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# Blastic Plasmacytoid Dendritic Cell Neoplasm: A 10-year Bibliometric Study

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## Abstract:

**INTRODUCTION:** Blastic plasmacytoid dendritic cell neoplasm (BPDCN) is a rare hematological disease. There is an incidence. We aimed to evaluate the scientometrics characteristics of the scientific production on BPDCN between 2011 and 2020.

**METHODOLOGY:** A cross-sectional and retrospective bibliometric study was performed. The search was executed in the Scopus database. Data were extracted based on a formula developed using thesaurus MeSH (Medline) and Emtree (Embase) terms. The retrieved papers received 11.2 citations per paper. Four of the ten most productive authors were from the United States. The institution with the highest impact (1064 citations) and the highest scientific output (46 papers) was the University of Texas MD Anderson Cancer Center (United States).

**RESULTS:** The journal "Movement Disorders" rated first with 18 articles and 643 citations in terms of productivity and impact, and articles published in Q1 journals surpassed the remaining quartiles. The most cited articles had national collaboration. Conclusion the number of papers on BPDCN has been rising, yet journals with a higher quality tend to keep the similar publication rates, although they have shown a slight increase in the recent years.

**CONCLUSIONS:** High-income countries' international collaboration is crucial for increasing publications impact; hence, greater collaborations between researchers and institutions from countries around the world are needed to expand knowledge on this subject.

## Keywords:

Bibliometric analysis, Blastic plasmacytoid dendritic cell neoplasm, cancer

## Introduction

Blastic plasmacytoid dendritic cell neoplasm (BPDCN) is a rare hematological disease with an incidence of about 0.04/100,000 inhabitants worldwide, and it usually occurs in elderly people.<sup>[1]</sup> The main clinical manifestations are cutaneous involvement, either through plaques and nodules or through violaceous skin lesions; it can also develop spinal cord infiltration or in some cases extramedullary infiltration.<sup>[2,3]</sup> Although it has been related with other hematologic neoplasms such as myelodysplastic syndrome, its etiology has not yet been confirmed.<sup>[4]</sup>

The BPDCN diagnosis is based on immunohistochemistry, with a positive result for four of the five main surface markers: CD4, CD56, CD123, TCL1, and CD303.<sup>[5]</sup> Although there is no standardized treatment, chemotherapy with high-dose methotrexate with asparaginase, acute myeloid leukemia (AML), and acute lymphoid leukemia type showed improved survival and remission over other treatments.<sup>[3]</sup>

The scientific contribution of researchers, institutions, a specific region, or a certain field has been measured using bibliometric analysis. Furthermore, it can allow research to be oriented toward previously

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unaddressed areas.<sup>[6-9]</sup> In addition, they have also been used to determine the citations influence between journals and to study the collaborative contribution in an multidisciplinary subject.<sup>[9-12]</sup> Although no previous bibliometric studies of BPDCN have been developed, previous bibliometric analyses of other hematologic malignancies have been reported. On AML, it was reported that between 1999 and 2018, the genetic domain was one of the most studies.<sup>[13]</sup> In addition, regarding the hematological tumor microenvironment, immunotherapy was reported to be the current approach with the most investigations.<sup>[14]</sup> The results of this study could be useful for future research on BPDCN, not only identifying which areas are the most investigated but also to identify potential collaborators and journals. In addition, assessing trends in BPDCN research may promote the development of future advances in the field of treatments or new diagnostic methods.

Therefore, the purpose of this study was to evaluate through a bibliometric analysis of the trends, the scars and impact of publications related to BPDCN between 2011 and 2020.

## Methodology

Manuscript metadata were downloaded from Scopus. It was decided to work with this database because it is one of the most prestigious and important at the international and multidisciplinary level.<sup>[15]</sup> The SciVal software was selected to complete the bibliometric analyses and to calculate the different metrics.

The MeSH terms in PubMed and the Emtree terms in Embase were used. With the help of the Boolean operators "AND" and "OR," the advanced search strategy was established: TITLE-ABS-KEY ("BPDCN" OR "Monomorphic NK-cell lymphoma" OR "Lymphoblastoid variant of NK-cell lymphoma" OR ("blastic\*" AND "agranul\*") OR "hematodermic neoplasm" OR "BPDCN").

For the sake of accuracy, the search was limited to journals in the subject area "Medicine." Letters to the editor, articles in press, and errata were excluded. The study period limit was from 2011 to 2020.

## Statistical analysis

The search was carried out on September 19, 2021. The data were saved from Scopus in CSV format and then exported to the SciVal tool. The most productive journals and authors publishing scientific articles on BPDCN, number of articles, number of institutions, collaboration, citations per publication, and CiteScore were analyzed.<sup>[16]</sup> Finally, VOSviewer (version 1.6.10, Leiden University, The Netherlands) was selected to evaluate the co-occurrence and co-citation collaborative networks.

