Original Article

Research on high anal fistula: a bibliometric analysis

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Background: High anal fistula (HAF) treatment is more complicated than low anal fistula treatment. Improper treatment can easily affect anal function. The main treatment of HAF is surgery; however, external medicine and systemic medicine may also be used as adjuvant treatments. This study used bibliometric methods to analyze the relevant literature and provide a rough outline of the current status of HAF research.

Methods: A subject word-search strategy was used to retrieve HAF-related documents from the Science Citation Index Expanded (SCI-E) database. CiteSpace software was used to analyze the exported raw data files, and draw a visual map, and BUSRT was used to detect and analyze the usage of keywords.

Results: A total of 1,020 documents were retrieved using "high anal fistula" as the search term, and the number of documents generally shows an increasing trend over time. These papers were mainly published in developed countries, such as Europe and the United States (US). The US is the country that has carried out the most relevant cooperative research; however, there is a lack of cooperation among a large number of authors. The keyword analysis showed that the current research focus is the treatment of HAF, while research on the prevention of HAF is largely lacking.

Conclusions: More international multi-center clinical research studies on HAF should be conducted, and research on HAF prevention should also be strengthened.

Keywords: High anal fistula (HAF); surgery; anal function; bibliometric analysis

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Introduction

With an incidence of approximately 12–28 cases per 100,000 people per year, anal fistula is a common perianal disease (1). When other diseases involve the perianal, the incidence of anal fistula is higher (2,3). This disease is more common in young and middle-aged men (1). It causes obvious pain to the patient and significantly affects their quality of life (3). The high anal fistula (HAF) and its branches pass through the levator ani muscle and above the anorectal ring (4). In recent years, the incidence rate of HAF has increased significantly (1). Additionally, the treatment of HAF is difficult, and the recurrence rate is high (1).

The local anatomy and physiological environment of the perianal area is special, which can easily lead to perianal infection, swelling, pain, and the formation of abscesses, which can affect the normal physiological functions of patients. In severe cases, HAF can lead to sepsis, endangering the life and safety of patients. Clinical studies suggest that the pathogenic factors of HAF mainly include anal gland infection, rectal and anus injury, perineal surgery, tuberculosis, ulcerative colitis, and blood infection, and the clinical manifestations mainly include perianal pus, itching, swelling and pain, and systemic symptoms (5). The treatment of HAF is more complicated than that of low anal fistula. Improper treatment can easily affect anal function; thus, choosing an effective treatment method is of great significance in improving clinical efficacy and improving anal function (6). The current clinical research mainly focuses on the surgical treatment of HAF. Careful

examination before surgery is necessary. The most used examinations include digital examination, magnetic resonance imaging (MRI) and ultrasonography. During the operation, surgeon make further examination by probe to ensure all fistula be found and treated.

At present, the main treatment for HAF is surgery; however, external medicine and systemic medicine are used as adjuvant treatments. There are a variety of surgical procedures for the treatment of HAF, of which sphincter preservation surgery has become the most popular operation (7-9). Common surgical procedures include sphincter-preserving thread-drawing surgery, sliding-flap surgery, transsphincteric fistula ligation (ligation) of the intersphincteric fistula tract (LIFT), video-assisted anal fistula treatment (VAAFT), laser ablation and closure of the anal fistula, and autologous fat-derived stem cell therapy. Though these treatments bring patients high cure rate, some patients still suffer from recurrence. The virtual and actual thread-hanging method proposed by this research group has also achieved good results in the treatment of HAF (10).

Bibliometrics is a literature research method that has gradually emerged in recent years. It can capture the current status of a specific research field and help researchers to understand the specific field. It can also provide a macro understanding of the research, which can help researchers to choose a reasonable research direction and suitable cooperation objects (11,12). To clarify the current status of research related to HAF, we use bibliometric methods to analyze the basic information of the relevant literature, including the author, the country of the research source, the research institution, and the use of keywords to provide a rough outline of the current status of HAF research.

Methods

Literature source

This research used the Science Citation Index Expanded (SCI-E) database in the Web of Science Core Collection (WOSCC), which is commonly used in bibliometrics research, as the data source to search. The SCI-E was established and is published by The Institute for Scientific Information. It contains more than 8,000 important journal paper titles and information cited in the literature. The literature comprises more than 170 natural science articles, including articles in the area of medicine and science. The SCI-E provides a citation retrieval function that allows quantitative analyses of documents in a certain field to be conducted. The SCI-E is an important basis and data source for current bibliometric research and scientific research evaluation.

