Worldwide panorama of bruxism and patients with disabilities: a bibliometric analysis in PubMed (1965-2021)

Panorama mundial do bruxismo e pacientes com necessidades especiais: uma análise bibliométrica no PubMed (1965-2021)

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Abstract
This study aimed to present a bibliometric analysis about the central topic bruxism and disabled people in a global panorama through PubMed database (1965-2021). It was included in this study only journal publications in English. Letters to editor, short communication, articles irrelelative to people with disabilities and bruxism or articles with unavailable abstract or full text were not included. The initial search was performed by three independent investigators who assessed the title, abstract and main text to determine the included or excluded articles. It was observed that the tendency of publications was rising, with an average of 1.9 per year. There were 27 countries that have contributed to publications on the studied field and the most productive ones are United States (28.2%) and Brazil (20.0%). The most productive authors in the field are Brazilian and among the most productive institutions, 4 are Brazilian and 3 are North American. Case reports were the most frequent type of article (28.1%), followed by cross-sectional studies (24.5%) and literature reviews (12.7%). The most productive journal in the studied field were Special Care in Dentistry (12.7%), followed by Journal of Oral Rehabilitation (3.6%). Brazil has a great scientific contribution to the field with a relevant number of publications, leading authors and institutions. There is a lack of high-quality evidence involving the main topic, reinforcing that further interventional studies and controlled trials must be conducted.

Keywords: Bruxism; Disabled persons; Bibliometrics.

Resumo
O objetivo deste trabalho foi apresentar uma análise bibliométrica sobre o tema central bruxismo e pacientes com necessidades especiais em um panorama global utilizando a base de dados PubMed (1965-2021). Foram incluídos apenas artigos no idioma inglês. Cartas ao editor, comunicações breves, artigos fora da temática proposta ou com o resumo ou texto completo indisponíveis não foram incluídos. A busca inicial foi realizada por três examinadores independentes que acessaram título, resumo e texto completo dos trabalhos a fim de incluí-los ou não no estudo. Foi observada uma tendência crescente no número de publicações, com média de 1,9 artigos por ano. Vinte e sete países contribuíram com publicações na temática, sendo os Estados Unidos (28,2%) e o Brasil (20,0%) os países mais produtivos. Os autores mais produtivos na área estudada são brasileiros e entre as instituições mais produtivas, 4 são brasileiras e 3 norte-americanas. Relatos de caso foram os artigos mais frequentes (28,1%), seguido de estudos transversais (24,5%) e revisões de literatura (12,7%). As revistas científicas que mais publicaram na temática estudada foram a Special Care in Dentistry (12,7%) e Journal of Oral Rehabilitation (3,6%). O Brasil mostrou-se com grande contribuição científica na temática estudada na presente revisão bibliométrica, apresentando relevante número de
Bruxismo puede ser definido como ‘una actividad muscular repetitiva caracterizada por clenching o grinding de los dientes y/o por bracing o thrusting de la mandíbula’. Bruxismo tiene dos manifestaciones circadianas distintas: puede ocurrir durante el sueño (indicado como bruxismo durante el sueño - SB) o durante la alerta (indicado como bruxismo durante la alerta - AB)’ (Lobbezoo et al., 2013).

Hay un amplio rango de prevalencia de bruxismo en los niños, siendo observado una varianza de 3.5 a 49.6% (Manfredini, Restrepo et al., 2013; Machado et al., 2014). En adultos, la prevalencia de SB varía de 8.0 a 31.4%, mientras que AB varía de 22.1 a 31% (Manfredini, Winocur et al., 2013). Relacionado a pacientes con deterioro cognitivo, se encontró que ha sido probado por varios estudios que el bruxismo es más prevalente en esta población (variendo desde 42.0 a 74.0%) (López-Pérez et al., 2007; Lai et al., 2021; Cabrita et al., 2022) y también puede ser asociado al uso de medicamentos con efectos centrales (Ortega et al., 2014).

La fiabilidad de la prevalencia de bruxismo es pobre desde estos datos son basados en informes de los individuos y es difícil aplicar patrones diagnósticos basados en informes de los individuos, exclusivamente, fue probado por Maluly et al. (2013). Los autores hallaron una prevalencia de SB de 12.5% utilizando cuestionarios, contra 5.5% confirmado por el examen polisomnográfico en una población adulta, siendo evidente la limitación de los informes de los individuos como criterio diagnóstico (Maluly et al., 2013).

En el otro lado, Restrepo et al., reafirmaron que los informes de los individuos son útiles como criterios diagnósticos en los niños, tanto en el consultorio y para investigación científica. Los informes de los individuos han no presentado superior precisión de los examen polisomnográficos comparados con los informes de los individuos en este estudio (Restrepo et al., 2017).

