

Hot topics in global perianal fistula research

A scopus-based bibliometric analysis

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Abstract

Background: The number of citations a scientific paper has received indicates its impact within any medical field. We performed a bibliometric analysis to highlight the key topics of the most frequently cited 100 articles on perianal fistula to determine the advances in this field.

Methods: The Scopus database was searched from 1960 to 2018 using the search terms “perianal fistula” or “anal fistula” or “fistula in ano” or “anal fistulae” or “anorectal fistulae” including full articles. The topic, year of publication, publishing journal, country of origin, institution, and department of the first author were analyzed.

Results: The median number of citations for the top 100 of 3431 eligible papers, ranked in order of the number of citations, was 100 (range: 65–811), and the number of citations per year was 7.5 (range: 3.8–40.1). The most-cited paper (by Parks et al in 1976; 811 citations) focused on the classification of perianal fistula. The institution with the highest number of publications was St Mark’s Hospital, London, UK. The most-studied topic was surgical management (n=47). The country and the decade with the greatest number of publications in this field were the USA (n=34) and the 2000s (n=50), respectively.

Conclusion: The 100 most frequently cited manuscripts showed that surgical management had the greatest impact on the study of perianal fistula. This citation analysis provides a reference of what could be considered the most classic papers on perianal fistula, and may serve as a reference for researchers and clinicians as to what constitutes a citable paper in this field.

Abbreviations: ERUS = endorectal ultrasonography, GS = Google Scholar, JCR = Journal Citation Reports, MRI = magnetic resonance imaging, WOS = Web of Science.

Keywords: bibliometric analysis, citations, hot topics, perianal fistula, surgery

1. Introduction

Perianal fistula is one of the most difficult surgical disorders. Protecting anal function and preventing anal incontinence are very difficult for surgeons, especially complex perianal fistulas (high, Crohn diseases and low fistulas with compromised sphincters). Although new minimally invasive surgical procedures continue to be developed, the problems of postoperative recurrence and incontinence have yet to be resolved. For more

than a century, the study of perianal fistula has produced a large body of clinical and scientific papers, which have led to a greater understanding of the etiology and pathology of the disease and advances in its surgical treatment and imaging examinations. In particular, treatment of perianal fistula in the presence of Crohn disease has also made many new advances, such as the use of stem cells. Since scientists Cole and Eales in 1917 proposed a quantitative comparison of the anatomical literature, bibliometric citation analysis, which examines the citation history of individual papers on a topic of interest, has developed gradually. A citation is received when a publication is referenced by another peer-reviewed paper. As a high number of citations is a proxy for a manuscript’s contribution to the current body of knowledge on a subject, the 100 most-cited articles represent the core works of the understanding and treatment of a given disease. Eigenfactor scores are used to determine a journal’s impact, which are listed in Thomson’s Journal Citation Reports (JCR).

Many medical and surgical specialties have utilized the citation rank analysis to identify the most influential papers in their field, which include burns,^[1] cardiac surgery,^[2] laparoscopic surgery,^[3] abdominal surgery,^[4] and cardiovascular medicine.^[5] To date, there have been no studies undertaken to determine the most influential papers in the field of perianal fistula. We aim to analyze the 100 most-cited papers on perianal fistula to provide a unique insight into how our understanding of perianal fistula has developed and changed the management of this disease.

2. Materials and methods

The Scopus database was searched to identify the top 100 most-cited manuscripts on perianal fistula using “perianal fistula” or

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“anal fistula” or “fistula in ano” or “anal fistulae” or “anorectal fistulae” as advanced search terms. The search was limited to full papers in all languages, and the results were ranked by citation number in descending order. Considering the potential bias of increased numbers of citations in older papers, we also calculated the citation rate to reflect the annual reference rate. Articles with the same number of citations were sorted by the citation rate. The number of citations obtained from each study was also evaluated using the Google Scholar database for a comparative analysis.

The title, first and senior author, institution and department of the first author, date of publication, topic, and the country of origin of each paper were recorded for further analysis. The 5-year impact factors and Eigenfactors of each journal publishing the manuscripts were recorded.