Retrieval method

We used a subject term to retrieve documents. The subject term used in this study was "high anal fistula." The publication time for the target documents was not limited; that is, the publication period for the documents ran from the earliest publication date of the database documents [1900] to the final retrieval date of this research (July 25, 2021).

Analysis method

All the retrieved records were exported, and the bibliographic information was cited in plain-text format to form the original data file. CiteSpace software was used to analyze the exported original data file. The following dimensions were included the analysis: annual changes in the number of publications, trends, annual changes in the number of times a document has been cited, the distribution of the source country and institution, the distribution of the journals published, the distribution of the authors, and the keywords used. The main indicators included the number of documents published per year, the number of citations per year, the number of documents published per country, institutions, and authors, the centrality scores of countries, institutions and authors, and the utility status of keywords.

Statistical analysis

This study is a descriptive study, and data are expressed in quantities and percentages. No statistical comparisons were conducted; thus, no P value was set.

Results

Retrieval results

A total of 1,150 records (see *Table 1*) were retrieved in the preliminary search, and 130 duplicate records were eliminated. The actual related research documents totaled 1,020, and the citation frequency was 25,451. The average number of citations for each item was 24.95, and the h-index was 76 (see *Table 1*). Of the 1,020 documents there were 869

Table 1 Type of literature

Literatures	Records	% of 1,020
Article	869	85.20
Proceedings article	120	11.76
Review	108	10.59
Letter	17	1.67
Editorial document	12	1.18
Meeting abstract	12	1.18
Early access	10	0.98
Note	2	0.20

The preliminary search result was 1,150 records, after excluding 130 duplicate documents, 1,020 documents remained.

original articles, 120 conferences, 108 reviews, 17 letters, 12 editorial documents, 12 conference abstracts, 10 papers for priority publication, and 2 notes (see *Table 1*).

Annual changes in the number of documents and the number of citations

The annual analysis of HAF research documents showed that the number of documents published in this field has generally increased over time, as has the number of citations of these documents (see *Table 2*, and *Figures 1* and *2*).

The distribution of countries and institutions

CiteSpace V software was used to analyze the original data and documents to generate a national visualization map (see *Figure 3*) that depicts the countries conducting research in this field and their mutual cooperation. The larger the circle representing a country, the more papers that country has published. The number of nodes in the graph was 76, and the relationship between the nodes was 136. Thus, 76 countries have published research in this field, and 136 cooperative relationships have been established. Similarly, the number of nodes in the organization visualization map was 453 (see *Figure 4*), and there were 279 interconnections. In terms of the quantitative statistics, the top 5 countries for the number of publications were the United States (US), the United Kingdom, Germany, China, and France (see *Table 3*). The 5 countries with the top centrality scores were the US, the United Kingdom, Australia, Canada, and Italy (see *Table 4*). The top 5 institutions for the number of

Table 2 Annual publication of literature on HAF

Years	Records	% of 1,020
2021	38	3.73
2020	60	5.88
2019	69	6.76
2018	45	4.41
2017	52	5.10
2016	46	4.51
2015	45	4.41
2014	40	3.92
2013	35	3.43
2012	47	4.61
2011	54	5.29
2010	49	4.80
2009	40	3.92
2008	41	4.02
2007	32	3.14
2006	32	3.14
2005	22	2.16
2004	22	2.16
2003	19	1.86
2002	23	2.25
2001	22	2.16
2000	22	2.16
1999	15	1.47
1998	26	2.55
1997	16	1.55
1996	25	2.45
1995	23	2.25
1994	17	1.67
1993	13	1.27
1992	6	0.59
1991	17	1.67
1990	1	0.10
1985	2	0.20
1984	1	0.10
1983	2	0.20
1981	1	0.10

HAF, high anal fistula.

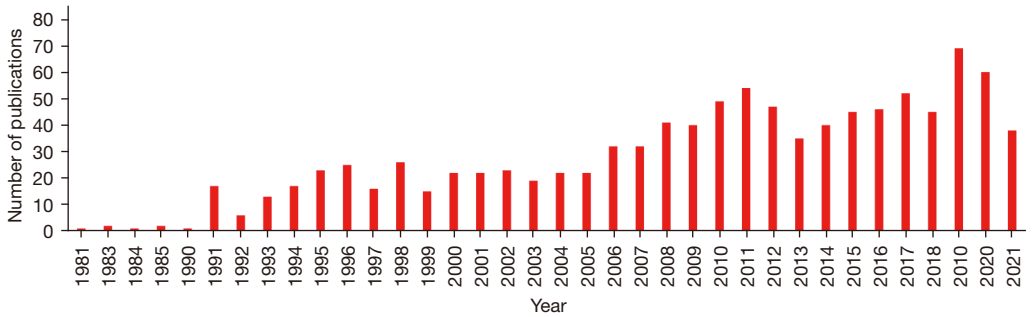

Figure 1 Annual changes in the number of publications.

