

Reviews

The Top 50 Most-Cited Articles in Orthostatic Tremor: A Bibliometric Review

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Abstract

Background: Article-level citation count is a hallmark indicating scientific impact. We aimed to pinpoint and evaluate the top 50 most-cited articles in orthostatic tremor (OT).

Methods: The ISI Web of Knowledge database and 2017 Journal Citation Report Science Edition were used to retrieve the 50 top-cited OT articles published from 1984 to April 2019. Information was collected by the Analyze Tool on the Web of Science, including number of citations, publication title, journal name, publication year, and country and institution of origin. Supplementary analyses were undertaken to clarify authorship, study design, level of evidence, and category.

Results: Up to 66% of manuscripts were recovered from five journals: *Movement Disorders* (n = 18), *Brain* (n = 4), *Journal of Clinical Neurophysiology* (n = 4), *Neurology* (n = 4), and *Clinical Neurophysiology* (n = 3). Articles were published between 1984 and 2018, with expert opinion as the predominant design (n = 22) and review as category (n = 17). Most articles had level 5 evidence (n = 26). According to their countries of origin, 34% of articles belonged to the United States (n = 17) leading the list, followed by United Kingdom (n = 15). University College London yielded the greater number of articles (n = 12), followed by the University of Kiel (n = 9). Most popular authors were G. Deuschl (n = 10), C.D. Marsden (n = 6), J. Jankovic (n = 5), P.D. Thompson (n = 5), J.C. Rothwell (n = 5), L.J. Findley (n = 4), and P. Brown (n = 4), who together accounted for 48% of them. All papers were in English.

Discussion: Publishing high-cited OT articles could be facilitated by source journal, study design, category, publication language, and country and institution of origin.

Keywords: Articles, bibliometrics, citation analysis, impact, orthostatic tremor, top-cited

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Introduction

Bibliometrics is a statistical analysis of books, articles, and/or other publications. Bibliometric analysis use data on numbers and authors of scientific publications and on articles and the citations therein, and in

patents, to evaluate the output of individuals/research teams, institutions, and/or countries; to identify national and international networks; and to delineate the development of novel multidisciplinary areas of science and technology.¹

One method to estimate the academic relevance of an article is the rate at which the work is cited by other authors. When an article is referenced by another peer-reviewed article receives a “citation”.² The number of times an article is quoted in scientific journals translates its impact on a specific biomedical field or specialty, and indicates the impact of the authors’ creativity.³⁻⁵ The Institute for Scientific Information (ISI) has been collecting citation and other academic impact information since 1945, and it has been available electronically since 1979. ISI (currently a subsidiary of Thomson Corp.) calls their newest journal citation system “Science Citation Index Expanded (SCIE)”, and it is one of the databases accessible under the banner of Web of Science.^{2,6,7} Citation data from peer-reviewed articles are indexed from more than 10,000 high-impact journals not only in the sciences and social sciences, but also in the arts and humanities.^{2,8}

In the last few years, several citation studies of the top-cited articles have been carried out in different areas of clinical neurology and neurosurgery, such as essential tremor (ET),^{2,9,10} dystonia,¹⁰ Parkinson’s disease (PD),¹¹ epilepsy,¹² multiple sclerosis,¹³ brain metastases research,¹⁴ and neurosurgery.¹⁵⁻²⁰ Nevertheless, no similar study has been conducted in the context of orthostatic tremor (OT). OT, also called “shaky leg syndrome”,⁸ is an uncommon neurological disease that was first coined by Heilman.²¹ It has an undetermined prevalence and incidence, and is typically distinguished by unsteadiness along with a uniquely coherent 13–18 Hz postural tremor of the leg, trunk and, less frequently, arm, neck and cranial muscles, while standing, and subsiding on movement or sitting. It constitutes one of the most unusual and enigmatic tremor syndromes, which is thought to be generated from so far not exactly known central oscillator located at the brainstem and/or cerebellum, without ruling out the possible involvement of ponto-cerebello-thalamo-cortical pathway, fronto-cerebellar loops, motor and sensory cortices, and the basal ganglia.^{8,22-34} Hence, more studies are warranted to shed light on the epidemiology and etiopathogenesis of this potentially disabling movement disorder.

Recent years have seen an exponential increase in the number of scientific publications on OT, mainly distributed through the fields of epidemiology, pathophysiology, clinical phenomenology, diagnostic tools, and treatment options.^{8,22-52}

We used the electronic version of the SCIE to determine which published OT articles have been most usually cited by other authors, ranking the 50 top-cited works. By analyzing the characteristics of these articles, we tried to provide a comprehensive review, identifying the most highly cited articles in OT research, including primary OT and OT plus, and determining what properties make these articles relevant for further studies and clinical practices.