3. Results

The Scopus database search returned 3431 full-length, English and non-English language papers. The 100 most-cited articles are listed in Table 1^[6–105]. The number of citations in these 100 papers ranged from 811 to 865, with a median number of 100; with Google Scholar (GS), the median was 159 (range: 1364–1389). The most cited paper, by Parks et al in 1976 focused on the classification of perianal fistula, which was published in 1976 and cited 811 times.^[6] The oldest paper in the top 100 most-cited articles was by Milligan and Morgan^[69] and was published in the *Lancet* in 1934. The most recent paper in the top 100 most-cited papers by Panés et al was published in the *Lancet* and studied stem cell treatment of complex perianal fistulas in Crohn disease, which was cited 101 times.^[54] The 2000s yielded the highest number of influential papers (n=50; 704 citations).

The 100 most-cited papers were published in 22 journals, with the number of manuscripts per journal ranging from 1 to 44. Impact factors of the 22 journals ranged from 53.254 (*Lancet*) to 2.031 (*Digestive Surgery*). The *Journal of the Diseases of the Colon and Rectum* published most papers (n=44; 5804 citations). The *Lancet* also had the highest 5-year impact factor (52.665) and Eigenfactor (0.40).

The 100 most-cited papers were from 18 countries, including 13 non-English-speaking countries. The greatest number of publications was from the USA (n=34; 4613 citations) (Fig. 1). The UK and Netherlands were responsible for 27 (4099 citations) and 10 papers (1188 citations), respectively. St. Mark's Hospital, London, UK had the highest number of papers in the top 100 with 14 papers generating 2598 citations. Professor Buchanan GN, from St Mark's Hospital, had the highest number of first authorships in the top 100 most-cited papers with a total of 4 manuscripts,^[17,24,40,63] one of which was published in the *Lancet* (impact factor 53.254). Three authors had 3 publications each in which they were first author, namely Schwartz et al,^[8,30,72] Williams et al,^[18,35,58] and Lunniss et al.^[25,33–34]

The citation rate for the top 10 most-cited manuscripts ranged from 50.5 for Panés et al in 2016 (“Expanded allogeneic adipose-derived mesenchymal stem cells (Cx601) for complex perianal fistulas in Crohn disease: a phase 3 randomized, double-blind controlled trial”)^[54] to 14.3 for Whiteford et al in 2005 (“Practice parameters for the treatment of perianal abscess and fistula-in-ano (revised)”)^[19]. The USA had the highest number of papers in the top 10 according to citation rate with 5, followed by Spain with 4 and the UK with 1. Interestingly, 3 in the top 10 papers according to citation rate were from Spain, which investigated stem cell therapy.

Table 1

The top 100 most-cited papers on perianal fistula.

Rank	First author	Scopus	Citation rate	Google Scholar
1	Parks, AG ^[6]	811	19.3	1364
2	Garcia-Olmo, D ^[7]	415	46.1	628
3	Schwartz, DA ^[8]	360	21.2	530
4	Garcia-Aguilar, J ^[9]	331	15	507
5	Hellers, G ^[10]	268	7.1	397
6	Parks, AG ^[11]	255	4.5	592
7	Mizrahi, N ^[12]	223	13.9	353
8	Johnson, EK ^[13]	212	17.7	343
9	Beets-Tan, RGH ^[14]	205	12.1	350
10	Choen, S ^[15]	201	7.4	275
11	Loungnarath, R ^[16]	197	14.1	285
12	Buchanan, GN ^[17]	192	13.7	347
13	Williams, JG ^[18]	189	7	310
14	Whiteford, MH ^[19]	186	14.3	384
15	Champagne, BJ ^[20]	183	15.3	280
16	Lindsey, I ^[21]	181	11.3	287
17	Kuijpers, HC ^[22]	170	5.2	267
18	Morris, J ^[23]	169	9.4	303
19	Buchanan, GN ^[24]	167	11.1	268
20	Lunniss, PJ ^[25]	167	7	245
21	Cintron, JR ^[26]	166	9.2	238
22	Law, PJ ^[27]	166	5.7	255
23	O'Connor, L ^[28]	161	13.4	240
24	Halligan, S ^[29]	151	12.6	299
25	Schwartz, DA ^[30]	151	8.9	235
26	Sentovich, SM ^[31]	150	10	248
27	Poen, AC ^[32]	146	7.3	209
28	Lunniss, PJ ^[33]	144	6	220
29	Lunniss, PJ ^[34]	143	5.5	198
30	Graham Williams, J ^[35]	143	5.3	210
31	Aguilar, PS ^[36]	141	4.3	209
32	Hussain, SM ^[37]	140	6.4	210
33	Goldberg, SM ^[38]	133	3.2	3
34	De La Portilla, F ^[39]	131	26.2	193
35	Buchanan, G ^[40]	129	8.1	216
36	Soltani, A ^[41]	128	16	188
37	García-Aguilar J ^[42]	128	6.4	229
38	Barker, PG ^[43]	126	5.3	181
39	Cheong, DMO ^[44]	125	5	189
40	Shanwani, A ^[45]	124	15.5	237
41	Deen, KJ ^[46]	121	5	188
42	Ky, AJ ^[47]	118	11.8	169
43	Herreros, MD ^[48]	112	18.7	187
44	Makowiec, F ^[49]	112	4.9	155
45	Seow-Choen, F ^[50]	111	4.3	Unable to find
46	Tio, TL ^[51]	107	3.8	121
47	Hämäläinen, KJ ^[52]	104	5	176
48	Marks, CG ^[53]	102	2.5	196
49	Panés, J ^[54]	101	50.5	159
50	Rojanasakul, A ^[55]	101	11.2	223
51	van der Hagen, SJ ^[56]	99	8.3	134
52	Spencer, JA ^[57]	99	4.5	166
53	Williams, JG ^[58]	98	8.9	184
54	Spencer, JA ^[59]	98	4.9	145
55	Ortiz, H ^[60]	97	10.8	158
56	Marks, CG ^[61]	94	2.5	155
57	Van Koperen, PJ ^[62]	93	9.3	140
58	Buchanan, GN ^[63]	92	6.6	199
59	Patrlj, L ^[64]	90	5	125
60	García-Aguilar, J ^[65]	90	5	134
61	Faucheron, J-L ^[66]	90	4.1	136
62	Sangwan, YP ^[67]	90	3.8	132