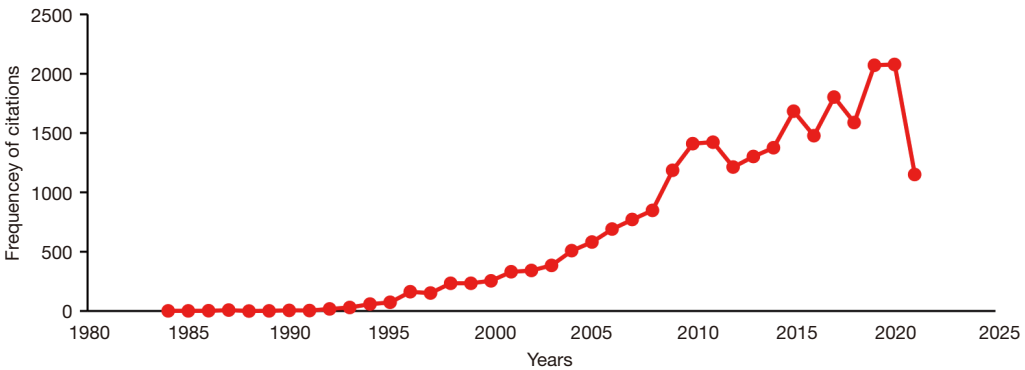

Figure 2 Annual changes in the frequency of citations.

Figure 3 Visualization map of countries.

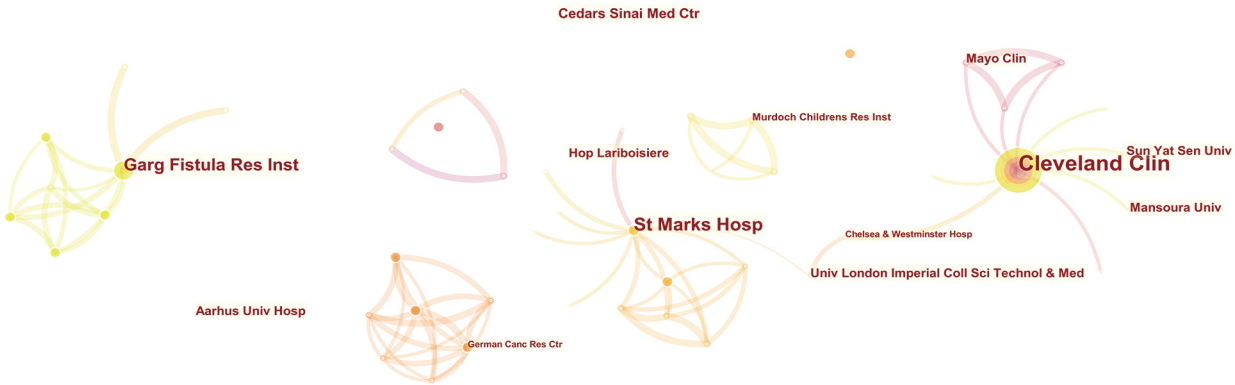

Figure 4 Visualization map of institutions.

publications were Thomas Jefferson University, the Mayo Clinic, the Hospital of Special Surgery, Rush University, and the Cleveland Clinic (see *Table 5*). The top 4 centrally ranked institutions were Thomas Jefferson University, Tel Aviv University, the University of Melbourne, and Rush University (see *Table 6*).

Authors

In the research field of HAF, the top 3 authors with the most articles published were Garg, Emile, and Kohler (see *Table 7*). The centrality score of all the authors did not reach 0.1, and there was little collaboration between authors (see *Figure 5*). The top 3 cited authors were Parks, Van Koperen, and Williams (see *Table 8*, *Figure 6*). The top 3 authors cited in relation to centrality scores were Parks, Fazio, and Williams (see *Table 9*).

Distribution of journals

The results of this study included 1,020 papers from 271 journals, of which 13 had published more than 10 papers (see *Table 10*). A total of 514 papers were published, accounting for 50.39% of the total number of papers (see *Table 10*). The top 3 journals based on the frequency of the citations were *Dis Colon Rectum*, *Brit J Surg*, and *Int J Colorectal Dis* (see *Table 11*). The top 3 journals based on the centrality of the citations were *Lancet*, *World J Surg* and *Surg Gynecol Obstet* (see *Table 12*).