Methods

A search was performed through the bibliometric database Web of Science for articles using the topic search term “orthostatic tremor*” (the asterisk was included as a wild card character). In early April 2019 (April 6, 2019), we found the 50 top-cited OT articles (Table 1) published in professional journals between 1900 and 2019 by the Web of Science. The full texts were mainly picked by PubMed, ScienceDirect, and ClinicalKey.

The search term for OT returned 508 articles with the earliest published in 1984.²¹ The results were separately sorted by number of citations from highest to lowest and were manually examined to identify the top 50 cited articles related to the topic. Works were regarded as “citation classics” if they received 400 or more citations.⁵³⁻⁵⁵ Key data regarding country of origin (based on the first author’s affiliation), institution, year of publication, publication name, and citations of the target articles were obtained from Web of Science using the Analyze Tool. Further analyses were then performed to ascertain authorship, article type, study design, and level of evidence. For each study, the level of evidence was graded according to the Oxford Centre for Evidence-Based Medicine (OCEBM) Levels of Evidence (2011).^{56,57}

Cited half-life is defined as the number of publication years, going back from the current year, that account for 50% of current citations received. This index helps to evaluate the age of the majority of cited articles published in a journal. Only those journals cited 100 or more times have a cited half-life.⁶ On the other hand, the h-index aims to measure the cumulative impact of a researcher’s output; the value of h is equal to the number of papers (n) that have n or more citations.⁵⁸

Results

The 50 top-cited OT articles (Table 1) were published in 18 journals. Journal title, number of articles, impact factor 2017, 5-year impact factor, and cited half-life are detailed in Table 2. Sixty-six per cent of articles were recovered from five journals: *Movement Disorders* (n = 18),^{22,59-75} *Brain* (n = 4),⁷⁶⁻⁷⁹ *Journal of Clinical Neurophysiology* (n = 4),⁸⁰⁻⁸³ *Neurology* (n = 4),⁸⁴⁻⁸⁷ and *Clinical Neurophysiology* (n = 3).⁸⁸⁻⁹⁰ The 50 top-cited articles on OT were published from 1984²¹ to 2018²² (Figure 1A). The largest number of top-cited articles (n = 25) were published between 1996 and 2002,^{59-63,65-67,70,76,78-85,87,89,91-95} and eight of the top 10-cited articles were published in the 1990s (n = 4)^{59,77,80,81} and the 2000s (n = 5)^{60,76,78,88,91} (Table 1 and Figure 1A).

Twenty-eight authors contributed to design two or more articles to the list, and seven authors contributed to four or more articles (Table 3). The number of authors per article varied from one^{21,23,62,66,80,82-84,93} to 28,⁵⁹ being the most usual figures one (nine articles),^{21,23,62,66,80,82-84,93} two (nine articles),^{63,68,69,73,76,96-99} four (eight articles),^{78,81,85,88,91,94,100,101} and three (six articles).^{61,67,70,72,79,86} The most common authors of the top-cited articles in OT were G. Deuschl (n = 10),^{22,59-61,63,69,81,89,91,92} C.D. Marsden (n = 6),^{59,76,77,95,102,103} J. Jankovic (n = 5),^{59,62,65,68,84} P.D. Thompson (n = 5),^{59,77,85,102,103} J.C. Rothwell (n = 5),^{59,64,77,102,103} L.J. Findley (n = 4),^{59,77,82,103} and P. Brown (4),^{64,94,95,104} and when combined, they accounted for 48% (n = 24) of the articles on the list^{22,59-65,68,69,76,77,81,82,84,85,89,91,92,94,95,102-104} (Table 3).

According to first author’s affiliation, 34% of articles were produced in the United States of America (USA) (n = 17),^{21,23,62,65-69,71,72,80,84,86,89,93,97,99} leading the list, closely followed by the United Kingdom (n = 15),^{22,64,76-79,82,85,94-96,100,102-104} (Table 1). The next most productive country was Germany (n = 10),^{59-61,63,69,81,89,90,91,92} followed by Italy (n = 3),^{83,98,101} Spain,^{75,87} the Netherlands,^{88,105} and Austria,^{64,96} each aided two articles, and Argentina,⁷³ France,⁷⁴ Sweden,⁹⁹ and Taiwan,⁷⁰ each contributed one article to the list (Table 1). The 28 leading institutions that provided

Table 1. The 50 top-cited OT articles (ranked by number of citations)

