such as "nutcracker phenomenon," researchers gradually began to study the disease itself. The contents of the articles were wide-ranging and covered all aspects of the NCS. However, it should be noted that the depth of research on each aspect was not the same in all cases. Researchers have unified the general definition of NCS, but their understanding of the pathogenesis and pathological mechanisms of the disease has differed.[49,50] Many clinical case reports have made the diagnostic criteria and treatment of NCS an enduring research topic, especially focusing on the investigation and application of imaging examinations and surgical treatment (**Fig. 1**).

5. Conclusion

NCS have attracted increasing attention, but the utilization rate of related literature is relatively insufficient. The pathogenesis and pathological mechanisms need to be further studied, and the diagnostic criteria and surgical methods will continue to be favored by clinicians. We hope that, as the first study to carry out bibliometric and visualization-based analysis of research into NCS, this can provide a meaningful reference and help for clinicians engaged in research related to this disease in the future.

Author contributions

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Software: Yuchang Jiang, Yong Jiang, Zaili Gan.
Supervision: Yong Jiang, Qinsheng Wang.
Writing – original draft: Yuchang Jiang, Yong Jiang.
Writing – review & editing: Yuchang Jiang, Yong Jiang, Zaili Gan.

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