Keywords

CiteSpace V software analyzed the use of keywords and generated a keyword co-occurrence map (see *Figure 7*), where node (N) =374 (i.e., 374 keywords appeared in these documents). The number of times these keywords appeared in the same articles was E =2,975. The top 3 keywords in terms of frequency were “fistula-in-ano”, “in-ano”, and management (see *Table 13*), and the top 3 keywords in terms of centrality were “fistula-in-ano”, “Crohn’s disease”, and “fistula” (see *Table 14*). CiteSpace was used to perform a burst detection on the keywords (see *Figure 8*).

Discussion

This study analyzed 1,020 articles and their cited data, and the results showed that the number of articles published and the number of citations on the subject of HAF had generally increased over time. These studies were mainly published in developed countries, such as Europe and the US. There were some cooperative relationships between countries and institutions; however, there was very little cooperation between authors. The literature in this field has mainly been published in the surgical journals of colorectal diseases. The keyword analysis showed that the research mainly focused on the management of anal fistula. Additionally, the distribution of keywords also suggested that in articles in this field, the keywords are relatively inconsistent, and multiple keywords with similar meanings coexist, which is

Table 3 Top 10 countries for publications

Rank	Countries	Publications
1	USA	215
2	England	95
3	Germany	85
4	China	76
5	France	64
6	Netherlands	61
7	Italy	55
8	Spain	47
9	India	40
10	Japan	33

Table 4 Centrality for countries

Rank	Countries	Centrality
1	USA	0.54
2	England	0.32
3	Australia	0.19
4	Canada	0.13
5	Italy	0.08
6	Spain	0.06
7	Germany	0.05
8	Netherlands	0.05
9	Sweden	0.05
10	Japan	0.05

Table 5 Top 10 institutions for publications

Rank	Institutions	Publications
1	Thomas Jefferson Univ	243
2	Mayo Clin	216
3	Hosp Special Surg	138
4	Rush Univ	114
5	Cleveland Clin	103
6	Duke Univ	73
7	Univ Calif San Francisco	71
8	NYU	60
9	Univ Penn	54
10	Sichuan Univ	46

Table 6 Top 10 institutions for centrality

Rank	Institutions	Centrality
1	Thomas Jefferson Univ	0.07
2	Tel Aviv Univ	0.05
3	Univ Melbourne	0.04
4	Rush Univ	0.04
5	Charite Univ Med Berlin	0.03
6	Univ Penn	0.03
7	Cleveland Clin	0.03
8	Hosp Special Surg	0.03
9	Mayo Clin	0.03
10	Univ Helsinki	0.02

Table 7 Top 10 authors based on publications

Rank	Authors	Frequency
1	Garg PJ	14
2	Emile SH	7
3	Kohler A	7
4	Herold A	6
5	Arroyo A	6
6	Schoetz DJ	5
7	Ailsa Hart A	5
8	Parello A	5
9	Remzi FH	5
10	Yagnik VD	4

not conducive to a bibliometric analysis.

With the increase in the incidence of related diseases (e.g., AIDS, tuberculosis, and Crohn's disease) or behavior (anal sex), the prevalence of HAF has also shown a significant increase (13). Among them, anal fistula is the most common perianal disease of Crohn's disease (14). The incidence of anal fistula is significantly increased in patients with active intestinal disease, and can be as high as 92% when the rectum is involved (14). However, most patients with specific anal fistula do not get a timely diagnosis before surgery, which leads to treatment failure. Thus, both scholars at home and abroad agree that the timely identification of the cause of anal fistula and adopting standardized and correct treatment measures can improve