Absolute number	Rank	Paper	Number of citations	Country of origin (based on first author's affiliation)	Number of authors	Study design	Category
1	1	Deuschl et al. ⁵⁹	1,206	Germany	28	Expert opinion	Clinical: Classification
2	2	McAuley and Marsden ⁷⁶	284	UK	2	Expert opinion	Review
3	3	Deuschl et al. ⁹¹	263	Germany	4	Expert opinion	Review
4	4	Bain et al. ⁷⁷	262	UK	7	Case series	Epidemiology
5	5	Elble ⁸⁰	193	USA	1	Expert opinion	Review
6	6	Visser et al. ⁸⁸	184	The Netherlands	4	Expert opinion	Review
7	7	Heilman ²¹	174	USA	1	Case series	Clinical: Classification
8	8	Alusi et al. ⁷⁸	144	UK	4	Case series	Epidemiology
9	9	Deuschl et al. ⁸¹	134	Germany	4	Expert opinion	Review
10	10	Raethjen et al. ⁶⁰	116	Germany	6	Case series	Laboratory: Pathophysiology
11	11	Wilms et al. ⁶¹	115	Germany	3	Expert opinion	Review
12	12	Jankovic ⁸⁴	108	USA	1	Expert opinion	Clinical: Classification
13	13	Deuschl and Bergman ⁶³	107	Germany	2	Expert opinion	Review
14	14	Thompson et al. ¹⁰²	98	UK	7	Case report	Laboratory: Pathophysiology
15	15	Katzenschlager and Lees ⁹⁶	97	UK and Austria	2	Expert Opinion	Review
16	16a	Gerschlagler et al. ⁶⁴	89	UK and Austria	8	Case series	Epidemiology
17	16b	McManis and Sharbrough ⁹⁷	89	USA	2	Case series	Clinical: Classification
18	17a	Ondo et al. ⁶⁵	84	USA	5	Randomized controlled trial	Clinical: Medical therapies
19	17b	Britton et al. ¹⁰³	84	UK	7	Case series	Clinical: Classification
20	18	Jankovic ⁸⁴	81	USA	1	Expert opinion	Clinical: Classification
21	19	Wills et al. ⁸⁵	72	UK	4	Case series	Laboratory: Pathophysiology
22	20a	Hallett ⁶⁶	69	USA	1	Expert opinion	Review
23	20b	Findley ⁸²	69	UK	1	Expert opinion	Review
24	21a	Abdo et al. ¹⁰⁵	67	The Netherlands	5	Expert opinion	Review
25	21b	Cantello ⁸³	67	Italy	1	Expert opinion	Review
26	22	Köster et al. ⁹²	61	Germany	7	Case series	Laboratory: Pathophysiology
27	23	Chouinard et al. ⁶⁷	58	USA	3	Expert opinion	Epidemiology
28	24	Wang et al. ¹⁰⁰	56	UK	4	Case report	Laboratory: Pathophysiology
29	25	Lauk et al. ⁸⁹	55	Germany and USA	6	Case series	Laboratory: Pathophysiology
30	26a	Nardone and Schieppati ⁹⁸	54	Italy	2	Expert opinion	Review
31	26b	Sethi ⁹³	54	USA	1	Expert opinion	Review

Table 1 continued

Table 1. (Continued) **The 50 top-cited OT articles (ranked by number of citations)**

Absolute number	Rank	Paper	Number of citations	Country of origin (based on first author's affiliation)	Number of authors	Study design	Category
32	27a	Katzenschlager et al. ¹⁰⁴	51	UK	11	Case-control study	Laboratory: Pathophysiology
33	27b	FitzGerald and Jankovic ⁶⁸	51	USA	2	Case series	Clinical: Classification
34	28a	Bhatia et al. ²²	50	UK	18	Expert opinion	Clinical: Classification
35	28b	Elble and Deuschl ⁶⁹	50	USA and Germany	2	Expert opinion	Review
36	28c	Wu et al. ⁷⁰	50	Taiwan	3	Case series	Laboratory: Pathophysiology
37	29	Wee et al. ⁸⁶	48	USA	3	Case series	Clinical: Classification
38	30a	Espay et al. ⁷¹	47	USA	14	Case series	Clinical: Surgical therapies
39	30b	Piboolnurak et al. ⁷²	47	USA	3	Case series	Clinical: Classification
40	31a	Papa and Gershanik ⁷³	46	Argentina	2	Case series	Clinical: Classification
41	31b	Martinelli et al. ¹⁰¹	46	Italy	4	Case series	Epidemiology
42	32a	Elble ²³	45	USA	1	Expert opinion	Review
43	32b	Semenescu et al. ⁷⁴	45	France	8	Case series	Clinical: Classification
44	32c	Benito-León et al. ⁸⁷	45	Spain	6	Case series	Clinical: Classification
45	33	Guridi et al. ⁷⁵	43	Spain	9	Case report	Clinical: Surgical therapies
46	34a	Fung et al. ⁷⁹	40	UK	3	Case series	Laboratory: Pathophysiology
47	34b	Yarrow et al. ⁹⁴	40	UK	4	Case series	Laboratory: Pathophysiology
48	35a	Puschmann and Wszolek ⁹⁹	39	USA and Sweden	2	Expert opinion	Review
49	35b	Krafczyk et al. ⁹⁰	39	Germany	5	Case-control study	Clinical: Classification
50	35c	Wills et al. ⁹⁵	39	UK	5	Case report	Clinical: Medical therapies

