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Bibliometric and visualized analysis of research on ossification of posterior longitudinal ligament

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Abstract

Objectives: Through the analysis of the publication years, countries, institutions, keywords, references, authors, and journals, we found the main directions and hot topics of Ossification of the posterior longitudinal ligament research.

Methods: Using the Science Citation Index Expanded (SCI E) database of the Web Of Science (WOS) Core Collection database, publications on OPLL research were retrieved from 1995 to 2019, and bibliometric methodology was used to research the source data. VOSviewer software were used to analyze the publication trend in OPLL research. Network maps were made by using VOS viewer, to discover the relationship between institutions, keywords, references, authors, and journals.

Results: 1525 literatures were retrieved in the field of OPLL by using the database of the Web Of Science (WOS). The country and institution with the largest number of publications are Japan and Hirosaki University (Japan). The most co-cited document was "Operative Results and Postoperative Progression of Ossification among Patients with Ossification of Cervical Posterior Longitudinal Ligament", authored by Hirabayashi K, published in the journal "Spine". Hirabayashi K was also the most co-cited author. The relationship between co-cited authors was also awfully close and active. The most articles about OPLL were published in SPINE, which is also the most co-cited journal.

Conclusions: Japan contributed the most publications. The overall trend of literature publication on OPLL was increasing year by year. By summarizing keywords, co-cited keywords, references, and co-cited references, it was not difficult to find the surgical method and the factors that affect the long-term postoperative effect were the hotspots of research.

Keyword: Ossification of the posterior longitudinal ligament (OPLL); Bibliometric analysis; Treatment; Laminoplasty.

Abbreviations: OPLL: Ossification of the posterior longitudinal ligament; SCI E: The Science Citation Index Expanded; WOS: Web Of Science.

Introduction

Ossification of Posterior Longitudinal Ligament (OPLL) occurs more frequently in East Asia, especially in Japan [1-4]. The incidence of cervical OPLL is estimated to be 1.9% to 4.3% in the Japanese general population [2,5-8]. OPLL is a disorder characterized by progressive ectopic ossification of the Posterior Longitudinal Ligament (PLL), with occurrence of 70% in cervical spine and 15% in thoracic vertebra as well as in lumbar vertebra [2,9,10]. OPLL is prone to 50-60 years old, and the male: female ratio is 2:1 [2,4,7,9,11,12]. The C4-C5 is most often involved in the cervical spine [2,5,13]. Pathogenesis of OPLL is a combination of many factors, include familial inheritance (genetic factors) and non-genetic factors including diet, obesity, physical strain on the posterior longitudinal ligament, age, and diabetes mellitus [8,9,12,14].

The essence of ossification of posterior longitudinal ligament is heterotopic ossification [13,15-17]. There are several classification systems to categorize patients with OPLL. The one established by The Investigation Committee for Ossification of the Spinal Ligaments (part of the Japanese Ministry of Health, Labor and Welfare) is widely accepted. Based on X-Ray, there are four radiographic types of OPLL: 1) segmental (39%), limited to the posterior surface of the vertebral bodies without crossing disc spaces; 2) continuous (27.3%), spanning multiple levels with involvement of vertebral body and disc spaces;3) mixed (26.2%), a combination of segmental and continuous; and 4) other (7.5%), limited to posterior disc spaces with some extension to the posterior vertebral body endplate [2,14,17-19]. X-ray examination of cervical spine is the first choice while 2- or 3-dimensional CT imaging is best diagnosed for OPLL [14,17]. Magnetic Resonance Imaging (MRI) is used to detect myelomalacia as a result of compression from OPLL as well as detect the extent of spinal cord compression [3]. OPLL could cause spinal canal or foraminal narrow, induce myelopathy or radiculopathy, and increase the risk of spinal cord injury after a traumatic event, while 5% of diagnosed patients are asymptomatic [3,9,12,17]. However, studies on characteristics of research on OPLL are limited and this topic needs more attention.