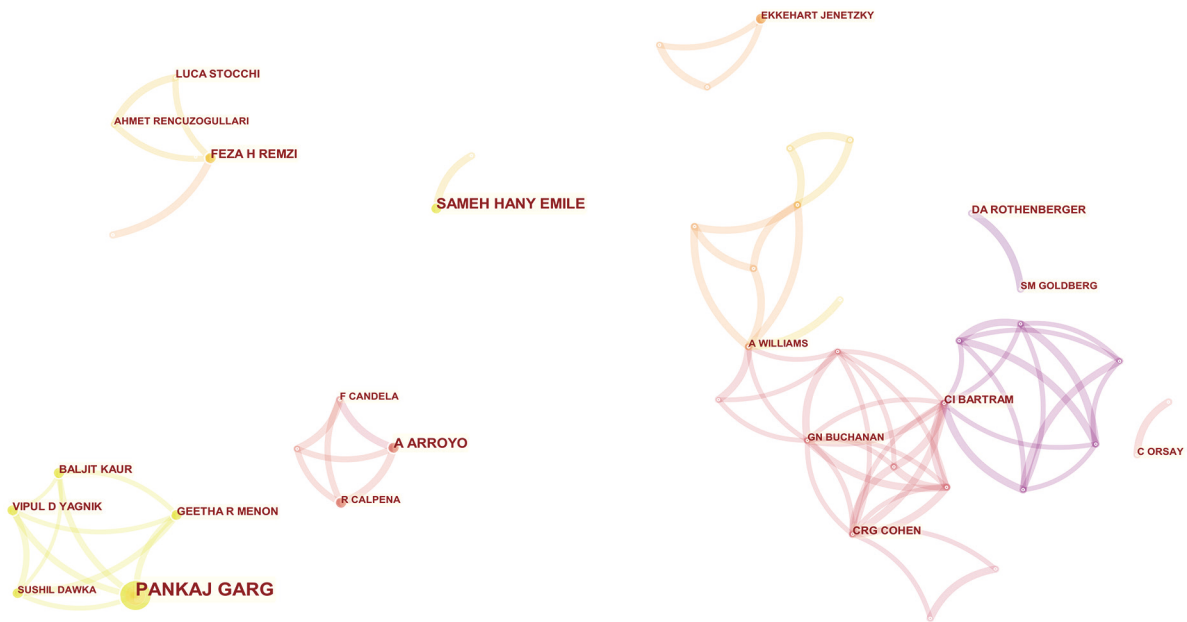

Figure 5 Visualization map of authors' collaborations.

Table 8 Top 10 authors by frequency of co-citations

Rank	Authors	Frequency
1	Parks AG	307
2	Van Koperen PJ	127
3	Williams JG	121
4	Buchanan GN	120
5	Garciaaguilar J	110
6	Lunniss PJ	110
7	Schouten WR	92
8	Ortiz H	90
9	Ellis CN	86
10	Van Der Hagen SJ	86

the cure rate of anal fistula (7-9).

The main themes of HAFs for patients are pain and local repeated infections and exudations, which reduce patients' quality of life. The long-term prognosis of HAF is relatively good, and generally does not have malignant consequences (15). HAF is not a rare disease; however, there are not many studies on HAF. In this study, the SCI-E database was searched from 1,900 to the present day, and only 1,020 articles were retrieved. Judging from the amount of publications, the medical community has not paid enough attention to this disease. In many medical centers, there are no specialized departments to deal with this kind of problem. Indeed, it is generally treated by general surgery or intestinal surgery departments. The focus of these 2 departments is often on internal organ tumors and acute abdomens. It is difficult for doctors to focus on research of HAF (16). Thus, we found that there were very few high-quality multi-center clinical studies.

There has still been great progress in the treatment of anal fistula; however, the treatment of complex HAF remains very difficult for clinicians. Traditional surgery has a high cure rate, but it has the disadvantage of damaging the anal sphincter. The sphincter-sparing surgery that has emerged over the decades has protected postoperative anal function, but the recurrence rate is significantly higher than that of traditional surgery, which has become a major obstacle to the promotion of sphincter-sparing surgery (17). Thus, looking for a new technology that can effectively retain the function of the sphincter and anus, but can also effectively reduce the recurrence rate has become a popular area of research in the treatment of anal fistula. According to previous studies, predictive factors for recurrence after surgical treatment of high anal