UK: United Kingdom; USA: United States of America.

the 50 top-cited OT articles are sorted in Table 4. University College London in the United Kingdom originated the highest number of top-cited OT articles ($n = 12$),^{22,64,76,77,79,85,94-97,103,104} followed by the University of Kiel in Germany ($n = 8$)^{59-61,63,69,81,89,91} and Baylor College of Medicine in USA ($n = 4$).^{62,65,68,84}

Among the 50 top-cited OT articles, the most prevalent design was expert opinion ($n = 22$),^{22,23,59,61-63,66,67,69,76,80-84,88,91,93,96,98,99,105} and the next most frequent design was case series ($n = 21$),^{21,60,64,68,70-74,77-79,85-87,89,92,94,97,101,103} and case-control study ($n = 2$)^{90,104} (Table 1). Only one article was a randomized controlled trial (RCT)⁶⁵ (Table 1). According to the OCEBM (2011),^{56,57} and in line with the study design, the most common level of evidence was that of a level 5 study, including expert opinion ($n = 22$)^{22,23,59,61-63,66,67,69,76,80-84,88,91,93,96,98,99,105} and case report ($n = 4$)^{75,95,100,102} studies, followed by level 4 with case series with or

without intervention ($n = 21$),^{21,60,64,68,70-74,77-79,85-87,89,92,94,97,101,103} level 3 with case-control study ($n = 2$),^{90,104} and finally level 1 with high-quality, properly powered and conducted RCT ($n = 1$)⁶⁵ (Table 5). All the 50 top-cited OT manuscripts were written in English.

For following analysis of the most-cited articles, each paper was classified into one of seven categories: epidemiology, clinical classification, laboratory pathophysiology, clinical medical therapies, clinical surgical therapies, and review articles. The number of articles in each category is shown in Figure 1B.

Epidemiology

Studies were incorporated in this category if they described the OT epidemiology such as prevalence, incidence, benchmarks, and trends over time. There were five articles in this category. One study

Table 2. Journals that published the 50 top-cited OT articles (ranked by number of articles)

Absolute number	Rank	Journal	Number of articles	Impact factor 2017	5-year impact factor	Cited half-life
1	1	<i>Movement Disorders</i>	18	8,324	7,523	7,7
2	2a	<i>Brain</i>	4	10,84	11,199	9,7
3	2b	<i>Journal of Clinical Neurophysiology</i>	4	1,982	1,813	>10
4	2c	<i>Neurology</i>	4	7,609	8,515	>10
5	3	<i>Clinical Neurophysiology</i>	3	3,614	3,638	9,6
6	4a	<i>Annals of Neurology</i>	2	10,244	10,744	>10
7	4b	<i>Archives of Neurology (currently known as JAMA Neurology)</i>	2	11,46	10,415	2,9
8	4c	<i>Current Opinion in Neurology</i>	2	401	4,427	7,3
9	4d	<i>Muscle & Nerve</i>	2	2,496	2,494	>10
10	5a	<i>Acta Neurologica Scandinavica</i>	1	3,126	2,877	>10
11	5b	<i>European Journal of Physical and Rehabilitation Medicine</i>	1	2,208	228	4,7
12	5c	<i>Gait & Posture</i>	1	2,273	2,971	8
13	5d	<i>Journal of Neurology</i>	1	3,783	3,805	7,5
14	5e	<i>Journal of Neurology Neurosurgery and Psychiatry</i>	1	7,144	6,923	>10
15	5f	<i>Journal of Neuroscience Methods</i>	1	2,668	2,571	>10
16	5g	<i>Nature Reviews Neurology</i>	1	19,819	20,888	4,3
17	5h	<i>Neurologic Clinics</i>	1	3,072	2,638	8,8
18	5i	<i>Seminars in Neurology</i>	1	1,87	2,19	8,2