## Results

A total of 486 articles were selected from SciVal: a total of 2662 authors, 5463 citations, and a mean of 11.2 citations per article. Most of the selected papers were published in these main subcategories: hematology ( $n = 203$ ; 41.8%), oncology ( $n = 135$ ; 27.8%), dermatology ( $n = 81$ ; 16.7%), pathology and forensic medicine ( $n = 68$ ; 14.0%), and general medicine ( $n = 63$ ; 13.0%).

### Top 10 most productive authors

Table 1 shows the most productive authors on the topic of BPDCN. Pemmaraju with the affiliation University of Texas MD Anderson Cancer Center had the highest number of published manuscripts<sup>[17]</sup> together with the highest number of citations (840), followed by author Lane with 16 published studies.

The principal institutions with the highest number of articles are shown in Table 2. The University of Texas MD Anderson Cancer Center (USA) was the institution with the highest academic production (46) and 23.1 citations per paper. Finally, the following institutions with more academic production were the Dana-Farber Cancer Institute (USA) and Harvard University (USA), respectively.

**Table 1: Top 10 authors publishing on Blastic plasmacytoid dendritic cell neoplasm**

Rank	Authors	Documents, $n$ (%)	Total citation	Citations per document	H-index	Field-weighted citation impact	Country
1	Pemmaraju, Naveen	25 (5.1)	840	33.6	39	4.0	
2	Lane, Andrew	16 (3.3)	333	20.8	25	3.6	
3	Khoury, Joseph	12 (2.5)	192	16.0	36	1.5	
4	Petrella, Tony	12 (2.5)	321	26.8	45	1.9	
5	Facchetti, Fabio	12 (2.5)	415	34.6	67	1.6	
6	Konopleva, Marina	11 (2.3)	610	55.5	89	6.4	
7	Medeiros, Leonard	10 (2.1)	253	25.3	86	2.1	
8	Pileri, Stefano	10 (2.1)	386	38.6	90	1.7	
9	Garnache-Ottou, Francine	10 (2.1)	293	29.3	22	1.4	
10	Angelot-Delettre, Fanny	9 (1.9)	292	32.4	9	1.5	

Table 3 shows the main journals with the highest number of BPDCN publications. The top three most productive journals were Blood, Annals of Hematology and Haematologica with an average of 18, 13 and 13 articles respectively. However, in terms of the highest citation, only the former keeps its place (643). The American Journal of Hematology obtained a great number of citations citations/paper.

Furthermore, in accordance with the CiteScore, Table 4 shows the amount of articles by journal quartiles published between 2011 and 2020. The high

concentration of the first quartile manuscripts suggests the high research quality.

Table 5 shows the collaboration type with bibliometric indicators. The majority of the articles had only institutional collaboration ( $n = 201$ ; 42.0%), followed by only national collaboration ( $n = 187$ ; 39.0%) and international collaboration ( $n = 61$ ; 12.7%). However, international collaboration (1641; 26.9 citations) exceeds both national (2397; 12.8) and institutional (1326; 6.6) in terms of impact. The remaining papers belong to the "single authorship" or "no collaboration" category ( $n = 30$ ; 6.3%).

**Table 2: Top 10 institutions on Blastic plasmacytoid dendritic cell neoplasm**

Institution	Documents, n (%)	Total citation	Authors	Citations per document	Field-weighted citation impact
University of Texas MD Anderson Cancer Center (United States)	46 (9.5)	1064	100	23.1	2.6
Dana-Farber Cancer Institute (United States)	19 (3.9)	356	29	18.7	3.2
Harvard University (United States)	17 (3.5)	279	57	16.4	1.5
Institut national de la santé et de la recherche médicale (France)	15 (3.1)	443	93	29.5	1.9
University of South Florida (United States)	14 (2.9)	311	21	22.2	2.9
Université de Paris (France)	13 (2.7)	469	18	36.1	2.2
Assistance publique-Hôpitaux de Paris (France)	12 (2.5)	381	33	31.8	2.1
Etablissement français du sang (France)	12 (2.5)	434	40	36.2	1.8
University of Brescia (Italy)	12 (2.5)	415	8	34.6	1.6
Stanford University (United States)	11 (2.3)	237	26	21.5	1.3

MD: Medical

**Table 3: Bibliometric indicators of impact and production on Blastic plasmacytoid dendritic cell neoplasm**

Journals	Quartile	SCImago Journal Rank	Documents	Citations	Citations per document	CiteScore 2020
Blood	Q1	5.5	18	643	35.7	18.5
Annals of Hematology	Q1	1.1	13	110	8.5	4.4
Haematologica	Q1	2.8	13	305	23.5	8.9
Hematology/Oncology Clinics of North America	Q2	1.1	13	22	1.7	5.3
British Journal of Haematology	Q1	1.9	10	232	23.2	7.4
American Journal of Hematology	Q1	2.5	9	322	35.8	10.3
Journal of Leukemia and Lymphoma	Q4	0.1	8	8	1.0	0.4
Leukemia and Lymphoma	Q2	1.0	7	44	6.3	4.1
Leukemia Research	Q2	0.9	7	157	22.4	4.7
Turkish Journal of Hematology	Q3	0.4	7	10	1.4	1.8