Our research aimed to provide all-round insights on the current state of OPLL. A comprehensive bibliometric analysis was conducted to determine the research landscape of Ossification of the Posterior Longitudinal Ligament (OPLL) in terms of publication language, document type, countries, institutions, the year, keywords, co-cited references, journals, co-cited journals and authors. In order to better understand the global trend of research and to discover the popular topics in this field, our research attempted to provide all-round insights on the current state of global Ossification of the Posterior Longitudinal Ligament (OPLL) research [20].

Methods

Data source and selection criteria

A search was performed online using the Science Citation Index Expanded (SCI E) database of the Web Of Science (WOS) Core Collection database on a single day, September 21, 2019 to avoid errors caused by daily updates. The search formula was [TS= ("ossification of posterior longitudinal ligament") OR ("ossification of the posterior longitudinal ligament") OR ("ossification of PLL") OR ("OPLL")] from 1995 to 2019. There was no restriction on document type, language or data category. Finally, the search retrieved 1525 literatures that met the inclusion criteria.

Bibliometric analysis

The network maps of institutions, keywords, co-cited references, co-cited authors and co-cited Journals were designed by VOSviewer (Leiden University, Leiden, Netherlands) to form a visual relationship map [20-23]. The hotspot map of keywords was also produced by VOSviewer, which helped to find the research hotspot in image [23-25]. In the network maps generated by VOSviewer, through cluster analysis, the elements that need to be analyzed, such as institutions, co-cited references, co-cited authors, etc., were divided into multiple nodes and classified with different colors. The size of the nodes reflected the number of publications or frequency. The links between nodes represented relationships such as collaboration, co-occurrence or co-citations [21,22,24,26]. The chart, line chart, histogram and pie chart are all made by Excel 2016. The Impact Factors (IF) of journals were obtained from the 2018 Journal Citation Reports (JCR).

Results

Publication language and document type

There are seven languages in total in 1525 literatures searched from WOS. Among the 1525 papers, 1491 were published in English, accounts for 97.7%, 11(0.72%) in French, 9(1.13%) in German, 5(0.07%) in Japanese, 5(0.07%) in Czech, and 2(0.03%) in Turkish. English is still the mainstream publishing language in the world. 1525 literatures contain 11 document types, in which articles (1313, 86.09%) are the most frequently used document type and second are reviews (100,6.56%).

Countries and institutions analysis

The search results refer to Taiwan are classified as China and those from England, Northern Ireland and Wales are classified as the United Kingdom, literatures on ossification of posterior longitudinal ligament are published by fifty-one countries. The top ten countries were Japan (623,40.85%), China (343,22.49%), USA (237, 15.54%), South Korea (158,10.36%), India (49,3.21%), Canada (43,2.82%), Germany ((24,1.57%), UK (22,1.44%), Turkey (21,1.38%), France (16,1.05%) and Italy (16,1.05%) (Table 1). The top four countries account for 89.25% of the total articles and Japan is far ahead.

The 1525 documents retrieved involved 1144 institutions. Eight of the top 10 institutions are from Japan and two are from China (Table 1). The top 10 institutions contributed 488(32.01%) articles. Among them, Japan has 398 articles, accounting for 26.11% and China has 90 articles, accounting for 5.90%, including 73(4.79%) from Hirosaki University (Japan), 56(3.67%) from Second Military Medicine University (China). In the network map of institutions, there were 134 nodes and 8789 links (Figure 1). The 134 institutions formed 4 clusters, and from the charts, there are intensive links between different organizations, especially prominent in the same cluster. Centrality analysis through charts, Hirosaki University (Japan) and Second Military Medical University (China) are the main research institutes.

Table 1: The top 10 countries and institutions contributed to publications of OPLL [n (%)].