JAMA: Journal of the American Medical Association.

examined the clinical and epidemiological phenotype of hereditary ET, without finding primary OT.⁷⁷ Another study investigated the clinical and epidemiological features of tremor in multiple sclerosis, and primary OT was not detected.⁷⁸ The remaining three examined the evolution and syndromic associations of OT,⁶⁴ different clinical characteristics of ET, including OT,¹⁰¹ and the frequency of inter-rater agreement and disagreement about ET diagnostic criteria, along with OT.⁶⁷

Clinical: Classification

This wide category included consensus statement on diagnostic criteria for OT, novel description of the disease, its classification into subtypes, as well as targeted measures, and diagnostic tools. There were 14 OT papers in this category,^{21,22,62,68,72-74,84,86,87,90,97,103} including one citation classic.⁵⁹

Laboratory: Pathophysiology

Works analyzing pathophysiological processes underlying OT were incorporated in this category. There were 10 studies, employing neurophysiological^{60,70,79,89,92,94,100,102} and neuroimaging^{85,104} techniques. There were six studies on physiology,^{60,70,92,94,100,102} two on pathology,^{79,104} and one in both.⁸⁹

Clinical: Medical therapies

Research articles related to the application of medical and nonsurgical treatments were included in this category. There were two articles on medical treatment, with one RCT on gabapentin,⁶⁵ and one case report on levodopa.⁹⁵

Clinical: Surgical therapies

Research articles regarding primarily surgical treatment were incorporated in this category. There were two articles about the effect of deep brain stimulation of the ventral intermediate nucleus of the thalamus (Vim-DBS) for drug-refractory OT, including one case series⁷¹ and one case report,⁷⁵ with positive results.

Review articles

There were included 17 review articles: seven about OT pathophysiology,^{61,63,66,76,80,81,91,108} and the rest reviewing general aspects,^{23,69,82,93} apart from diagnosis,^{88,96,98,99} treatment,⁹⁹ and the effect of transcranial magnetic stimulation.⁸³

Time trends and journals

The publication years of the most cited articles are outlined in Figure 1A. This revealed a peak generation of the most cited OT articles for

