

Gender Dysphoria Publication Trends: A Bibliometric Analysis between 1900 and 2018

Tendencias de publicación sobre disforia de género: Un análisis bibliométrico entre 1900 y 2018

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Abstract

Objective Research on gender dysphoria (GD) has been growing over the last decades with increasing interest in understanding and characterizing the causal relationships between psychological, genetics, hormonal, and sociocultural factors. Changes and acceptance of this condition as non-pathologic have led to significant changes in general perspective and its management over time. Our objective is to carry out a bibliometric analysis to know the publication trends and quality of evidence related to gender dysphoria.

Methods A systematic search and critical review of the literature was carried out between January 1900 and December 2018 to perform a bibliometric analysis. Research was done in the following databases: OVID, PubMed, EMBASE, Scopus, Web of Science and Google Scholar. The medical subject headings (MeSh) terms used were: *gender dysphoria*; and *surgery and psychology*. The results were plotted using the VOSviewer version 1.6.8. Statistical analyses were performed with the IBM SPSS, Version 25.0.

Keywords

- gender dysphoria
- bibliometrics
- publications
- information sciences

Results A total of 1,239 manuscripts were identified, out of which 1,041 were selected. The average number of cited times per year per manuscript is 1.84 (interquartile range [IQR] 0-2.33). The average impact index was 47.8 (IQR 20-111.6). The median of total citations per manuscript was 3 (IQR 0-33.1), and the highest number of citations per manuscript was 484. Most publications focus on the psychological aspects of GD, and there is a significant amount of manuscripts related to social and anthropological issues. Most articles have a low level of scientific evidence.

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Address for correspondence Nicolas Fernandez, M.D., PhD, Division of Urology, Seattle Children's Hospital, University of Washington, 4800 Sand Point Way NE, Seattle, WA 98105, United States (e-mail: Nicolas.fernandez@seattlechildrens.org). **Conclusion** There is a great amount of published literature on GD; however, there is a significant level of disagreement in many respects on this topic. Regarding surgical gender-affirmation, there is a lack of information supported by high level of evidence to uphold the emerging expansion of medical practices.

Resumen Objetivo La investigación sobre la disforia de género (DG) ha crecido en las últimas décadas con un interés creciente por comprender y caracterizar las relaciones causales entre factores psicológicos, genéticos, hormonales y socioculturales. La aceptación de esta condición como no patológica ha llevado a cambios significativos en la perspectiva general y su manejo a lo largo del tiempo. Nuestro objetivo es realizar un análisis bibliométrico para evaluar las tendencias de publicación y la calidad de evidencias relacionadas con la disforia de género.

Métodos Se realizó una búsqueda sistemática y revisión crítica de la literatura entre enero de 1900 y diciembre de 2018 para hacer un análisis bibliométrico. Se utilizaron los criterios de los Ítems de Informe Preferido para Revisiones Sistemáticas y Meta-análisis (Preferred Reporting Items for Systematic Reviews and Meta-analysis, PRISMA, en inglés), y la investigación se realizó en las siguientes bases de datos: OVID, PubMed, EMBASE, Scopus, Web of Science y Google Scholar. Los términos de encabezados de temas médicos (*medical subject headings*, MeSh, en inglés) utilizados fueron: *disforia de género*; y *cirugía y psicología*. Los resultados se trazaron utilizando VOSviewer, versión 1.6.8. Los análisis estadísticos se realizaron con el IBM SPSS, versión 25.0.

Resultados Se identificaron un total de 1.239 manuscritos, y se seleccionaron 1.041. El número promedio de citas por año por manuscrito fue de 1,84 (rango intercuartil [RIC]: 0–2,33). El índice de impacto promedio fue de 47,8 (RIC: 20–111,6). La mediana del total de citas por manuscrito fue de 3 (RIC: 0–33.1), y el mayor número de citas por manuscrito fue de 484. La mayoría de las publicaciones se centran en los aspectos psicológicos de la DG, y hay una cantidad significativa de manuscritos relacionados con temas sociales y antropológicos. La mayoría de los artículos tienen un bajo nivel de evidencia científica.

Palabras clave

- disforia de género
- bibliometría
- publicaciones
- ciencias de la información

Conclusión Existe una gran cantidad de literatura publicada sobre disforia de género; sin embargo, existe un nivel significativo de desacuerdo en muchos aspectos sobre este tema. Con respecto a la afirmación quirúrgica, hace falta información respaldada por un alto nivel de evidencia que argumente la expansión emergente de las prácticas médicas.