Rank	Country	N(%)	Institute	N(%)
1	Japan	623(40.85%)	Hirosaki University (Japan)	73(4.79%)
2	China	343(22.49%)	Second Mil Med University (China)	56(3.67%)
3	USA	237(15.54%)	Tokyo University (Japan)	55(3.61%)
4	South Korea	158(10.36%)	Keio University (Japan)	53(3.48%)
5	India	49(3.21%)	Chiba University (Japan)	52(3.41%)
6	Canada	43(2.82%)	Kagoshima University (Japan)	46(3.02%)
7	Germany	24(1.57%)	Tokyo Med Dent University (Japan)	42(2.75%)
8	UK	22(1.44%)	Nagoya University (Japan)	40(2.62%)
9	Turkey	21(1.38%)	Osaka University (Japan)	37(2.43%)
10	France	16(1.05%)	Peking University (China)	34(2.23%)
11	Italy	16(1.05%)		

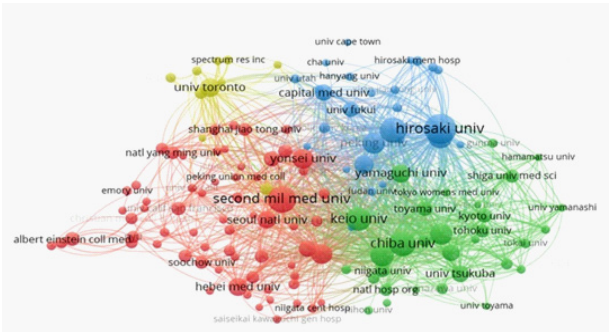

Figure 1: The network map of institutions in research of OPLL.

Publication years analysis

In total of 1525 documents are retrieved from 1995 to 2019. The number of articles published increased year by year and occasionally decreased (Figure 2). There are three stages of growth, first stage, from 1995 to 1999, with an average annual growth rate of 24.91%; second stage, from 2010 to 2013, with an average annual growth rate of 23.90%; third stage, from 2015 to 2018, with an average annual growth rate of 20.06%. From 2000 to 2009, the number of articles published annually tends to stabilize. Through trend analysis, OPLL has been paid attention to since 1995, and the research results show a growing trend.

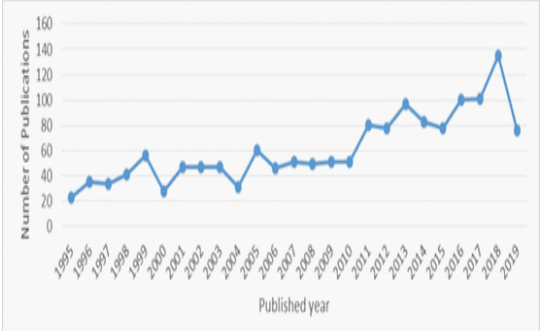

Figure 2: Number of publications of OPLL from 1995 to 2019.

Co-occurrence keywords analysis

In the 1525 articles, the top ten most common occurrences of key word are analysed. Using VOSviewer software, the network map about the keywords of OPLL are made. As was shown in Figure 3, the most commonly used keyword was “Posterior longitudinal ligament” when studying OPLL. Spondylotic myelopathy caused by OPLL is also the focus of researchers’ attention. In the treatment of OPLL, more emphasis is placed on surgical treatment and fusion. In the network map, 38 main items are divided into 3 clusters, distinguished with different colors. There are 647 links each other. The main keywords in red cluster are “Posterior longitudinal ligament” “Ossification” “Myelopathy” “Ossification of the posterior longitudinal ligament” “Spine” and so on. In green cluster, the main keywords are “Laminoplasty” “Spondylotic myelopathy”. In blue cluster, the main keywords are “Fusion” “Surgery” and “Decompression” (Figure 3).