Para operationalizar el diagnóstico de bruxismo, Lobbezoo et al. (2013) propusieron un sistema de gradación diagnóstico de ‘posible’, ‘probable’ y ‘definitivo’. En todos los niveles es necesario un informe de los individuos, comúnmente presentado por padres o cuidadores de los niños y pacientes con necesidades especiales. En el primer nivel de diagnóstico ‘posible’, solo un informe de los individuos es necesario. El segundo nivel ‘probable’, una asociación entre informes de los individuos y evaluación clínica es realizada. Finalmente, el último nivel ‘definitivo’ debe incluir un examen polisomnográfico, considerado el estándar en la población adulta.

Un análisis detallado de los antecedentes y los hallazgos clínicos son altamente recomendados para el diagnóstico de bruxismo en los niños. Síntomas de bruxismo son desgaste dental, sonidos durante el sueño (para SB), sensibilidad a la hiperactividad dental, fatiga muscular, indentación de la lengua y dolor de cabeza. Es importante determinar los hábitos orales y su frecuencia e intensidad, así como los períodos de grinding and/or
clenching teeth (Emodi-Perlman et al., 2012). Furthermore, diagnose which forms of bruxism is occurring is needed, during sleep (SB) or during wakefulness (AB), since etiologic factors involved are different in which condition. Both, SB and AB are modulated by central nervous system neurotransmitters (Bayar et al., 2012).

Huang et al. have described SB etiology via central pathways. This condition involves especially dopaminergic system leading to higher cortical activity, higher heart frequency, increased muscle tone and involuntary jaw excursions (Huang et al., 2014).

Many factors may be associated with bruxism including genetics basis, stress and anxiety, obstructive sleep apnea syndrome and gastroesophageal reflux (Bulanda et al., 2021). Furthermore, it has been described by literature a correlation between bruxism and children diagnosed with attention-deficit hyperactivity disorder (ADHD) (Bulanda et al., 2021), children with neurological impairment (Ortega et al., 2014; Cabrita et al., 2022) and using drugs with central effects (Ortega et al., 2014; Bulanda et al., 2021).

Disabled people represent a population with a higher risk to bruxism development and also a challenge to clinical management. It has been recorded that people with cerebral palsy, Rett syndrome, Down syndrome and autism spectrum disorder (ASD) are more likely to present bruxism compared to general population (Carneiro et al., 2020; Bagattoni et al., 2021; Mahdi et al., 2021; Cabrita et al., 2022).

Bibliometrics is an important tool for the quantitative analysis of the productivity of scientific research and also can be applied to track development trends, access the influences of publications and compare academic performance between different countries related to central topic (Yao et al., 2018).

There is a tendency of increasing number of dental journals, as well as, the number of published articles (Jayaratne and Zwahlen, 2015). Global trends in several dentistry fields have been revealed by bibliometric analyses. Moreover, can be applied to determine top-cited articles in a field (Kammer et al., 2021) or even to honor a remarkable author (Santos et al., 2021).

As far as we know, there is only one published bibliometric analysis involving bruxism (Ribeiro-Lages et al., 2021) and two involving disabled people and dentistry (Kammer et a., 2021; Gutierrez et al., 2021). Publications on bruxism, especially in children and adolescents, have increased over past 20 years (Ribeiro-Lages et al., 2021). Despite bruxism being worldwide studied and strongly related to a higher prevalence in disabled patients compared to general population, no bibliometric data have been published to date on central topic bruxism and disabled people.

The current study aimed to present a bibliometric analysis in PubMed of the published papers regarding the central topic bruxism and patients with disabilities in a global panorama.

2. Methodology
2.1 Search strategy

The bibliometric data were acquired through PubMed database, since it is a database widely used and internationally recognized. For this bibliometric review, it was performed articles search related to bruxism and people with no period restriction. Search strategy was conducted by using “bruxism” and “disabled people”, including their related terms and three specific disabilities. The search was performed on January 26, 2022. The keyword regarding bruxism was sleep bruxism. The keywords related to people with disabilities were: Down syndrome, cerebral palsy, disabled persons, disabled children, autism spectrum disorder, genetic diseases, intellectual disability, developmental disabilities and mentally disabled persons. The final search query was set as follows: ((Down syndrome) OR (cerebral palsy)) OR (disabled persons) OR (disabled children)) OR (autism spectrum disorder)) OR (genetic diseases)) OR (intellectual disability)) OR (developmental disabilities)) OR (mentally disabled persons)) AND ((bruxism) OR (sleep bruxism)).
It was included in this study only journal publications in English. Letters to editor, short communication, articles irrelelative to people with disabilities and bruxism or articles with unavailable abstract or full text were not included. The initial search was performed by three independent investigators who assessed the title, abstract and main text to determine the included or excluded articles (Figure 1).

