https://doi.org/10.1590/0004-282X20200016

VIEW AND REVIEW

Deep brain stimulation treatment in dystonia: a bibliometric analysis

Estimulação cerebral profunda no tratamento de distonia: uma análise bibliométrica

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ABSTRACT

Background: Dystonia is a heterogeneous disorder that, when refractory to medical treatment, may have a favorable response to deep brain stimulation (DBS). A practical way to have an overview of a research domain is through a bibliometric analysis, as it makes it more accessible for researchers and others outside the field to have an idea of its directions and needs. **Objective:** To analyze the 100 most cited articles in the use of DBS for dystonia treatment in the last 30 years. **Methods:** The research protocol was performed in June 2019 in Elsevier's Scopus database, by retrieving the most cited articles regarding DBS in dystonia. We analyzed authors, year of publication, country, affiliation, and targets of DBS. **Results:** Articles are mainly published in *Movement Disorders* (19%), *Journal of Neurosurgery* (9%), and *Neurology* (9%). European countries offer significant contributions (57% of our sample). France (192.5 citations/paper) and Germany (144.1 citations/paper) have the highest citation rates of all countries. The United States contributes with 31% of the articles, with 129.8 citations/paper. The publications are focused on *General outcomes* (46%), followed by *Long-term outcomes* (12.5%), and *Complications* (11%), and the leading type of dystonia researched is *idiopathic or inherited*, *isolated*, *segmental or generalized dystonia*, with 27% of articles and 204.3 citations/paper. **Conclusions:** DBS in dystonia research is mainly published in a handful of scientific journals and focused on the outcomes of the surgery in idiopathic or inherited, isolated, segmental or generalized dystonia, and with *globus pallidus internus* as the main DBS target.

Keywords: Bibliometrics; Deep Brain Stimulation; Dystonia.

RESUMO

Introdução: A distonia é uma doença heterogênea que, quando refratária ao tratamento medicamentoso, pode ter uma resposta favorável à estimulação encefálica profunda (EEP). Uma forma prática de ter uma visão desta área de pesquisa é por meio de análise bibliométrica, pois permite aos pesquisadores e terceiros a terem uma ideia das tendências e necessidades da área. **Objetivo:** Analisar os 100 artigos mais citados no tratamento de distonia pelo uso de EEP nos últimos 30 anos. **Métodos:** O protocolo de pesquisa foi realizado em junho de 2019 através da base de dados Scopus da Elsevier, em que se obteve os artigos mais citados na área de tratamento de distonia com EEP. Analisaram-se variáveis como autores, ano de publicação, país, afiliação, e alvos de EEP. **Resultados:** Os artigos foram principalmente publicados principalmente na *Movement Disorders* (19%), no *Journal of Neurosurgery* (9%), e na *Neurology* (9%). Os países europeus oferecem contribuições significativas (57% da amostra). A França (192,5 citações/artigo) e a Alemanha (144,1 citações/artigo) possuem as mais altas taxas de citações dentre todos os países. Os Estados Unidos contribuem com 31% dos artigos da amostra (129,8 citações/artigo). As publicações focaram em *Desfechos gerais* (46%), seguido de *Desfechos a longo prazo* (12,5%), e *Complicações* (11%). O principal tipo de distonia pesquisado foi distonia *generalizada ou segmentar, idiopática ou hereditária, isolada*, abrangendo 27% dos artigos e 204,3 citações/artigo. **Conclusões:** A pesquisa de EEP em distonia é publicada em seletos periódicos científicos e foca nos desfechos da cirurgia, nas distonias generalizadas ou segmentares, idiopáticas ou hereditárias, isoladas, sendo o *globus pallidus internus* o principal alvo da EEP.

Palavras-chave: Bibliometria; Estimulação Encefálica Profunda; Distonia.

INTRODUCTION

Dystonia is a heterogeneous movement disorder characterized by sustained or intermittent muscle contractions leading to abnormal movements and postures[1]. It can be classified by its clinical characteristics, including body distribution (focal, segmental, multifocal, generalized, or hemidystonia) and associated features (isolated or combined); and etiology (idiopathic, inherited or acquired)[1]. The treatment of dystonia is aimed at providing symptomatic relief for the

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Conflict of interest: There is no conflict of interest to declare.