Introduction

The term *gender dysphoria* (GD) was first introduced as a medical term by Fisk, in 1974,¹ to describe discomfort with an individual's biological sex and the willingness to undergo medical intervention for gender reassignment. Research in gender development and gender dysphoria has been growing over the years as a sign of the intention to understand and characterize the causative relationships between the psychological sphere, genetic alterations, hormone conditions, and sociocultural factors. In addition, the will to describe variants in psychosexual differentiation and possible treatments, like surgical and hormone, for gender affirmation have led to find a wide range of different types of studies describing and exploring this condition.^{2–5}

The issues currently under discussion abound, taking into account the multifactorial origin of this condition and the

controversy concerning especially the social and phycological aspects.⁶ Despite the abundance of published literature, there is a significant level of discordance in many aspects of this subject, but, above all, a lack of literature with appropriate evidence that allows decisions to be made in clinical practice with a scientific basis.

There are multiple theories that explain the etiology of the gender dysphoric behavior. No biological anomalies have been found that may be associated with GD;⁷ however, it is suggested that the etiology could be explained by hormonal influences and genetic factors rather than environmental and behavioral factors.⁸ There are also several descriptions of neural basis and sexual orientation variability in GD^{7,9} This rapidly evolving topic has allowed us to understand how to approach individuals and offer support and management. The published literature has also tried to keep up with these rapid changes. Nonetheless, the current management of individuals with GD include gender-affirming surgery, which is technically challenging and should only be performed by specialized multidisciplinary teams. Our aim is to perform a bibliometric analysis of the literature, looking for the quality of the published literature that supports the current management as well as to make a critical review of the literature on the most discussed topics and to propose future trends in the field of GD.

Methods

Systematic Literature Search

A systematic search and critical review of the literature was performed in 2020 including articles published between January 1900 and December 2018 to perform a bibliometric analysis. The manuscript search was performed in the following databases: OVID, PubMed, EMBASE, Scopus, Web of Science, and Google Scholar. The medical subject headings (MeSh) terms used were: gender dysphoria, sexual nomenclature, surgery, and psychology. Due to the nature of the study, only articles published up to 2018 were included, owing to the fact that some articles published in 2019 are still in press or under review.

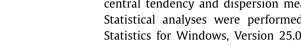
An initial screening of all references excluded duplicated publications. A second screening was performed assessing abstracts; these were read to categorize which topic they corresponded to. Manuscripts that discussed topics that were not directly related to GD were excluded. The level of evidence was defined by classifying manuscripts according to the type of methodology. In case of being considered nonscientific literature, a description of the text type was assessed in order to classify it. The categories and level of evidence reviews were carried out by four of the authors, and disagreements were resolved by consensus between them.

Data Analysis

Analysis was then performed, and citation counts per manuscript were taken from Scopus, Web of Science, and Google Scholar. The highest citation count per manuscript was selected. Citation data extraction was performed on December 20th 2019.

For the present study, we used a recently described impact index analysis (IIA) in order to adjust the citation



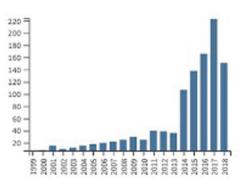


Results

A total of 1,239 manuscripts were identified. After the screening process and final review 1,041 manuscripts were selected. The average number of citation counts per year per manuscript was 1.84 (interguartile range [IQR] 0-2.33). The average impact index was 47.8 (IQR 20-111.6). The median for total citations per manuscript was 3 (IQR 0-33.1), and the highest number of citations per manuscript was 484, corresponding to the term consensus statement on 6 of intersex disorders, which also had the highest average per year, with 37.2 citations.¹¹ The results of elements and citations published every year are shown in **Figure 1**. The year with the highest number of publications was 2017, with 197. The Archives of Sexual Behavior was the journal with the highest number of publications, with 118 during the study period. Publication trends have shown a steady increase in the amount of publications per year (~Fig. 1).

Most publications focus on the psychological aspects of GD, and there is a significant number of manuscripts related to social and anthropological issues (the percentage of articles and their main topics are presented in **-Table 1**). Regarding the trends of recent years, in the last 5 years, 215 (31.3%) articles were written related to the psychiatric and psychological aspects of the condition. Publications about surgical management amounted to 62 (9%), and those discussing endocrinology totaled 63 (9.2%).

We also noted that most articles have a low level of scientific evidence. Of the 1,041 articles evaluated, we found that 256 (27%) were cross sectional studies, 255 (26.9%) were



Citations published by year

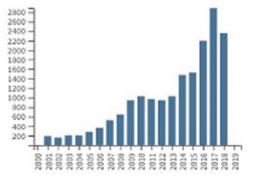


Fig. 1 Elements published and citations every year.

counts based on the time since publication to reduce the effect of time available since publication and the influence in the community.¹⁰ Search results were graphically represented using VOSviewer version 1.6.8. (Centre for Science and Technology Studies [CWTS], Leiden, Netherlands). A descriptive analysis of interest variables was carried out. For quantitative variables, the Kolmogorov-Smirnov normality test was applied to define distribution; the corresponding central tendency and dispersion measures were reported. Statistical analyses were performed with the IBM SPSS Statistics for Windows, Version 25.0 (IBM Corp., Armonk, NY, USA).