2.2 Data analysis

The articles included in this study and categorized accordingly: year publication, leading universities and facilities, leading countries, leading authors, leading journals, keywords, study design and impact factor. First, data analysis was performed manual handling by two investigators (N.B.D. and G.M.G.) assessing full texts of the included articles. In case of some discrepancy, the third investigator (C.F.B) was consulted to compared the results and, if necessary, a discussion involving the three investigators was made. These data were brought in Microsoft Excel 2019 (Microsoft) to further statistical procedures. The main journals, countries, authors and institutions were classified according to the standard classification of the competition (SCR).

Data was also analyzed by software process (VOSview version 1.6.17) to create scientific landscapes and networks based on a plain text impoted from PubMed, according to the instruction of the software.

Impact factors (IF) of the journals were determined using Thomson Reuters in 2015 Journal Citation Reports (JCR). The study design was classified into the following categories: descriptive study, case report, case-control study, cohort study, cross-sectional study, randomized controlled trial, literature review and systematic literature review.

3. Results

After applied proposed search strategy a total of 185 articles were found ranging from 1965 (Adelson, 1965) to 2021 (Lansdown et al., 2022 - Epub 2021 Oct 27). Thus, there was no year restriction regarding inclusion in this study. Seventy-five articles were excluded, being: not English (n=8), letter to editor or short communication (n=3), articles irrelative to disabled
people and bruxism (n=45) and unavailable for abstract or full text (n=19). Finally, 110 articles were included for full reading and bibliometric analysis (Figure 1).

3.1 Articles published per year

From 1965 to 2021, a total of 110 articles related to disabled people and bruxism were indexed in PubMed, with an average of 1.96 per year. In this period, the tendency of publications in general was rising. The highest number of publications occurred in 2021, with 9 published articles (Figure 2).

![Figure 2 - Number of publications per year.](image)

Source: Authors.

3.2 Country and publication

There were 27 countries that contributed to publications on the studied field as shown in the density word map (Figure 3 A). Among displayed countries, only 4 countries had over 5 publications. United States (US) was the most productive country with 31 publications (28.2%) followed by Brazil with 22 publications (20.0%) and Italy with 7 publications (6.36%). The top 10 most productive countries are shown in Figure 3 B.
3.3 Authors contribution

Within the 110 articles included, a total of 482 authors have been contributed. The top three productive authors in the field of bruxism and disabled people are shown in Table 1. There are 8 authors in the top three list because 6 authors published an equal number of articles. Santos, MTBR was the most productive author in the field with 6 publications, followed by Ortega, AOL with 5 publications, both from Brazil and Cruzeiro do Sul University.
Table 1 - Top 3 productive authors in the field of bruxism and disabled people.

| Author          | Number of publications | Affiliation                     |
|-----------------|------------------------|---------------------------------|
| Santos, MTBR    | 6                      | Cruzeiro do Sul University      |
| Ortega, AOL     | 5                      | Cruzeiro do Sul University      |
| Ferri, R        | 3                      | University of Rome              |
| Jankovic, J     | 3                      | Baylor College of Medicine      |
| Machuca-Portillo, G | 3                  | University of Seville           |
| Silvestre, FJ   | 3                      | University of Valencia          |
| Ciamponi, AL    | 3                      | University of São Paulo         |
| Percy, AK       | 3                      | Baylor College of Medicine      |

Source: Authors.

3.4 Institutions involved

The top three institutions that have contributed in publishing papers on oral health and cerebral palsy are presented in Table 2. The most active institutions were *Cruzeiro do Sul University* in Brazil (4 publications) and *University of São Paulo* in Brazil (4 publications), followed by *Baylor College of Medicine* in the US (3 publications) and *Federal University of Minas Gerais State* (3 publications) in Brazil. A total of 86 organizations have published only one article in the field. Among the top three most active institutions, four are in Brazil, three are in US, one is in Poland and Spain.