Received on November 15, 2019; Received in its final form on December 30, 2019; Accepted on January 27, 2020.

motor symptoms, improving pain, and avoiding musculoskeletal complications such as joint contractures[2]. Medical treatment is usually limited to the side effects and has small symptomatic relief[3]. Botulinum toxin is a good option for focal dystonia; however, in generalized and segmental dystonia, it may have a limited effect due to its dose limits.

In general, most dystonic patients are selected for surgical treatment when the pharmacological treatment has revealed inadequate response; therefore, an individualized approach is the norm. Ablative procedures were largely performed in dystonia, before the advent of deep brain stimulation (DBS). Nowadays, pallidotomy is usually an option, for instance in acquired static dystonia and in status dystonicus. DBS should be considered in the inherited or idiopathic generalized dystonias that do not have reasonable symptomatic control with medication and in which disabilities impact patient's QoL. In inherited or idiopathic segmental and focal dystonias and other acquired forms like tardive dyskinesia or cerebral palsy, it should be considered when refractory to pharmacological treatments[2]. DBS, usually targeting the *globus pallidus internus* (GPi), has a response in idiopathic or inherited isolated segmental or generalized dystonia that varies between 43–65%[2]. GPi is the most common target; however, initially, thalamic targets were used[2]. Recently, the *subthalamic nucleus* (STN) is also being considered a viable target[4,5].

A practical way to identify which are the most influential authors, journals, and countries in a particular field is through a bibliometric analysis[6]. It makes it more accessible for researchers and others outside the field to have an overview of its directions and needs[7]. The literature does present bibliometric analysis on various themes, such as neurocritical care[6], back pain[8], essential tremor[7], Parkinson's disease[9], and deep brain stimulation[10].

We analyzed the 100 most cited articles on the use of DBS for dystonia treatment in the last 30 years. We evaluated authors' information, their affiliation, and the country of the corresponding author. Additionally, we investigated which were the most cited journals and their impact factors, the used DBS targets (when applicable), and the dystonia classification (also, when applicable). Articles were divided into primary or secondary (i.e., reviews and guidelines) articles.

METHODS

Search protocol

The used database for article selection was Elsevier's Scopus, and the search protocol was performed in June 2019. The exact input was *TITLE ("dystonia" AND ("DBS" OR "Deep brain stimulation")) OR ABS ("dystonia" AND ("DBS" OR "Deep brain stimulation")) AND PUBYEAR>1988*. This terminology translated to publications which possessed the terms *dystonia* and *DBS* or *deep brain stimulation* in either the article's title or abstract and have been published in the last 30 years (i.e., since 1989). After screening the search results based on pertinence, the 100 most cited were used in this study. Impact factor (IF) data of journals were retrieved from InCites Journal Citation Reports (Clarivate Analytics), and both 2017 IF and 5-year IF were collected. Lastly, the *h* index of authors was obtained from Scopus, as well.

Bibliometric analysis

After selecting the 100 most cited articles, we obtained additional information regarding these publications within our sample. At first, all the authors, year of publication, journal and its impact factor, country, and affiliation of the corresponding author were retrieved. All articles were also categorized, when mentioned, regarding the targets of DBS. The publications were divided into primary researches (i.e., original articles) and secondary researches, such as reviews, and guidelines; the classification of dystonia, if applicable; and into specific themes, such as complications, outcomes, pathophysiology, physiology, surgical approach, targets, ethics, types of stimulation and treatment. Categorized articles in *outcomes* were further classified into general aspects, long-term outcomes, dystonic tremor outcomes, and predictors of outcome.

RESULTS

General results

Our search led to 337 hits ranging from zero to 679 citations (mean of 30.2). The 100 most cited articles have a mean of 130.4 citations (70–679), and the top 10, a mean of 311.2 (203–679), which have been detailed in Table 1. The most cited article was from Vidailhet et al.[11].

Journals and impact factor

Movement Disorders (2017 IF=8.324) accounts for 19 of the articles in our sample, summing up 2071 citations, and 12 articles were from primary research (1,262 citations). *New England Journal of Medicine* (2017 IF=79.260) was the second most cited journal while having only two articles in the top 100, both of them from primary research; they were cited 1276 times. The *Journal of Neurosurgery* was the third most cited journal, with nine publications, all of them from primary research. This material accounted for 1,247 citations.