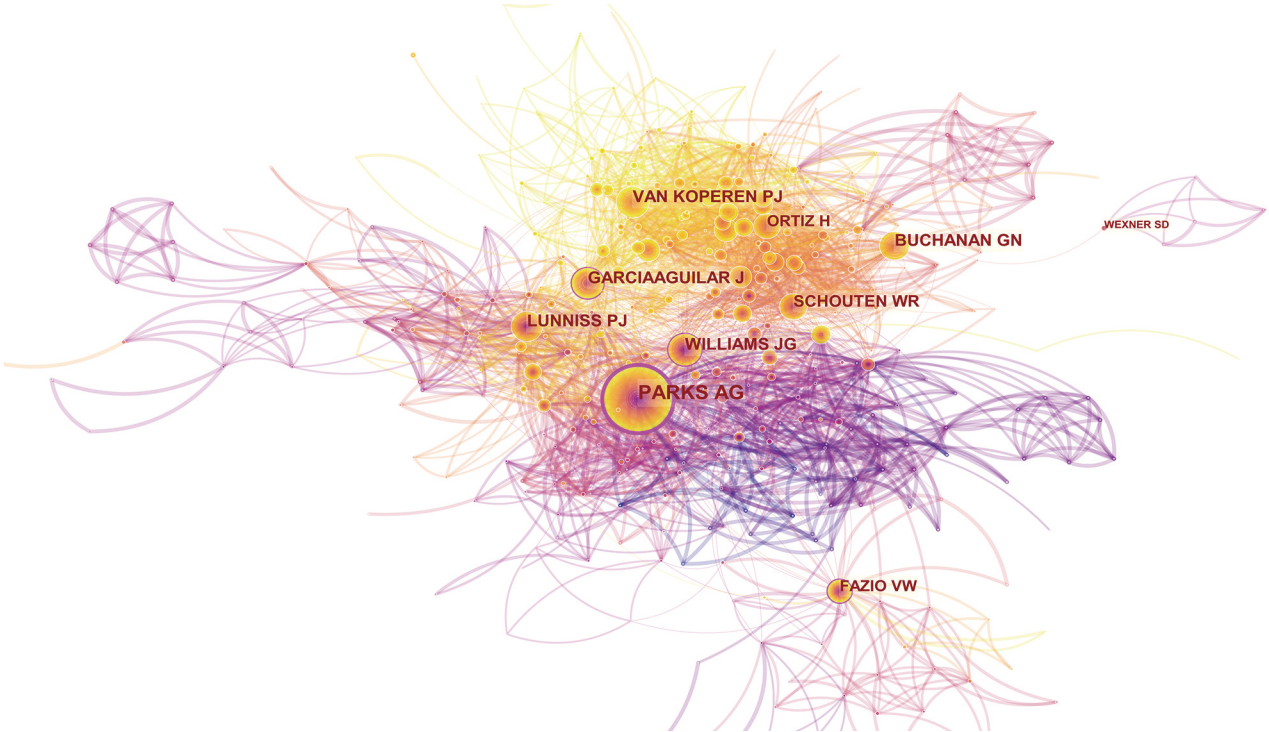

Figure 6 Visualization map of authors by co-citations.

Table 9 Top 10 authors by centrality of co-citations

Rank	Authors	Centrality
1	Parks AG	0.45
2	Fazio VW	0.16
3	Williams JG	0.15
4	Garciaaguilar J	0.11
5	Van Koperen PJ	0.09
6	Aguilar PS	0.09
7	Lunniss PJ	0.08
8	Sainio P	0.08
9	Ozuner G	0.07
10	Law PJ	0.07

Table 10 Top 10 journals based on the number of articles

Journals	Records	% of 1,020
Diseases of the Colon Rectum	172	16.863
Colorectal Disease	72	7.059
International Journal of Colorectal Disease	58	5.686
Techniques in Coloproctology	44	4.314
Journal of Pediatric Surgery	37	3.627
British Journal of Surgery	30	2.941
Pediatric Surgery International	21	2.059
Chirurg	16	1.569
Journal of Gastrointestinal Surgery	15	1.471
World Journal of Gastroenterology	14	1.373

fistula include: internal opening unidentified, horseshoe extensions, prior anal surgery, seton placement surgery, and multiple fistula tract (18).

At present, sphincter-preserving thread-drawing operation, sliding-flap operation, and transsphincteric fistula ligation are the most widely used in clinical practice. Sphincter-preserving thread-drawing surgery can be used to treat complex HAF, and has the advantage of also reducing the risk of anal incontinence (18). This method uses the drainage and foreign body stimulation effects of thread-hanging, and discards the chronic strangulation effect that

Table 11 Top 10 journals by frequency of citation

Rank	Journals	Frequency
1	*Dis Colon Rectum*	773
2	*Brit J Surg*	631
3	*Int J Colorectal Dis*	446
4	*Colorectal Dis*	414
5	*Ann Surg*	290
6	*Am J Surg*	284
7	*Tech Coloproctol*	270
8	*Gut*	244
9	*Gastroenterology*	242
10	*Lancet*	199

Table 12 Top 10 journals by centrality of citation

Rank	Journals	Centrality
1	*Lancet*	0.16
2	*World J Surg*	0.10
3	*Surg Gynecol Obstet*	0.10
4	*Am J Roentgenol*	0.09
5	*Gastroenterology*	0.08
6	*Radiology*	0.08
7	*Acta Chir Scand*	0.08
8	*New Engl J Med*	0.07
9	*J Pediatr Surg*	0.07
10	*Brit Med J*	0.07