Table 2 - Top 3 institutions that published in the field of bruxism and disabled people.

| SCR* | Institution                                | Country   | Documents |
|------|--------------------------------------------|-----------|-----------|
| 1st  | Cruzeiro do Sul University                 | Brazil    | 4         |
| 1st  | University of São Paulo                    | Brazil    | 4         |
| 2nd  | Baylor College of Medicine                 | US        | 3         |
| 2nd  | Federal University of Minas Gerais State   | Brazil    | 3         |
| 3rd  | Boston Children’s Hospital                 | US        | 2         |
| 3rd  | Federal University of Juiz de Fora         | Brazil    | 2         |
| 3rd  | Indiana University                         | US        | 2         |
| 3rd  | Medical University of Warsaw               | Poland    | 2         |
| 3rd  | University of Seville                      | Spain     | 2         |

*SCR, standard competition ranking. Source: Authors.

3.5 Keywords

There were 402 keywords in the 110 articles. The visualization of keywords is shown in Figure 4. This analysis was performed based on the terms extracted from the title and abstract fields of retrieved publications; a number of 51 terms met the threshold with a minimum number of 5 occurrences. The term with the highest frequency was human (n=102), followed by female (n=66), child (n=63) and bruxism (n=54).
Figure 4 - Word frequency analysis. The density visualization map of the top 25 keywords. The larger circle size indicates higher occurrence.

Source: Authors.

3.6 Publications analysis based on study design and journals

Among 110 included studies, case reports were the most frequent type of article (31, 28.18%), followed by cross-sectional studies (27, 24.54%) and literature reviews (14, 12.72%). All the retrieved documents were published in 75 different journals. The top 3 most productive journals are presented in Table 3, comprising 22.72% (25/110). The most productive journals in this field were Special Care in Dentistry (n=14), Journal of Oral Rehabilitation (n=4), Medicina Oral Patologia Oral y Cirurgia Bucal (n=4) and Journal of Applied Behavior Analysis (n=3).

Table 3 - Top 3 journals with publications on bruxism and disabled people.

| SCR* | Journal                                               | Documents | % n=110 | IF*    |
|------|-------------------------------------------------------|-----------|---------|--------|
| 1st  | Special Care in Dentistry                             | 14        | 12.72   | 1.28   |
| 2nd  | Journal of Oral Rehabilitation                        | 4         | 3.63    | 3.837  |
| 2nd  | Medicina Oral Patologia Oral y Cirurgia Bucal         | 4         | 3.63    | 1.596  |
| 3rd  | Journal of Applied Behavior Analysis                  | 3         | 2.72    | 2.108  |

*SCR, standard competition ranking; *IF, impact factor. Source: Authors.

4. Discussion

Bibliometric analysis is current used to reveal the global panorama in a research field. In this study, the relationship between bruxism and patients with disabilities were analyzed using a bibliometric approach. To the best of authors’ knowledge, this is the first bibliometric study involving bruxism and patients with any kind of disability.

Previous studies have pointed to a tendency of increasing number of publications over time, specially over the past
two decades (Jayaratne and Zwahlen, 2015; Celeste et al., 2016; Ribeiro-Lages et al., 2021). These findings corroborate with our data, although our results presented a less expressive absolute number, being 9 published articles, the highest number reached in a single year (2021).

Less expressive absolute numbers were also observed by Gutierrez et al. in recent bibliometric analysis involving patients with cerebral palsy and dentistry (Gutierrez et al., 2021). It was verified that scientific literature raised all over the years, although, a maximum number of published papers was 17 in 2018. On the other hand, regarding bruxism, literature shows a growth of publications accompanying more expressive absolute numbers compared to people with disabilities topic. According to Ribeiro-Lages et al. (2021), a significant turn point on publication numbers was found over the past two decades, demonstrated by a growth rate of 189% in observational studies in the 2000’s. The authors suggest that it may be explained since this condition is not yet fully know and a better understand about etiology, prevalence and its effects are needed.

The great amount of etiology and prevalence studies concerning bruxism topic reinforces that this area has been recent explored by researchers. Thus, observational studies higher production is in agreement with the level of knowledge in the field.

Over 1 billion people worldwide experience disability, which represent about 15% of the global population (WHO). Even though people with disabilities represent a significant group in world population, it is still needed to better explore this research field in dentistry. In an altmetric study that aimed to assess trends in pediatric dentistry, among 200 publications analyzed there were 13 studies (6.5%) related to patients with special needs (Martin et al., 2021).

Perazzo et al. (2019) have analyzed the top 100 most-cited papers in pediatric dentistry journals and observed the most cited area was cariology, followed by dental growth and development and behavioral and epidemiological science. Other bibliometric analysis demonstrated a minor contribution of papers including patients with special needs (Bhalla and Chockattu, 2020). These data can explain the low absolute number of publications in the researched topic found in this study.

However, the research field regarding people with disabilities has become important not only scientifically, but socially. As verified by Kammer et al. (2021) in a bibliometric overview of the 100 most-cited papers in dentistry for individuals with neurodevelopmental disorders, all continents presented at least one article on the list (Antarctica excepted). The authors reinforce the relevance of this theme in a global level, even in a scenario where more research is required.