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Table 1 Distribution of main topic discussed per manuscript

	N = 1,041
	(n / %)
Psychiatry/psychology	349 (35.4)
Management	139 (14.1)
Surgery	96 (9.7)
Andrology/sexology	88 (8.9)
Endocrinology	86 (8.7)
Sociocultural/anthropology/psychosocial	77 (7.8)
Physiology/biology/genetics	56 (5.7)
Other	37 (3.8)
Epidemiology	30 (3)
Ethology	18 (1.8)
Ethical issues	10 (1)

topic reviews, 73 (7.7%) were case reports, 72 (7.6%) were prospective cohorts, 63 (6.6%) were cases and controls, 49 (5.2%) were retrospective cohorts, 42 (4.4%) were systematic reviews, 35 (3.7%) were case series, 16 (1.7%) were letters to

the editor, 5 (0.5%) were qualitative studies, 2 (0.2%) were clinical trials, and 80 (8.4%) were classified as others, among which are chronicles, essays, and opinion articles, among others.

► Figures 2 and 3 were created using the VOSviewer term co-occurrence network. In these figures, each circle represents a term. The size of a circle indicates the number of publications that have the corresponding term in their title or abstract. Terms that co-occur a lot tend to be located close to each other in the visualization. In **Figure 2**, the main cluster corresponds to the word dysphoria and its associations with other terms. It demonstrates a close relationship with terms such as *psychiatry*, *concept*, *respect*, *implication*, and a considerable distance from terms such as hormonal treatment, surgical treatment, and practice care. The colors indicate that towards the blue area are terms related to childhood and adolescence; red corresponds to human or social study areas, such as *literature* and *history*, but also it also includes terms such as *psychiatry* and *guideline*; in the green area, there are terms related to endocrinology, like hormonal treatment, testosterone, and transsexual; and, finally, in the yellow area, there are various terms that are related and overlap transversely to other clusters, such as man, woman, symptoms, and analysis.

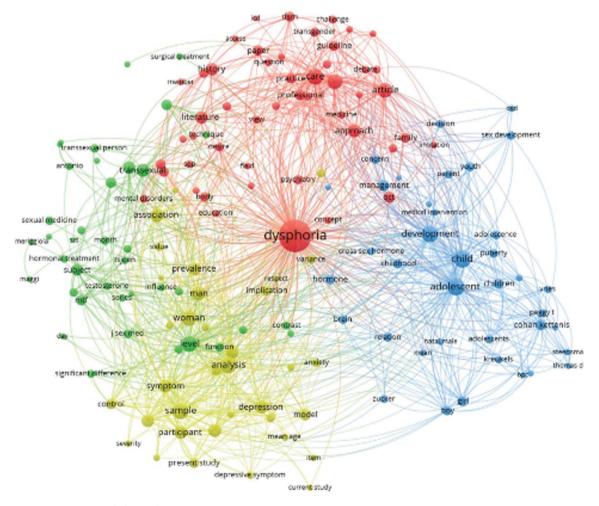


Fig. 2 Co-occurrence network figures by term.

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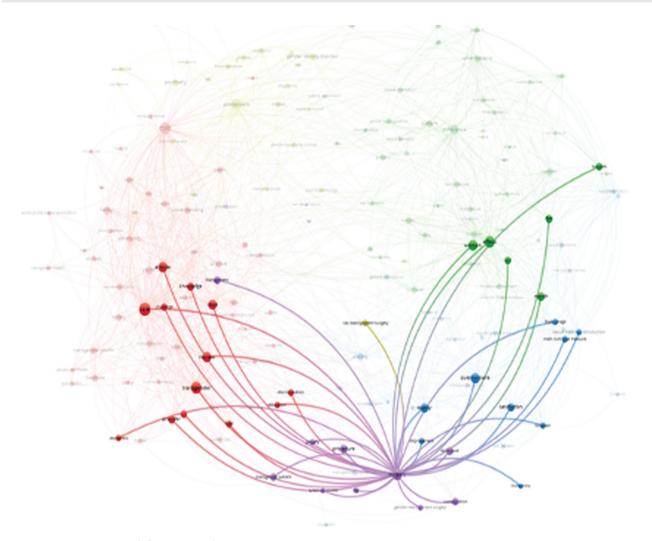


Fig. 3 Co-occurrence network figures using the term: "surgery".

In **Figure 3** we clustered results around the topic "surgical treatment" and found that there is poor publishing correlation to other topics.