Country and affiliation

All of the corresponding author countries in our samples were from the Northern Hemisphere. They were divided primarily in Europe and North America, but also in Asia. "DBS in dystonia" research is highly prevalent in Europe, which accounted for 57 articles in our sample. Germany, France, and the United Kingdom display a large production of material. Germany accounted for 25 articles (3,203 citations), 18 from primary research (2,438 citations). France summed up to 15 articles (2,647 citations), 12 from primary research (2,309). Moreover, the United Kingdom had 10 articles (1,086 citations), in which eight were from primary research. The United States was another country in which publications were majorly present. The country has the most articles than any other in the sample: 31 articles (3,834 citations), 19 from primary research (2,466 citations).

The most prolific affiliations are the University of Sorbonne (France), the Kiel University (Germany), and the University of Montpellier (France). The three institutions account for 16% of the 100 most cited articles in "DBS in dystonia". Both University of Sorbonne and Kiel University display five articles, although the first has four primary research publications, and the latter two. However, the total citations from the University of Sorbonne are 1,300 (1,221 from primary research), and the Kiel University 1,036 (696 from primary research). The University of Montpellier has six articles, all of them from primary research, summing up 789 citations.

Year

Although we researched articles since 1989, the most cited ones were only included from 1999 on. After then, all years, until 2014, had highly cited publications, with a peak from 2005–2007, in which 33% of our sample's articles reside, summing up to 39% of total citations.

Authors

The three most cited *first* authors were Vidailhet (*h* index of 66), Kupsch (*h* index of 63), and Volkmann (*h* index of 61), as seen in Table 2. The three altogether represent 18% of all citations in our sample. Vidailhet displayed four articles (1,222 citations), three from primary research (1,143 citations). Kupsch has a single highly cited, primary research article with 597 citations. Lastly, Volkmann had four articles (547 citations), two of which were from primary research (256 citations).

Another analysis was performed according to any placement of authors during publications. In this analysis, the most cited authors were Pollak (*h* index of 83), Volkmann (*h* index of 61), and Benabid (*h* index of 88). Pollak appeared in seven articles (1,646 citations), six from primary research (1,520 citations). Volkmann was included in nine publications (1,625 citations), five of which are primary research articles (1,334 citations). Benabid was in six articles (1,568 citations), five of which were from primary research (1,442 citations).

Table 1. Top ten cited publications in deep brain stimulation in dystonia.

First author	Title	Year	Journal	Country	Citations
Vidailhet[11]	Bilateral deep-brain stimulation of the globus pallidus in primary generalized dystonia	2005	New England Journal of Medicine	France	679
Kupsch[12]	Pallidal deep-brain stimulation in primary generalized or segmental dystonia	2006	New England Journal of Medicine	Germany	597
Coubes[13]	Electrical stimulation of the globus pallidus internus in patients with primary generalized dystonia: Long-term results	2004	Journal of Neurosurgery	France	287
Vidailhet[14]	Bilateral, pallidal, deep-brain stimulation in primary generalised dystonia: a prospective 3-year follow-up study	2007	Lancet Neurology	France	266
Vercueil[15]	Deep brain stimulation in the treatment of severe dystonia	2001	Journal of Neurology	France	227
Kumar[16]	Globus pallidus deep brain stimulation for generalized dystonia: Clinical and pet investigation	1999	Neurology	USA	220
Albanese[17]	EFNS guidelines on diagnosis and treatment of primary dystonias	2011	European Journal of Neurology	Italy	214
Volkmann[18]	Introduction to the programming of deep brain stimulators	2002	Movement Disorders	Germany	213
Okun[19]	Management of referred deep brain stimulation failures: A retrospective analysis from 2 Movement Disorders Centers	2005	Archives of Neurology	USA	206
Beric[20]	Complications of deep brain stimulation surgery	2002	Stereotactic and Functional Neurosurgery	USA	203

Categories

The articles were then categorized, and primary research articles accounted for 72% of our samples. The most present categories were articles that discussed *General outcomes* from DBS, which included 33 articles (5,044 citations). We separated from these general overviews the ones that investigated the *Long-term Outcomes* (more than 18 months), which was the second most discussed topic (9 articles, 1,327 citations). The third most present category was *Complications*, which had eight articles summing 996 citations.

The other 28 articles in our sample were from secondary research; most of them were Reviews, and only one a Guideline. Most of the secondary researches focused on *Treatment* aspects using DBS (13 articles, 1,508 citations), and other minor focus were given to *Pathophysiology* (3 articles, 388 citations) and *General outcomes* (3 articles, 294 citations).