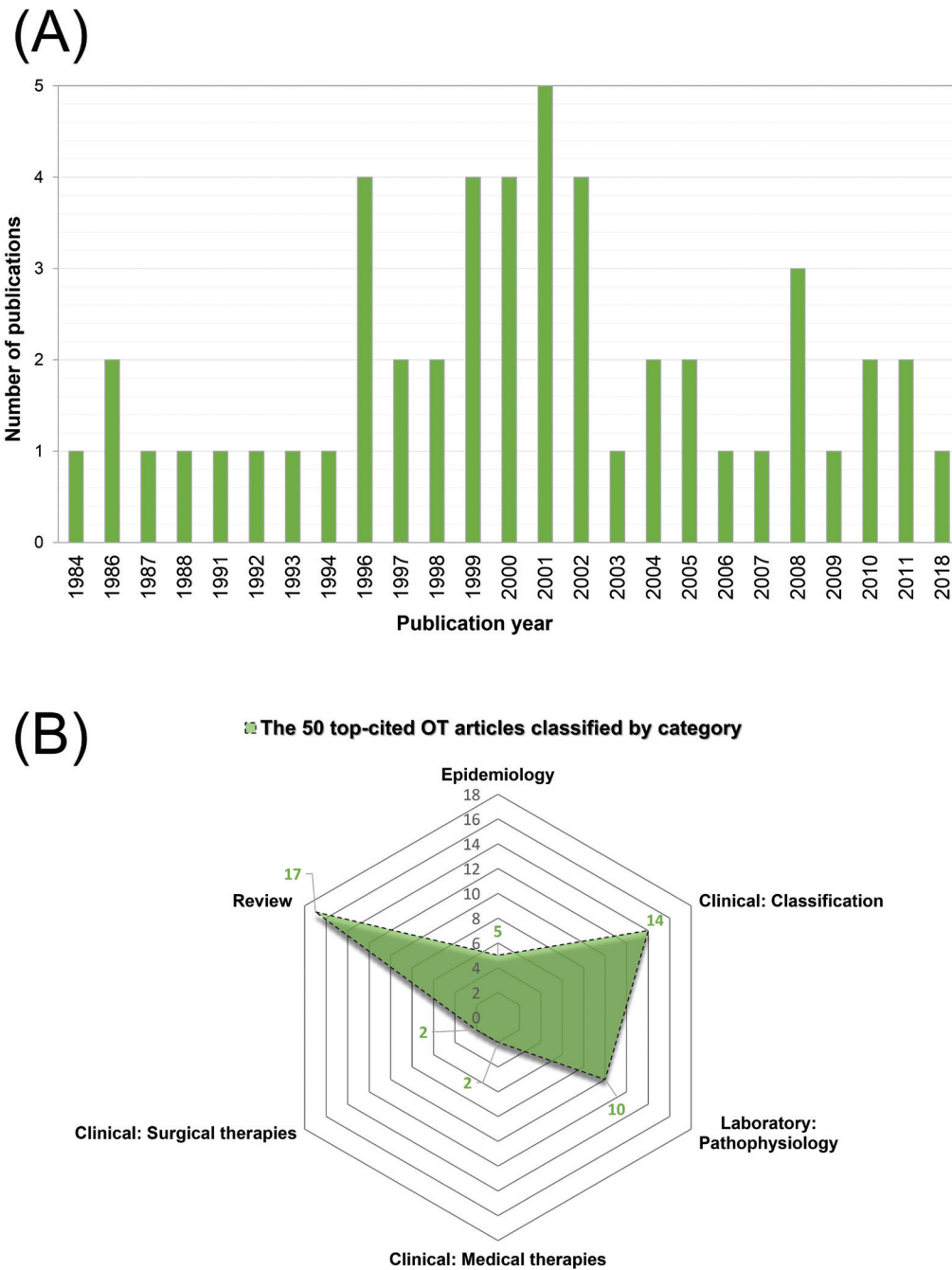


Figure 1. (A) Publication years for the 50 top-cited orthostatic tremor (OT) articles. (B) Plot showing the number of OT articles by category.

works published between 1996 and 2002.^{59-63,65-67,70,76,78-85,87,89,91-95} The most cited articles were published in 18 journals. The top 5 journals accounted for 33 (66%) of the 50 most-cited OT articles^{22,59-90} (Table 2).

Discussion

In the medical literature, the study of the number of times authors reference an article is one measure of the influence of the publication,

and this type of citation analysis is widespread.³⁻⁵ The assessment of specialty-wide citation analysis has been notified in other areas of the neurosciences.^{2,9-19,106} By ranking the 50 most-cited papers, we sought to establish which published journal articles in OT have greater influence on citation impact. The top article was cited 1,206 times.⁵⁹ This figure is considerably lower than that found in PD, in which the top-cited article earned 4,327 citations.¹¹

Table 3. Authors with two or more of the 50 top-cited OT articles (ranked by number of citations)

Absolute number	Rank	Author	Number of citations	Number of articles		h-index
				As first author	As co-author	
1	1	Deuschl G	10	4	6	10
2	2	Marsden CD	6	0	6	6
3	3a	Jankovic J	5	2	3	5
4	3b	Thompson PD	5	1	4	5
5	3c	Rothwell JC	5	0	5	5
6	4a	Findley LJ	4	1	3	4
7	4b	Brown P	4	0	4	4
8	5a	Katzenschlager R	3	2	1	3
9	5b	Elble RJ	3	1	2	3
10	5c	Lauk M	3	1	2	3
11	5d	Raethjen J	3	1	2	3
12	5f	Day BL	3	0	3	3
13	5g	Lees AJ	3	0	3	3
14	5h	Timmer J	3	0	3	3
15	6a	Wills AJ	2	2	0	2
16	6b	Bain PG	2	1	1	2
17	6c	Gerschlager W	2	1	1	2
18	6d	Hallett M	2	1	1	2
19	6e	Koster B	2	1	1	2
20	6f	Bain P	2	0	2	2
21	6g	Bhatia KP	2	0	2	2
22	6h	Bloem BR	2	0	2	2
23	6i	Gresty MA	2	0	2	2
24	6j	Guschlbauer B	2	0	2	2
25	6k	Krack P	2	0	2	2
26	6l	Lindemann M	2	0	2	2
27	6m	Louis ED	2	0	2	2
28	6n	Lucking CH	2	0	2	2

Most of the top-cited OT articles were usually published in specialized journals such as *Movement Disorders*, *Brain*, *Journal of Clinical Neurophysiology*, *Neurology*, and *Clinical Neurophysiology* (Table 2). In 2002, Callahan et al.¹⁰⁷ noticed that the strongest predictor of article's citations per year was the impact factor of the original publishing journal, instead of the methodology or quality of the research. By contrast, as our study has demonstrated, the current citation value of the individual paper is not positively correlated with the journal's impact factor. An example would be articles published in the *Journal of Clinical Neurophysiology*, representing the third journal of those that published the

50 top-cited OT articles, much higher than expected by the journal's impact factor. This is presumably attributable to contributions on pathophysiology^{80,81,83} and classification⁸² of OT from these papers. This implies that publishing remarkable neurophysiology and classification works in specialized journals is also able to achieve significant impact.