Discussion

Historically, the first reference to GD was made in 1966, and it was in a book written by Harry Benjamin,¹² who was the first widely known physician (endocrinologist and sexologist) recognized for his work on transgender people. In this book, multiple topics related to etiology, physiology, general management, and hormonal and surgical treatments were addressed. At that point in time, the definition of dysphoria has not been used, but later, during the early 70's, the term gender dysphoria syndrome was used to describe transsexualism in addition to other gender identity disorders.¹ This terminology continues to be valid to date. Nonetheless, from a psychiatric perspective, there have been great advances since the update of the Diagnostic and Statistical Manual of Mental Disorders, fourth edition (DSM-IV) to the last DSM-V, in which it was decided to stop using the term gender identity disorder and only use gender dysphoria, thus emphasizing that it is not in itself a mental disorder.^{13,14} We found that

35.4% of the total articles reviewed correspond to the psychiatry/psychology category. Our results show that the topic that had been assessed in greater depth in most of the publications is psychiatry and its relation to the changes in terminology.

These individuals are known to carry a high emotional burden due to external factors (discrimination, value judgments, isolation), which puts them at risk of developing psychiatric illness;^{15,16} many adults face these challenges with social support, but it is known that social and family support is very limited in childhood and adolescence.¹² A higher rate of psychopathology has been found among groups of trans individuals compared to their cis counterparts.¹⁷ Some studies recognize the high prevalence of suicide among transgender individuals, and a higher risk or life-threatening behavior has been observed in the youth population.^{18,19} Gender dysphoria groups showed elevated levels of depressive distress compared to non-gender minority respondents, and this behavior was also seen when evaluating symptoms of anxiety.²⁰ These individuals, in addition to having a greater chance of presenting these conditions, also have a greater possibility of being diagnosed but not being adequately treated or having unmet needs of mental health care.²¹ Other psychiatric disturbances, such as eating disorders, are prevalent in these communities. Standard screening for these symptoms should be considered.^{19,22}

More recently, there has been an increasing interest in publications about gender-affirmation procedures and therapies.^{23–25} An increment trend of publications about gender-affirmation surgery have been identified over recent years. The first surgical technique for gender-affirmation surgery in an individual with GD was described in 1948 by Harold Gillies and RJ Harrison.²⁶ The goals of surgical male-to-female genital surgery have been reported with general successful outcomes by the minimization of complications and the creation of a functional and aesthetically perineo-genital complex, feminine in appearance and functional to allow regular sexual intercourse and to achieve orgasm.²⁷ Nonetheless, our results put in evidence that the current surgical techniques lack good quality research, and they are supported with lowquality literature in this field. The published literature also lacks long-terbm follow-up for the relatively recent proposed surgical techniques. This needs to be acknowledged by surgeons involved in GD surgery. For female-to-male genital affirming surgery, the operative procedures are usually performed in different stages, including hysterectomy-ovariectomy and genital transformation that includes a vaginectomy, scrotoplasty, penile reconstruction and testicular prosthesis delayed implantation.²⁸ Based on our bibliometric analysis, there is a limited amount of literature evaluating the physiologic implications of these procedures.

The average number of citations per year per manuscript was 1.84 (IQR 0–2.33), which indicates how small the target of these publications is, compared to other subjects. Considering the prevalence of this condition in the general population, it is clear when comparing the scientific evidence in other research categories,²⁹ that there is an absence of interest in the replication of this information in scientific fields.

There is an overwhelming amount of literature regarding GD; however, in the medical field, it is only from the 1970s on that studies began to be published, making GD a very recent topic. Previous approaches are made from a social and human perspective, but not with the intention of understanding this condition physiologically. Also, there is confusion in the literature about the current terminology used by GD individuals and how this spectrum needs to be taken into consideration at the moment of surgical intervention. The medical field has delayed the study of this condition and, therefore, of the optimal treatment of these individuals. This is why the scientific study of this condition and its treatments are still in an early exploration phase. It is hard to believe that 50 years after the start of literary publications on this topic, we still experience a lack of evidence. This situation might be rare in relation to other medical conditions, which leads us to wonder if this is indeed because of an inherent difficulty due to the complexity of this condition or a lack of interest in the subject and limited investments in this type of research; finally, this explains the evident lack of agreement in the medical management of this condition that persists through the years.

Conclusion

It is essential to be aware of the lack of thoroughness and rigor in the literature regarding GD, which prevents the acceleration of knowledge about this entity, knowing results of high methodological quality on which medical practice in this field could be based. The present study results show that the quality of evidence of most of the studies is low. About 16.1% are case reports and case series and 26.9% correspond to topic reviews.

Concerning gender-affirmation procedures, there is a gap in high-quality literary evidence that supports the emerging expansion of medical practices and the monitoring of their possible complications in the long term. Furthermore, there is also a need to acknowledge the differences between patients, knowing their history, and making case-by-case decisions based on long-term evidence that mitigates adverse events.

Conflict of Interests

The authors have no conflict of interests to declare.

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