In this study, the most productive countries were US (31 publications, 28.2%), Brazil (22 publications, 20.0%) and Italy (7 publications, 6.36%). It is expected that developed countries lead publications ranking, since its budget to research and new technologies are high. Incredibly, Brazil, which is a developing country (according to the definition of the United Nations) appears among the top productive countries in several scientific literature (Liu et al., 2020; Kammer et al., 2021; Ribeiro-Lages et al., 2021; Gutierrez et al., 2021). This data may indicate that scientific collaborations between researchers and institutions increase the possibilities of scientific production, although it is known that scientific collaborations network between Brazil and developed countries are below the average compared to other developing countries (Cross et al., 2017).

A recent bibliometric analysis about bruxism in children and adolescents pointed Brazil with the highest number of publications in this area (131 publications, 18.06%). The authors highlight that this condition may be seen as a prevalent health problem in Brazil explaining the research focus (Ribeiro-Lages et al., 2021). It was also observed that Brazilian authors compose the top authors, presenting more publications in bruxism field than authors from other nationalities.

Concerning most productive authors, our findings are in agreement with previous studies (Ribeiro-Lages et al., 2021; Gutierrez et al., 2021). Santos, MTBR and Ortega, AOL were the most productive authors in the field. The main institutions that published in the field of bruxism and disabled people were Cruzeiro do Sul University and University of São Paulo, both from Brazil. Secondly, it was mentioned Baylor College of Medicine and Federal University of Minas Gerais State, from USA and Brazil, respectively. Brazil is a country where many institutions and recognized postgraduate programs related to special care dentistry are placed (Faulks et al., 2012). Moreover, special care dentistry is recognized as a specialty, which may
influence the productivity and lead authors and institutions in Brazil.

There is a lack of high-level evidence in the studied topic, once case reports (28.1%), cross-sectional studies (24.54%) and literature reviews (12.72%) were the most frequent type of study designs. Similar results were observed in the literature regarding patients with disabilities (Kammer et al., 2021; Gutierrez et al., 2021) and bruxism (Ribeiro-Lages et al., 2021). The majority of observational studies related to bruxism investigated the etiology and risk or prevalence and incidence (Ribeiro-Lages et al., 2021), while for patients with disabilities epidemiology data about oral health status and prevalence of dental trauma were mentioned (Kammer et al., 2021). These results reinforce that controlled trials must be encouraged in order to optimize this research field and make available new high-quality evidence. In addition, it is important to highlight the low costs involved to conduct observational studies compared to controlled trials, which also may influence the obtained results.

Moreover, still regarding studies design, it is important to mention the challenge for researchers to conduct trials with patients with disabilities, since groups standardization and allocation may be an obstacle.

It is crescent the number of journals in dentistry, however, there is only a few specialized journals in patients with disabilities. Special Care in Dentistry is one of the main journals in dentistry publishing about patients with disabilities. It was not surprising that our findings showed Special Care in Dentistry as the most productive journal, which reinforces the great contribution of this journal in the studied field, also confirmed by others (Gutierrez et al., 2021).

This study presented some limitations since patients with disabilities may represent a wide variety of conditions. We minimize the bias including similar global terms such as “disabled persons” and “disabled children”, but also focusing in prevalent conditions related to a high prevalence of bruxism like “Down syndrome”, “cerebral palsy” and “autism spectrum disorder”.

In addition, we selected only PubMed publications since it is the main database publishing articles in dentistry and it is widely used, including by previous bibliometric analysis (Gutierrez et al., 2021; Liu et al., 2020). However, PubMed favors English publications which we may consider a bias. Brazil, as well as its authors and institutions were placed in this study with a great scientific contribution on the central topic even excluding journals and publications in Portuguese. The authors considered that it could underestimate the data presented in this bibliometric review.

There is a lack of high-level evidence regarding central topic, once in this study case reports were the most frequent type of article. This information leads us to reinforce that the level of knowledge in this area is scarce, since is has been mostly studied from 2000’s. Further investigations and controlled trials must be carried out in order to optimize the level of available evidence and to promote better health conditions to disabled people.

5. Conclusion

Our findings suggested that dental research focusing on disabled people and bruxism seemed to be worldwide interest. Brazil has a great scientific contribution to the field not only due to the relevant number of publications, but also presented the majority of leading authors and institutions publishing on the central topic. High-quality evidence involving disabled patients and bruxism is needed, so further interventional studies and controlled trials are encouraged to be conducted.

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