**Table 4: Documents published according to CiteScore quartile on blastic plasmacytoid dendritic cell neoplasm**

CiteScore quartile	2011	2012	2013	2014	2015	2016	2017	2018	2019	2020	Total
Q1	10	10	17	17	13	21	17	19	23	40	187
Q2	7	15	10	8	6	11	16	18	13	17	122
Q3	3	7	3	9	9	7	12	4	13	8	75
Q4	4	8	11	8	9	3	9	8	12	10	82
Total	24	40	41	42	37	42	54	49	61	75	406

**Table 5: Type of collaboration on Blastic plasmacytoid dendritic cell neoplasm**

Collaboration	Percentage	Documents	Citations	Citations per document	Field-weighted citation impact
International	12.7	61	1641	26.9	1.6
Only national	39.0	187	2397	12.8	1.1
Only institutional	42.0	201	1326	6.6	0.7
Single authorship (no collaboration)	6.3	30	99	3.3	0.7

## Discussion

In the recent years, there has been a significant augmentation in the number of publications related to BPDCN. However, this is the first worldwide bibliometric analysis on this field. This research shows that the study impact is mainly conveyed through certain indicators of collaboration, impact, and quantity or production. Bibliometric analyses allow a broad evaluation of research, especially in specialized fields of knowledge.<sup>[18,19]</sup> Scopus is a database that allows this broad analysis, since it has an extensive repertoire of tools for the description of citations and authors, and in turn, has a large number of papers and references compared to other databases such as the Web of Science, Dimensions, or Crossref, thus providing a broader perspective.<sup>[20]</sup>

The subcategories with the highest number of articles are hematology and oncology, which agrees with the previous result of Zhong *et al.* in a bibliometric analysis of a topic similar to BPDCN, reported that the topics with the highest number of publications are hematology, oncology, and immunology.<sup>[21]</sup>

The author with more articles and citations than any other in BPDCN was Pemmaraju and among his most recent articles he has published are reviews regarding treatment for this neoplasm.<sup>[22,23]</sup> Although no previous bibliometric studies on BPDCN have been identified, Noor *et al.* mentioned the influence of social networks<sup>[24]</sup> on the dissemination of publications in oncology and hematology.<sup>[17,25]</sup> The main reason is because the author has recently published many reviews on BPDCN therapies in the first quartile (Q1) journals, which would increase their possibility of being cited. Regarding the country of origin, half of the 10 authors with the highest academic production are from the United States. This could be explained by the fact that previous bibliometric studies of other hematologic malignancies<sup>[13]</sup> or myeloma<sup>[26]</sup> have reported many articles from that country.

Regarding the top 10 productive institutions, the US institutions reported the highest productivity. The institution with the highest frequency of citations, publications, and authors was the University of Texas MD Anderson Cancer Center. This result was also reported on bibliometric research by Iqbal *et al.* They found that this institution was the most productive in the immunotherapy childhood leukemia field, which is part of a similar line of research to BPDCN. In addition, it should be mentioned that the Establishment français du sang in France was the institution with more citations per paper than any other.

The journal "Blood" has the highest number of documents (18) and citations (643). This is consistent

with the results of other bibliometric study on acute myeloid leukemia which reported the highest number of publications and h-index.<sup>[13]</sup> It can also be added that in another bibliometric analysis both Blood and the British Journal of Haematology, reported having had greater publication regarding immunotherapy in childhood leukemia.<sup>[21]</sup> This has been an area quite studied for BPDCN, being this latter a subtype of leukemia,<sup>[27]</sup> and having reported cases of occurrence in pediatric population.<sup>[28]</sup> Furthermore, half of the articles were presented in Q1 journals. This confirmed the scientific community interest toward BPDCN. Regarding the type of collaboration, institutional-only publications predominated over the rest, and the highest was the amount of citations with the national collaboration. However, the international collaboration had a greater impact on BPDCN research. The international collaboration should be encouraged to address new research horizons and identify new priorities, as well as to exchange relevant information.

Regarding the limitations and strengths of this study: first, the inclusion of only the most recent principal publications (2011–2020), which represents more than 80% of all articles on Scopus; second, articles published in journals not indexed in Scopus may have been omitted which does not reflect the total of articles on the area; although it should be clarified that Scopus presents a strict peer review process when including a journal, this to ensure high quality for research.<sup>[20,29]</sup> Nevertheless, this is the first bibliometric study on BPDCN.

## Conclusions

The number of papers on BPDCN has been increasing in the last decade, especially for high-quality journals. The national and institutional collaborations are important for the publications impact. Researchers and institutions from around the world need to joint efforts to ensure networking and future research on this topic.

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## Conflicts of interest

There are no conflicts of interest.

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