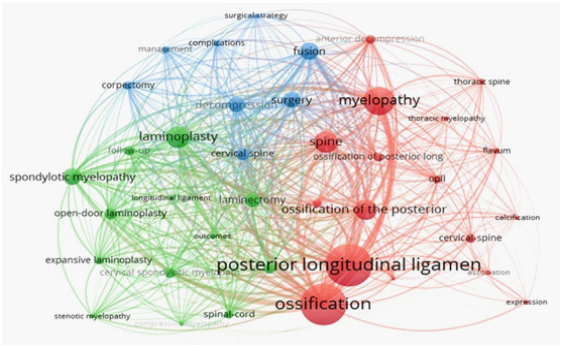

Figure 3: The network map of keywords for OPLL research..

Co-cited references analysis

The first 10 co-citations are tabulated in Table 2. The most co-cited document is “Operative Results and Postoperative Progression of Ossification Among Patients With Ossification of Cervical Posterior Longitudinal Ligament”, authored by Hirabayashi K, published in Spine. A total of 308 articles have been cited more than 20 times. Through analysis of references by VOSviewer, 308 references are divided into 4 clusters (Figure 4). There were active collaborations between the references and the clusters.

Table 2: Top 10 co-cited references for OPLL research.

Rank	Co-cited reference	Co-cition
1	Hirabayashi K, 1981, SPINE, V6, P354	223
2	Kato Y, 1998, J NEUROSURG, V89, P217	172
3	Tsuyama N, 1984, CLIN ORTHOP RELAT R, P71	159
4	Iwasaki M, 2002, J NEUROSURG, V96, P180	135
5	Hirabayashi K, 1983, SPINE, V8, P693	117
6	Iwasaki M, 2007, SPINE, V32, P647	111
7	Koga H, 1998, AM J HUM GENENT, V62, P1460	83
8	Chiba K, 2006, SPINE, V31, P2998	82
9	Ogawa Y, 2004, J NEROSURG-SPINE,V1, P168	73
10	Kawaguchi Y, 2001, J BONE SURG AM, V83a, P1798	69

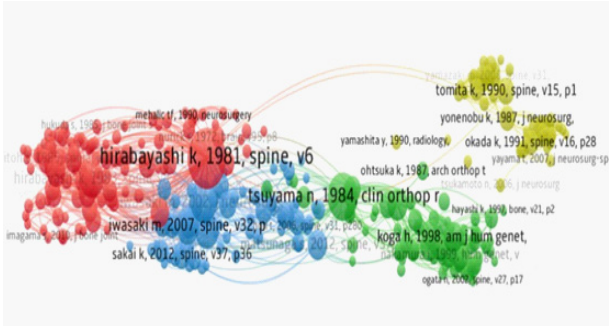

Figure 4: The network map of Co-cited references for OPLL research.

Authors and co-cited authors

There are 5070 authors in 1525 articles published, of which the top 10 are listed in Table 3. The top 10 authors are from Japan and China, both in East Asia, where OPLL is prevalent. 23% of 1525 articles published by top 10 authors. Okawa A 50(3.279%) published the most literatures, followed by Yamazaki M48(3.148%), all of which were from Japan. Three of the top 10 authors are from Second Military Medical University, located in Shanghai, China. Among the top 10 co-cited authors, Hirabayashi K has 548 citations, ranking first, followed by Matsunaga S (524 citations). 351 co-cited authors with more than 20 citations were analyzed by VOSviewer, and the co-cited authors were divided into 5 clusters (Figure 5). Cluster 1(red) has a maximum of 99 co-cited authors, including Hirabayashi K, Epstein Ne, Yonenobu K and Baba H. There maining six of the top 10 are distributed in Cluster 2(green) and Cluster 3(blue).