DBS Targets

As reported in Figure 1, we detailed how DBS targets were applied in the different publications listed in the 100 most cited articles in "DBS in dystonia". There were 31 articles in which this analysis was not applicable, as the target was either not mentioned or DBS treatment was not specified in a general manner. Doubtlessly, the most mentioned target was the GPi, which appeared in 60 articles (8,255 citations), 54 of which being from primary research (7,662 citations). Thalamic targets were, then, the most present ones. In total, 10 articles mentioned thalamic targets: five of which did not specify a precise target, three focused on the *Ventral intermediate nucleus* (VIM), one on the *Ventrolateral thalamic nucleus* (VLp), and one on the *Ventral-oralis complex* (Vo). The total citations for thalamic targets were 1,126. Additionally, seven articles were focusing on the *Subthalamic nucleus* (STN, 663 citations), five of which were from primary research (461 citations). One primary research article focused on the *Caudal Zona Incerta* (cZi) and had 143 citations.

Dystonia classification

In the most cited articles, the description of dystonia classification was only observed in primary articles (Table 3). A fraction of them (18, under *Miscellaneous*) did present different kinds of dystonia in the study. The most common dystonia investigated was *idiopathic or inherited, isolated, segmental or generalized dystonia*, which presented 16 articles (3,269 citations), followed by *idiopathic, isolated, focal dystonia*, with 11 publications (1,238 citations).

DISCUSSION

Our analysis indicated that the 10 most cited journals in DBS in dystonia accounted for almost three-quarters of

Table 2. Ten most cited first authors and all authors in deep brain stimulation in dystonia research.

Type of authorship	First author	Number of articles in the top 100 (primary research)	Number of citations (primary research)	h index
First authorship	Vidailhet[11,14,21,22]	4 (3)	1,222 (1,143)	66
	Kupsch[12]	1 (1)	597 (597)	63
	Volkmann[18,23,24,25]	4 (2)	547 (256)	61
	Krauss[26,27,28,29]	4 (2)	514 (271)	50
	Jankovic[30,31,32]	3 (0)	424 (0)	126
	Albanese[17,33]	2 (0)	412 (0)	63
	Coubes[13,34]	2 (2)	363 (363)	32
	Ostrem[35,36,37]	3 (2)	354 (228)	31
	Cif[38,39,40]	3 (3)	294 (294)	20
	Kumar[16,41]	2 (1)	284 (220)	44
Any authorship	Pollak[11,14,15,21]	7 (6)	1,646 (1,520)	83
	Volkmann[12,18,23,24,25,42,43,44,45]	9 (7)	1,625 (1,334)	61
	Benabid[11,14,15,21,46,47]	6 (5)	1,568 (1,442)	88
	Vercueil[11,14,15,46,47,48,49]	7 (5)	1,524 (1,322)	25
	Schneider[12,24,25,43,45,50,51,52,53]	9 (9)	1,430 (1,430)	47
	Vidailhet[11,14,21,22,47,49]	6 (5)	1,372 (1,293)	66
	Kupsch[12,24,25,43,45,51,52,53]	8 (7)	1,356 (1,356)	63
	Krauss[17,24,26,27,28,29,33,50,54,55,56,58]	10 (6)	1,302 (647)	50
	Agid[11,14,21,49]	4 (3)	1,221 (1,023)	121
	Ardouin[11,14,21,49]	4 (4)	1,221 (1,221)	26

all articles and citations in our sample. They are majorly published in *Movement Disorders* (19%), the *Journal of Neurosurgery* (9%), *Neurology* (9%), *Brain* (8%), and *The Lancet Neurology* (7%). However, the journals with most cited articles were the *New England Journal of Medicine* (average of 638 citations per paper), *The Lancet Neurology* (152.1 citations/paper), and the *European Journal of Neurology* (146.5 citations/paper).

DBS in dystonia is mainly researched in the Northern Hemisphere. In the top 100 most cited articles in this theme, there were no countries outside of it. European countries offer significant contributions (57% of our sample). France and Germany have the highest citation rates of all countries. When considering primary research articles, France displays 192.5 citations/paper, and Germany 144.1 citations/paper. The United States also contributes to 31% of the articles, with 129.8 citations/paper.