Overall, as can be seen from Figure 1A, the 50 top-cited articles in OT were articles that have been available for 20 or more years, and only one target article was published more recently (in 2018). It has been reported that scientific articles begin to be cited 1 or 2 years after publication and reach a maximum citation rate of 7 to 10 years

Table 4. Institution of origin of the 50 top-cited OT articles based on first author's affiliation (ranked by number of articles)

Absolute number	Rank	Institution of origin	Number of articles
1	1	University College London, London, UK	12
2	2	University of Keil, Keil, Germany	8
3	3	Baylor College of Medicine, Houston, Texas, USA	4
4	4a	Southern Illinois University School of Medicine, Springfield, Illinois, USA	3
5	4b	University of Freiburg, Freiburg, Germany	3
6	5a	Columbia University, New York, New York, USA	2
7	5b	Radboud University Nijmegen, Nijmegen, The Netherlands	2
8	5c	University of Eastern Piedmont "Amedeo Avogadro", Novara, Italy	2
9	6a	Imperial College School of Medicine, London, UK	1
10	6b	University of Oxford, Oxford, UK	1
11	6c	Oldchurch Hospital, London, UK	1
12	6d	King's College London, London, UK	1
13	6e	Boston University, Boston, Massachusetts, USA	1
14	6f	Mayo Clinic, Rochester, Minnesota, USA	1
15	6e	National Institutes of Health, Bethesda, Maryland, USA	1
16	6h	University of Florida College of Medicine, Gainesville, Florida, USA	1
17	6i	Medical College of Georgia, Augusta, Georgia, USA	1
18	6j	University of Mississippi Medical Center, Jackson, Mississippi, USA	1
19	6k	University of Cincinnati, Cincinnati, Ohio, USA	1
20	6l	Mayo Clinic, Jacksonville, Florida, USA	1
21	6m	Sección de Enfermedades Extrapiramidales, Centro Neurológico, Hospital Francés, Buenos Aires, Argentina	1
22	6n	Hospital Universitario 12 de Octubre, Madrid, Spain	1
23	6o	Clínica Universitaria, Universidad de Navarra, Pamplona, Spain	1
24	6p	University of Bologna, Bologna, Italy	1
25	6q	Saint-Antoine Hospital, AP-HP, Paris, France	1
26	6r	Lund University, Lund, Sweden	1
27	6s	University of Munich, Munich, Germany	1
28	6t	Chang-Gung Memorial Hospital, Taipei, Taiwan	1

AP: Assistance Publique; HP: Hôpitaux de Paris; UK: United Kingdom; USA: United States of America.

after publication.¹⁰⁸ However, an interval of 10 to 20 years is needed for the maximal recognition of prominent articles in a field.^{54,109} This may explain why recently published articles were seldom cited and few appeared on the list.

Among the 50 top-cited OT articles, the most widespread design was the expert opinion ($n = 22$), followed by the case series ($n = 21$) (Table 1), illustrating that descriptive and observational studies, respectively, are most frequent for OT. This fact indicates the relative ease to accomplish simpler study designs in OT. In order of importance of research, study designs, systematic reviews, meta-analyses, and well-conducted RCTs

yield the highest quality of evidence for most clinical or interventional questions, and the lowest value corresponds to expert opinions. Among the 50 target articles, there were only one RCT, being among the top 20 OT articles.⁶⁵ This is consistent with the contributions of the studies that have analyzed the most-cited papers in other fields.^{2,9,108,110–118} There are several potential reasons for the low numbers of RCTs. Firstly, RCTs are time- and money-consuming studies. Secondly, it is hard to recruit large sample and control groups. Thirdly, it is possible that RCTs were published comparatively recently, and thus, they still have not attained a representative number of citations.

Table 5. Level of evidence of the 50 top-cited OT articles

Level of evidence	Study type	Number of articles
1	High-quality, properly powered and conducted RCT	1
	Systematic review of these studies	0
	Meta-analysis of these studies	0
2	Well-designed controlled trial without randomization	0
	Prospective comparative cohort trial	0
3	Retrospective cohort study	0
	Case-control study	2
	Systematic review of these studies	0
4	Case series with or without intervention	21
	Cross-sectional study	0
5	Expert opinion	22
	Case report	4
	Bench research	0

Adapted from the Oxford Centre for Evidence-Based Medicine (2011).⁵⁶
RCT: Randomized controlled trial.