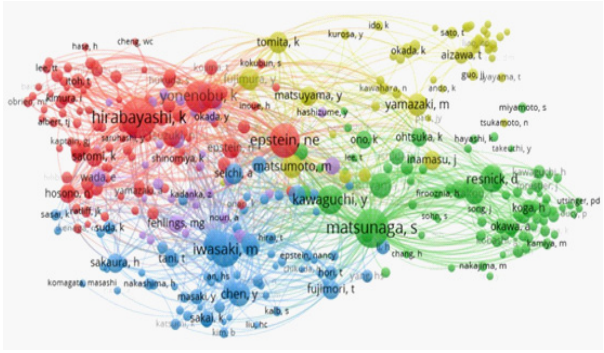

Figure 5: The network map of co-cited authors for OPLL research.

Journals and co-cited journals analysis

1525 papers have been published in 253 magazines, of which SPINE (IF=2.903) is the most published journal. Nearly half of the articles published on the top 10 journals. SPINE has the most articles, followed by European Spine Journal. However, through the analysis of VOSviewer, 143 journals have only one article on OPLL. In the top 10 co-cite journals, 7 journals also appear in the top 10 journals (Table 4). Spine is also the most co-cite journal, followed by Journal of Neurosurgery. Cluster analysis of 184 co-cited Journals cited more than 20 times showed that there were six clusters in this field (Figure 6). Cluster 1(red) has a maximum of 83 items. Cluster 2 (green) contains 42 items including Spine and Journal of Neurosurgery, ranking second.

Table 3: The top 10 author and co-cited authors contributed to publications of OPLL.

Rank	Author	N (%)	Country	Institute	Co-cited author	Citations	Country	Institute
1	Okawa A	50(3.279%)	Japan	Tokyo Med & Dent Univ	Hirabayashi K	548	Japan	Univ Miyazaki
2	Yamazaki M	48(3.148%)	Japan	Univ Tsukuba	Matsunaga S	524	Japan	Kyoto Univ
3	Kawaguchi Y	35(2.295%)	Japan	Univ Toyama	Epstein Ne	395	USA	Albert Einstein Coll Med
4	Chen DY	33(2.164%)	China	Second Mil Med Univ	Iwasaki M	390	UK	Imperial Coll London
5	Koda M	33(2.164%)	Japan	Univ Tsukuba	Yonenobu K	342	Japan	Osaka Yukioka Coll Hlth Sci
6	Yuan W	33(2.164%)	China	Second Mil Med Univ	Kawaguchi Y	285	Japan	Univ Tokyo
7	Toyama Y	32(2.098%)	Japan	Toshiba Co Ltd	Baba H	226	Germany	Univ Duisburg Essen
8	Chen Y	31(2.033%)	China	Second Mil Med Univ	Chiba K	214	Japan	Yokohama Univ Pharm
9	Harata S	30(1.967%)	Japan	Hokkaido Univ	Tsuyama N	213	Japan	Japanese Fdn Canc Res
10	Matsumoto M	29(1.902%)	Japan	Kagoshima Univ	Chen Y	212	China	Second Mil Med Univ

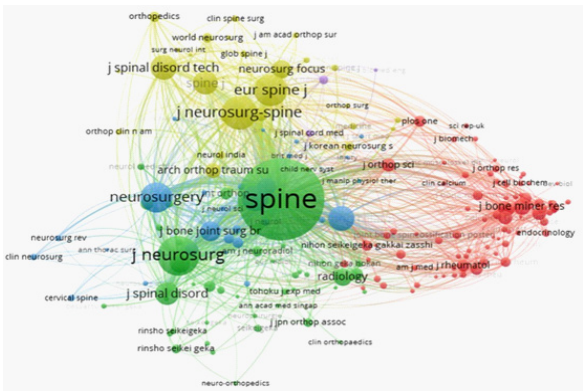

Figure 6: The network map of co-cited Journals for OPLL research.

Table 4: The top 10 Journal and Co-cited Journal contributed to publications of OPLL.