(continued)

Table 1
(continued).

Rank	First author	Scopus	Citation rate	Google Scholar
63	Weisman, RI ^[68]	90	3.3	141
64	Milligan, ETC ^[69]	90	1.1	309
65	Ellis, CN ^[70]	88	7.3	141
66	Garcia-Olmo, D ^[71]	87	8.7	137
67	Schwartz, DA ^[72]	86	6.6	103
68	Sentovich, SM ^[73]	86	5.1	135
69	O'Riordan, JM ^[74]	85	14.2	118
70	Sahni, VA ^[75]	85	8.5	142
71	Ellis, CN ^[76]	85	7.7	128
72	Hammond, TM ^[77]	85	6.1	141
73	Christoforidis, D ^[78]	84	8.4	134
74	Van Koperen, PJ ^[79]	83	7.5	115
75	Singer, M ^[80]	83	6.4	127
76	Ardizzone, S ^[81]	83	5.9	98
77	Meinero, P ^[82]	82	11.7	147
78	Ritchie, RD ^[83]	82	9.1	144
79	Malik, AI ^[84]	82	8.2	136
80	Schwandner, O ^[85]	82	8.2	127
81	Nordgren, S ^[86]	82	3.2	125
82	Cintron, JR ^[87]	81	9	117
83	Halligan, S ^[88]	81	4.1	130
84	Myhr, GE ^[89]	81	3.4	116
85	West, RL ^[90]	80	5.3	143
86	Makowiec, F ^[91]	80	3.5	109
87	Arseneau, KO ^[92]	79	4.6	89
88	Chapple, KS ^[93]	79	4.4	119
89	Cho, D-Y ^[94]	78	4.1	132
90	Beckingham, IJ ^[95]	78	3.5	136
91	Guadalajara, H ^[96]	77	12.8	115
92	Grimaud, J ^[97]	77	9.6	111
93	Safar, B ^[98]	77	8.6	133
94	Ratto, C ^[99]	77	4.3	127
95	Kennedy, HL ^[100]	76	2.7	127
96	Lawes, DA ^[101]	73	7.3	116
97	Christoforidis, D ^[102]	71	7.9	107
98	Christensen, A ^[103]	71	2.2	114
99	Navarro-Luna, A ^[104]	66	4.7	117
100	Van Der Hagen, SJ ^[105]	65	5	89

The topics covered in the top 100 most-cited papers were wide-ranging. Surgical management was the topic with the highest number of publications in the top 100 (n=47), followed by imaging examinations (n=28). Twenty-six papers on anal fistula were related to Crohn disease, while 5 papers were dedicated to stem cells, 2 papers focused on Infliximab, 2 articles studied the etiology and pathology of perianal fistula, and 1 focused on the classification of perianal fistula.