By category, the most cited OT studies were review articles ($n = 17$),^{23,61,63,66,69,76,80-83,88,91,93,96,98,99,108} followed by clinical classification ($n = 14$),^{21,22,59,62,68,72-74,84,86,87,90,97,103} laboratory pathophysiology ($n = 10$),^{60,70,79,85,89,92,94,100,102,104} epidemiology ($n = 5$),^{64,67,77,78,101} clinical medical ($n = 2$)^{65,95} and surgical ($n = 2$)^{71,75} therapies (Table 1).

Compared to ET^{9,10} and PD,¹¹ articles concerning epidemiology, clinical classification, genetics, pathophysiology, and medical and surgical treatment are scarce, demonstrating that OT is a smaller area of research in neurology.

Trends over time unveiled that the peak period when most cited OT papers were published was between 1996 and 2002. The proposed explanations for the peak during this time are that this period was especially active and profitable with outstanding success in clinical and neurophysiological characterization, as well as development of new medical and surgical therapies, assisting in the improvement of diagnosis and treatment of OT, respectively. Another potential explanation for the aforementioned peak may be a critical attribute of modern research as older articles are no longer cited because they have been replaced by more recent studies that have replicated their findings and produced more accurate information.¹⁸

In parallel, the latest studies have had not been given sufficient time to become settled as most cited OT articles, for instance, “Smartphone apps provide a simple, accurate bedside screening tool for orthostatic tremor”.³⁹

Finally, our review of the most cited OT articles might be worthwhile on the basis of different grounds. Firstly, our observations suggest that

the authors of citation classics in OT have produced more highly cited articles. Secondly, we have noticed several qualities contributing favourably to article citation, indicating that journal, country, and institution of origin are major factors. Finally, we identified only one Level 1 study among the most-cited OT articles.⁶⁵ Although the future of OT citation may be in higher level of evidence literature, nonetheless, this has not yet been a pivotal element of citation in the OT scenario.

Limitations

Seven major limitations of this review article must be noted. Firstly, notwithstanding the choice of Web of Science over Google Scholar which indexes a broader range of academic papers that could have had any influence on results, earlier reviews in other disciplines disclosed very similar results using these two search engines when the study field was small, as in our case with OT.^{18,106} Secondly, search term may not have brought all possible results despite our inclusion of an extensive keyword as “orthostatic tremor”. Likewise, for the Web of Science search engine, it may be possible to carry out either a title- or topic-based search. Given the small volume of the OT field, a topic-based search was preferred to recover all possible matches as stated above in the methods section. Conversely, a citation analysis study on OT by using a title-based search would have achieved fewer results.⁹ Thirdly, this kind of citation analysis does exclude citations in textbooks and lectures, and an author’s or authors’ potential bias to cite articles in the journal they intend to publish their manuscripts.¹¹⁹ Fourthly, there is a definite time effect in citation analysis, with recent studies earning fewer citations than the older ones.^{120,121} Fifthly, the language of publication has a key role, with an inherent bias for articles published in English being overrepresented. Sixthly, applying Kuhn’s philosophy of science¹²²⁻¹²⁴ to the focus of this review, and therefore, considering a paradigm as a core concept, authors of a scientific community would be prone to cite a paradigmatic paper because of its high citations instead of its content or quality. Seventhly, the classification of most-cited papers represents a dynamic state, modifying as time goes by, and hence is a snapshot of a point in time, reflecting an overview about the current situation of research on the matter.

Conclusions

Writing highly cited articles in OT may be facilitated primarily by appropriate choice of source journal (e.g., *Movement Disorders*), study design (e.g., expert opinion), category (e.g., review), and language of publication (English). And at later stage, it could be also important taking into account country (e.g., USA or UK) and institution (e.g., University College London) of origin.

To the best of our knowledge, this is the first bibliometric study to illustrate the most-cited articles in OT research. Noteworthy, the peak of citations has decreased since 2008. Contrary to ET^{9,10} and PD,¹¹ there are much fewer articles, as well as absence of citation classics, on epidemiology, clinical classification, genetics, pathophysiology, and medical and surgical treatment fields intended for further development of knowledge about this puzzling and challenging condition, which may severely impact on health-related quality of life. Sustaining that OT is a smaller field of research in neurology.

Although progress has been made in the diagnosis and treatment of OT, unfortunately, the correct diagnosis is often overlooked, delaying early administration of appropriate therapy, which is usually not completely successful. Therefore, these findings demonstrate that more studies are warranted to gain further insights into the nature and management of OT. Ultimately, it is essential to acknowledge top-cited OT articles because they involve landmarks and advances in OT.

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