Rank	Journal Abbreviation	N	Citations	Country	IF2018	Co-cited Journal	Co-Citations	Country	IF2018
1	SPINE	218	7323	USA	2.903	SPINE	8945	USA	2.903
2	EUR SPINE J	101	1596	USA	2.513	J NEUROSURG	2102	USA	4.13
3	J NEUROSURG-SPINE	79	1930	USA	2.998	J SPINAL DISORD TECH	1550	USA	2.31(2017)
4	WORLD NEUROSURG	58	190	USA	1.723	J NEUROSURG-SPINE	1525	USA	2.998
5	J SPINAL DISORD TECH	51	1072	USA	2.31(2017)	EUR SPINE J	1335	USA	2.513
6	NEUROSURGERY	48	957	USA	4.605	NEUROSURGERY	1228	USA	4.605
7	J CLIN NEUROSCI	46	348	UK	1.593	J BONE JOINT SURG AM	897	USA	4.716
8	SPINE J	42	579	USA	3.196	CLIN ORTHOP RELAT R	845	USA	4.154
9	SPINAL CORD	34	492	UK	1.898	SPINE J	729	USA	3.196
10	NEUROSURG FOCUS	29	539	USA	2.891	J BONE JOINT SURG BR	525	UK	3.309(2014)
10	J NEUROSURG	29	1695	USA	4.13				

Discussion

In this study, 1525 literatures about OPLL were retrieved from the WOS database, which involved 1144 institutions in 51 countries. The top fourth countries are Japan, China, USA and South Korea, three of which are in East Asia. Key CA [27] first reported OPLL in 1838, and then Tsukimoto H [28] elaborated in 1960. Due to its prevalence in East Asia, especially in Japan, it provides a large number of case samples for OPLL research. Japan was the first country to report OPLL and conduct ongoing research. It is in the leading position in OPLL research field. Japan accounts for almost half of the total articles, and eight of the top 10 research institutions are from Japan. Among the top four countries, only China is a developing country and among the top 10 institutions, 2 universities are shortlisted. It shows China's strong interest and scientific research strength in OPLL field.

Analysis of data from 1995 to 2019 by VOSviewer, a total of 3644 keywords are found. Of the 1,525 literatures, 2,427 keywords appeared only once, accounting for 66.6%. Screening out the top 10 keywords for analysis, it is not difficult to find that most of the keywords about OPLL treatments, such as "Laminoplasty", "Fusion", "Surgery" and "Decompression". In the density map of the keyword, the hotspot keywords located in red and yellow are mostly related to surgical treatment. It is not difficult to find through analysis that the surgical treatment of OPLL is a hot trend at this stage. For patients with neurological symptoms such as myelopathy or radiculopathy, surgical intervention is effective. According to the different approaches, it is mainly divided into anterior, posterior, and combined anteroposterior approaches [12]. Laminoplasty is an important surgical procedure for the posterior approach [13]. Kirtia first proposed laminectomy in 1968, through the posterior decompression of the spinal canal [29,30]. Laminoplasty is an improvement of Kirtia's technique for laminectomy, to avoid problems with laminectomy such as postoperative segmental instability, kyphosis, perineural, adhesions and late neurological deterioration [13,29-31]. Hirabayashi Kproposed expansive open-door laminoplasty for ossification of the posterior longitudinal ligament of the cervical spine in 1977 published in Japanese and expansive open-door laminoplasty was described in detail again in Spine [13,19,30,31]. As another important development of laminoplasty, spinous process splitting double-door laminoplasty is described by Kurokawa in 1982 [29,31]. These two surgical methods have far-reaching effects, and subsequent modifications are mostly based on Hirabayashi K' expansive open-door laminoplasty or Kurokawa's double-door laminoplasty [29,31,32].