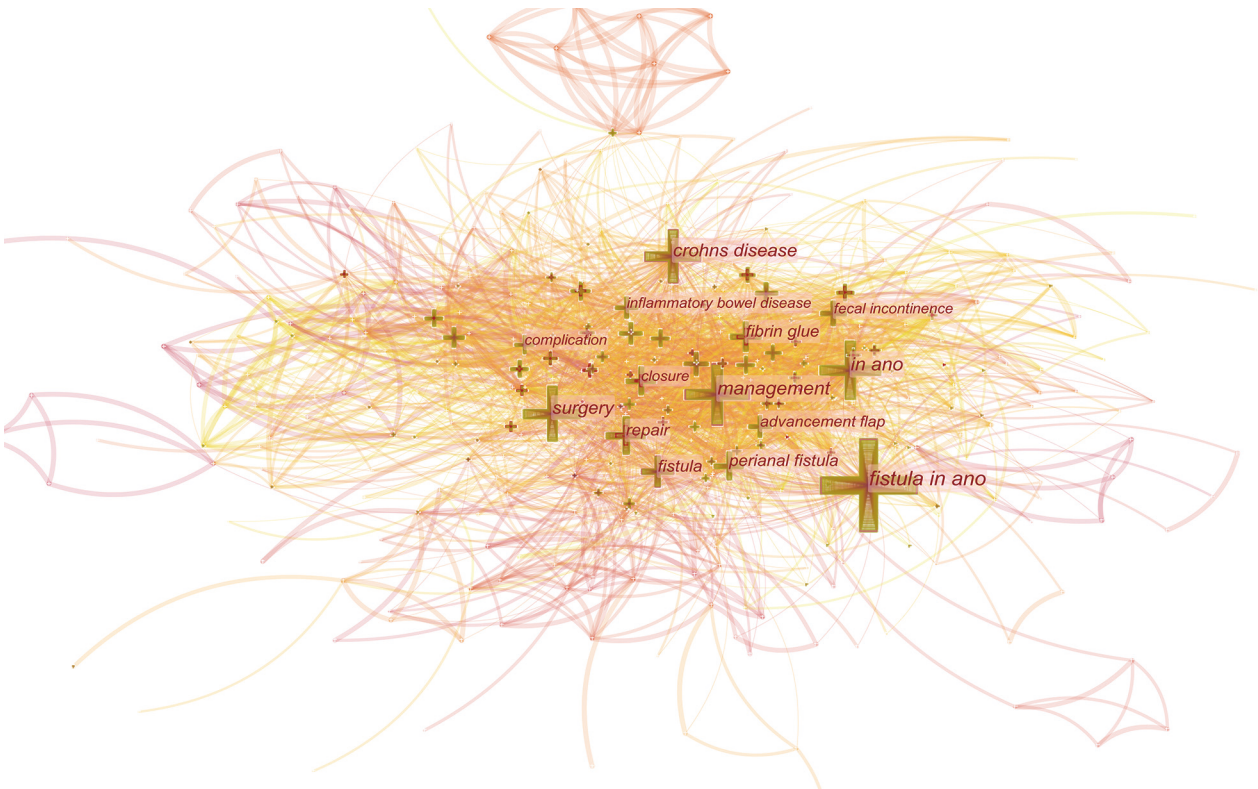

Figure 7 Visualization map of keywords co-occurrence.

causes muscle damage, postoperative pain, and prolonged treatment. It adopts a different view to the traditional one that traditional thread-hanging therapy must cut off the muscle tissue. Under the premise of removing the primary infection foci, thread-hanging without breaking the sphincter has the advantage of protecting the anal function, which is better than the traditional incision thread-hanging method. It achieves the goal of not only curing anal fistula, but also preserving the external anal sphincter completely, thereby protecting the anal function to the utmost extent,

Table 13 Top 10 keywords by frequency

Rank	Keywords	Frequency
1	Fistula-in-ano	364
2	In-ano	225
3	Management	213
4	Crohn's disease	178
5	Surgery	160
6	Repair	101
7	Fistula	97
8	Fibrin glue	94
9	Perianal fistula	93
10	Advancement flap	71

Table 14 Top 10 keywords by centrality

Rank	Keywords	Centrality
1	Fistula-in-ano	0.18
2	Crohn's disease	0.14
3	Fistula	0.13
4	Management	0.12
5	Surgery	0.1
6	Complication	0.09
7	Repair	0.07
8	Fecal incontinence	0.07
9	Anorectal fistula	0.07
10	Experience	0.07