4. Discussion

The treatment of anal fistula is difficult as recurrent or unhealed events occur, which seriously influence patient quality of life. Although surgery is the only treatment to cure perianal fistula, surgical problems such as protecting anal function, and preventing recurrence and incontinence have confused clinicians for centuries. Research on these aspects is constantly developing. Lundqvist et al^[106] carried out the first study on resource use, costs and sick leave related to anal fistulas in Sweden, and showed that anal fistula is a costly social disease, especially in patients undergoing multiple surgeries which results in a heavy social burden.

Unsurprisingly, according to the results of this bibliometric analysis, surgical management is the most discussed topic and was represented in 47 of the 100 most-cited papers. Research contents included “fistula fibrin glue” (11 articles, 1270 citations),^[15,24,26,31,64,72,73,75,82,87,97] “perianal fistula plug” (10 articles, 1031 citations),^[20,28,47,74,75,78,79,85,98,101] “seton-treatment” (6 articles, 628 citations),^[18,52,63,66,83,103] “advancement flap” (5 articles),^[12,36,41,49,62] “fistulotomy” (2 articles),^[62,100] “ligation-of-intersphincteric-fistula-tract” (2 articles),^[45,55] “video-assisted-anal-fistula-treatment” (1 article),^[82] comparison of operations (6 articles), and others (4 articles). The comparisons between operations were as follows: “perianal fistula plug” versus “fistula fibrin glue,”^[13] “fistula fibrin glue” versus “seton treatment” and “fistulotomy,”^[21] “seton treatment” versus “fistulotomy,”^[35,42] and “perianal fistula plug” versus “advancement flap.”^[60,102] Fistula fibrin glue was the most frequently studied form of surgical treatment. In the early 1980s, Kirkegaard and Madsen^[107] reported the successful closure of various fistulas with fibrin sealant. It is suitable for both cryptoglandular perianal fistula and perianal fistula with Crohn disease, and is safe, simple, and easy to perform by the surgeon. Although the reported success rates vary, should this operation fail, it can be repeated or changed to another type of sphincter retention surgery. All the studies on surgery lacked randomized controlled trials, which will be the focus of future research by clinicians.

With regard to perianal fistula with Crohn disease, this represents a greater challenge to surgeons due to poor healing, risk of incontinence, and the need for fecal diversion or proctectomy in some patients.^[108] There were 26 papers in the 100 most-cited manuscripts on this topic, 4 of these articles focusing on stem cell treatment, including 2 randomized controlled trials, which received 415 citations (Garcia-Olmo et al^[7]) and 101 citations (Panés et al^[54]), respectively. Therefore, this novel stem cell treatment of perianal fistula with Crohn disease is a hot research topic. Professor Dryden^[109] stated that Crohn disease remains a life-long disease, and mesenchymal stem cells may serve as a candidate therapy for patients who have failed to respond to biological therapy.

With regard to the topics covered in the 100 most-cited papers, imaging examinations were also well studied, with 28 articles in total, which included magnetic resonance imaging (MRI), endorectal ultrasonography (ERUS), and fistulography. Of these, MRI was the most studied. It is universally acknowledged that MRI has advantages in aspects such as accuracy, preoperative staging, and evaluation of the primary tract, location of the internal opening, and predicting postoperative recurrence. Yildirim et al^[110] conducted a study to assess the contribution of various MRI sequences, compared with readers with varying levels of experience. The results showed that there was statistically significant agreement between the readers for fistula classification, internal opening location, and the presence of sinus tracts, abscess, a horseshoe component and inflammation. ERUS has some advantages in perianal fistula staging. However, fistulography is inaccurate and unreliable, and is not recommended for the diagnosis of perianal fistula.