By analyzing the top 10 co-cited references, 7 of all are about expansive open-door laminoplasty and long-term follow-up results, 1 for review about OPLL, 1 for long-term follow-up results of laminectomy, and 1 for genetic mapping of OPLL. Obviously, long-term follow-up results and postoperative process after Hirabayashi' expansive open-door laminoplasty has received much attention and has been studied in depth. In the most co-cited literature, Hirabayashi studied 53 patients with OPLL and relied on the JOA score to get an overall recovery rate of 70%. The calculation formula of recovery rate: Recovery rate (%) =[postoperative JOA score-preoperative JOA score]/[17-preoperative JOA score] /100 was proposed by Hirabayashi to evaluate the postoperative effect [19,33]. Good operative results could not be obtained in patients over 60, those with myelopathy for more than two years, those with severe (more than 60%) stenosis of the spinal canal, or those operated on by inadequate surgical methods [19,34]. Hirabayashi also found that postoperative progressions were noted in 75% of the patients with continuous or mixed-type OPLL [14,19]. Iwasaki confirmed that the most significant predictors of postoperative neurological improvement were a higher preoperative JOA score and younger age at operation [14]. Early surgical treatment for OPLL would be advisable [35]. These were consistent with Kato's 1998 research of the postoperative neurological recovery of laminectomy [33]. In 2007, Iwasaki found that the most significant predictor of poor outcome after laminoplasty was hill-shaped ossification followed by lower preoperative JOA score by multiple regression analysis of another group of patients [34,36]. Further research, Iwasaki and Ogawa found that cervical kyphosis is not a contraindication to laminoplasty [13,35,37]. Postoperative complications of laminoplasty, for example, segmental motor paralysis, kyphosis, whether it has established before surgery or it has developed after surgery, and progression of OPLL, are also the focus of research [32,37].

In the analysis of authors and co-cited authors, it is found that scholars from Japan have in-depth research on OPLL and have worked closely with academics in the professional field. Cluster analysis shows that the co-cited authors are divided into 5 clusters, 31134 links and there are intensive contacts and cooperation among scholars in the field of OPLL. Seven of the top 10 authors are from Japan and six of the top 10 co-cited authors are from Japan. Japan is in a leading position in OPLL research, mainly due to the long history of OPLL research, the number of disease sources caused by regional differences in incidence,

and the consistency and continuity of research. The most articles about OPLL are published in SPINE, which is also the most co-cited journal. The research on the top 10 and co-cited top 10 journals found that the vast majority of journals from the United States have a profound international influence.

As time has passed, some new frontiers attracted attention. A majority of researches were related to the diagnosis and treatment of OPLL while many publications also emphasized the surgical treatment. According to the bibliometric analysis, researches on best operation way for OPLL and myelopathy caused by OPLL between are promising areas.

Conclusion

In research of OPLL, Japan is far ahead, with 623 literatures, accounting for 40.85%, followed by China, the United States, and South Korea. Eight of the top 10 research institutions are from Japan, with Hirosaki University at the top of the list. Seven of the top 10 authors contributed to publications of OPLL are also from Japan. Okawa A is the author who published the most articles on OPLL from 1995 to 2019. Hirabayashi K is the top 1 co-cited author who first proposed expansive open-door laminoplasty and became the typical posterior approach operation of OPLL, which is of great significance and far-reaching influence. The top two productive journals are SPINE and EUROPEAN SPINE JOURNAL. SPINE and JOURNAL OF NEUROSURGERY are the top two co-cited journals. By summarizing keywords, co-cited keywords, references, and co-cited references, it is not difficult to find the surgical method and the factors that affect the long-term postoperative effect are the hotspots of research.

Declarations

Data availability statement: The datasets generated during and analyzed during the current study are available from the corresponding author on reasonable request.

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Author contributions: DS was responsible for gathering and analyzing the data and drafted the paper.

JS and CS helped to conceived and designed the study. PN ang YJ helped to revise the manuscript. HZ and ZZ helped to design the study, draft and revise the manuscript, and approve the final manuscript.

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Ethical approval: All procedures used in this research were approved by the Ethics Committee of the Army Military Medical University.

Competing interests: The authors declare no competing interests.

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