Most primary research articles focus on *General outcomes* (46%), followed by *Long-term outcomes* (12.5%), and *Complications* (11%). Few publications in our sample tried to find *Predictors of outcome* (2.8%) or compared different *Targets* (1.4%). When we analyzed the used DBS targets for dystonia treatment, the classic *GPi* corresponded to 77% of the publications, *STN*, which is a hopeful new option of treatment[4,5,56], totalized 9%, and thalamic targets 13%, though mainly used for dystonic tremor[56,57].

As expected, most articles analyzed *idiopathic or inherited, isolated, segmental or generalized dystonia* (27% with an average of 204.3 citations/paper), which is the most studied kind of dystonia and has, usually, the most improvement after DBS treatment[58]. *Idiopathic, isolated, focal dystonia* is also highly prevalent (18.5%, 112.6 citations/paper) in our sample.

Similarly to other bibliometric analysis, our study has limitations. Scopus possesses greater coverage and specificity when compared to Web of Science and Google Scholar; however, we employed it as a single database for article retrieval[6,59,60]. Moreover, articles published after 2014 did not displayed in our sample, possibly because more recent articles are still accumulating citations. Nevertheless, this context does not undervalue their potential.

DBS in dystonia research is mainly focused on selected, Northern Hemisphere countries. They are mostly published in a handful of scientific journals and mainly focusing on outcomes of the surgery, with GPi as DBS target, and in idiopathic or inherited, isolated, segmental or generalized dystonia. This bibliometric analysis might assist unfamiliar researchers and practitioners in obtaining an overview of this particular domain.

Table 3. Citation and publication profile according to the type of dystonia in the 100 most cited articles in deep brain stimulation in dystonia research.

Dystonia classification	Number of articles (primary research)	Number of citations (primary research)
Idiopathic or inherited, isolated, segmental or generalized dystonia	16 (16)	3,269 (3269)
Idiopathic, isolated, focal dystonia	11 (11)	1,238 (1238)
Acquired dystonia	7 (7)	759 (759)
Combined dystonia	5 (5)	416 (416)
Dystonia associated with other neurological or systemic manifestations	2 (2)	250 (250)
Miscellaneous	18 (18)	2,133 (2133)
Not applicable	41 (13)	4,973 (1644)

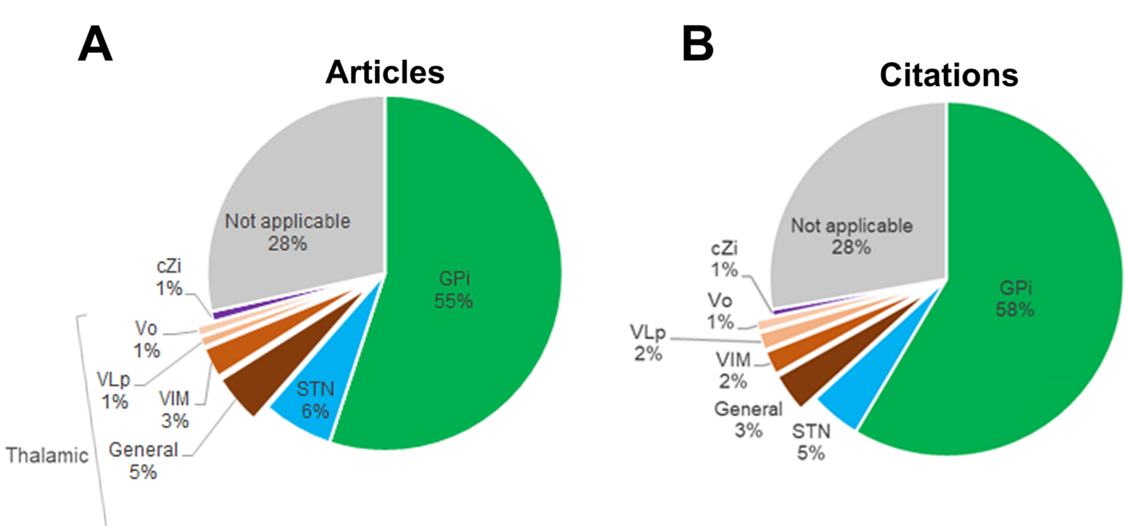

Figure 1. Pie charts indicating the distribution of (A) articles and (B) citations for different deep brain stimulation targets.

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