Scopus, Web of Science (WOS), Google Scholar (GS), and PubMed are the main databases for academic information sources. Scopus includes a broader spectrum of journals than PubMed and WOS, and its citation analysis is faster and includes more articles than the citation analysis of WOS.^[111] Moed et al^[112] demonstrated that the linear correlation between GS and

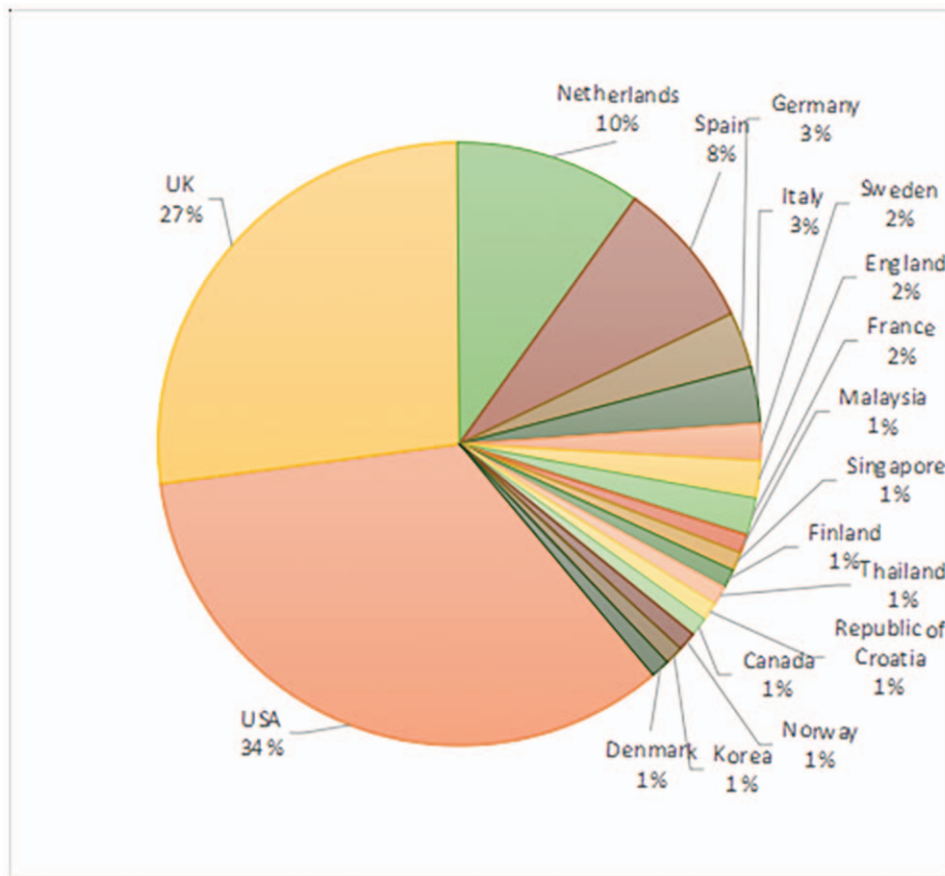


Figure 1. Countries of origin of the top 100 most-cited papers. Numbers of papers in the top 100 are shown as percentages.

Scopus citation counts at the article level is high, with Pearson correlation coefficient being in the range 0.8 to 0.9. Therefore, GS was also chosen to reflect the citation frequency of the top 100 most-cited papers listed in Scopus.

The main limitation of this study is that the Scopus database was used to search for the most-cited articles, as the number of citations is known to differ between GS and WOS. In this paper, although all studies were also evaluated based on the number of citations identified by GS, they were not sorted accordingly. Also the search strategy may not have included all articles on perianal fistula. Furthermore, as suggested by Schoonbaer and Roelants,^[113] the use of citation analysis and journal impact is controversial, due to technical limitations, database selectivity, time and discipline-related biases, language and publication-type biases, multiple authorship merits, and citing motivations. With regard to self-citation, several of the manuscripts in the top 100 are authored by multiple researchers (such as “Fibringlue is effective healing perianal fistulas in patients with Crohn Disease”^[97] which is authored by 20 coauthors), making it difficult to accurately track and calculate self-citations. Another limitation is that older manuscripts have a greater opportunity for citation than more recent manuscripts. In addition, the names of only the first and senior authors and the institution of only the first author are captured. Thus, several authors in the top 100 may in fact have contributed toward multiple papers, although in a lesser role than the first or senior authors.

5. Conclusions

The most-cited papers highlighted in the current work can be considered the classic works in the field of perianal fistula study, which describe surgical techniques, imaging examinations, basic science, drug therapy (stem cells, Infliximab), and other topics. The majority of papers were published in journals with an impact factor of less than 10. This article may serve as a reference for researchers and clinicians as to what constitutes a citable paper in this field. A few of the more recent papers now have higher rates of citation than those mentioned in this study. The topics covered in these papers can expect significant developments in the next 10 years.

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