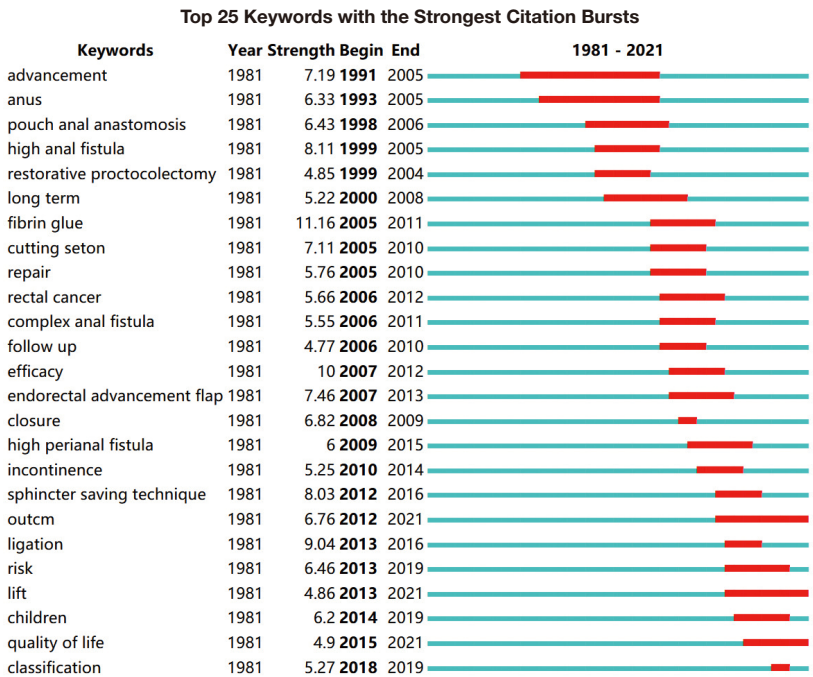

Figure 8 Top 25 keywords with the strongest citation bursts.

which is in line with the development trend for modern minimally invasive surgery (19).

Push-flap surgery is suitable for patients with impaired anal function due to fistula incision, and can also be used to close rectal-urethral or recto-vaginal fistulas (20). This operation cuts off the fistula connected to the rectum after the infection is controlled, and takes normal autologous healthy tissue to cover the internal mouth. A good blood supply and tension-free anastomosis of the moving valve are key to the operation (21).

Another technique involves the transsphincteric fistula tract (the LIFT). The LIFT technique is mainly based on the idea of closing the internal mouth and removing the infected crypt tissue from the intersphincteric plane (22). A

meta-analysis showed that the success rate of standard LIFT surgery for the treatment of anal fistula is between 61% and 91%, the healing time is usually 4 to 8 weeks, and there are few complications and very few anal incontinences (23). Both simple and complex transsphincteric anal fistulas can be treated with LIFT surgery (24,25).

VAAFT is an emerging treatment method that brings hope to the treatment of anal fistula (26). It is still unclear whether video assistance is critical to the success of the surgery; some supporters believe that even the lesions missed by MRI can be found using VAAFT. However, there is currently a lack of evidence to support this view, and more detailed research needs to be conducted (27,28).

The laser-ablation treatment of fistula is still in its infancy, and the surgical methods applied at different centers are quite different, a standard technical process has not been established, and there are also great differences in the treatment of the internal mouth (29,30).

Autologous fat-derived stem cells are also a new option for the treatment of complex anal fistula, but to date, there is not enough evidence to prove their effectiveness (31). This technique can be used alone to treat anal fistula by injecting autologous stem cells around the fistula or into the fistula, or it can be combined with fibrin glue or push flaps (31,32).

Thus, many novel surgical methods have emerged in recent years that have brought new hope to the surgical treatment of anal fistula; however, the effectiveness of these emerging surgical methods cannot be evaluated based on the current data or compared with the classic sphincter-sparing surgery. As we are still in the early days, multi-center, large sample, parallel randomized controlled trials need to be conducted (33).

This study had a number of limitations. This study only searched for articles in the SCI-E, which is a common databased used for bibliometric analysis; however, other databases may include articles that do not appear in the SCI-E. Thus, some documents may have been missed. Second, inconsistences in the keywords of different publications may have had an effect on the analysis of utility and the centrality of the keywords.

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Footnote

Conflicts of Interest: All authors have completed the ICMJE uniform disclosure form (available at https://dx.doi.org/10.21037/apm-21-3190). The authors have no conflicts of interest to declare.

Ethical Statement: The authors are accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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