



Review article

Research productivity in catamenial epilepsy: A bibliometric analysis of worldwide scientific literature (1956–2022)

Diah Kurnia Mirawati^{a,*}, Nanang Wiyono^b, Muhana Fawwazy Ilyas^a, Stefanus Erdana Putra^a, Muhammad Hafizhan^a

^a Department of Neurology, Faculty of Medicine, Universitas Sebelas Maret, Surakarta, Indonesia

^b Department of Anatomy, Faculty of Medicine, Universitas Sebelas Maret, Surakarta, Indonesia

ARTICLE INFO

Keywords:

Bibliometric analysis
Catamenial epilepsy
Review
Scientific literature
Scopus
Trends

ABSTRACT

Objectives: To perform a bibliometric analysis as a comprehensive review of publications associated with catamenial epilepsy and discuss the current state of knowledge in the field.

Methods: Publications published between 1956 and 2022 were retrieved from the Scopus database. Bibliometric analysis was performed using the R package and VOSviewer to show the data and network of journals, organizations, authors, countries, and keywords. The analysis conducted on October 15, 2022, yielded a total of 320 refinement studies.

Results: The number of publications has escalated significantly, particularly in the last 20 years. Catamenial epilepsy-related publications originated mostly from medicine and other subject areas, with the United States having the largest publication output. Collaboration is low at the author, organizational, and national levels, especially in the Asian continent. Publications remain scarce, particularly on practice guidelines, risk assessment, and medication-related research. Based on a keyword analysis, a bibliometric analysis identified possible themes for future investigation.

Conclusion: Catamenial epilepsy-related literature is crucial but still insufficient, and further studies are required.

1. Introduction

The prevalence of epilepsy is high and increasing worldwide. A systematic review revealed that the annual incidence of epilepsy is approximately 50 % for men and 46 % for women [1]. Additionally, epilepsy affects approximately 1.7 million women in the United States (US) [2], and half of epileptic women worldwide are between the ages of 15 and 49 years [3]. A breakdown of common epilepsy syndromes in women included catamenial epilepsy [3], idiopathic generalized epilepsy [4], cryptogenic localization-related epilepsy [5], PCDH19-related epilepsy [6], childhood absence epilepsy [7], and photosensitive epilepsy [8]. Epilepsy in women—including catamenial epilepsy—requires special considerations [9].

Catamenial epilepsy is characterized by increased seizure frequency and the worsening of seizures during specific phases of the menstrual cycle among women with epilepsy [10]. The molecular pathophysiology of catamenial epilepsy remains unclear. However, cyclical changes in the circulating levels of sex hormones including estrogens and progesterone that have neuroactive effects, may play a central role in the development of catamenial epilepsy [2,10]. Subsequently, those particular hormones may also explain different

* Corresponding author.

E-mail address: diahkm@staff.uns.ac.id (D.K. Mirawati).

patterns of seizure distribution during the menstrual cycle of women with epilepsy [11–13]. C1: perimenstrual seizures are associated with an increase in seizure frequency during the menstrual phase and arise from a drastic decrease in both progesterone and estrogen levels. C2: periovulatory seizures are associated with a high seizure frequency in the ovulatory phase and are attributed to a high estrogen-progesterone ratio. Estrogen can exacerbate seizures, and progesterone (which is an anticonvulsant) levels are low during this phase, making it more susceptible to seizures. C3: luteal phase seizures occur during the luteal phase of the menstrual cycle, when progesterone levels are high but estrogen levels low. A reduction in progesterone levels premenstrually and reduced secretion during the luteal phase are implicated in catamenial C1 and C3 patterns.

The prevalence of catamenial epilepsy remains controversial. Herzog et al. (1997) indicate that the prevalence of catamenial epilepsy reaches 42.3 % among women with epilepsy; within this percentage, ovulatory cycles are responsible for 35.7 % of cases (C1), while 28.5 % of cases are attributed to C2 cycles [10]. Anovulatory cycles—specifically C3—account for 41.4 % of cases. Meanwhile, Herzog et al. (2015) provide slightly different prevalence figures based on the patterns of catamenial epilepsy: 39.8 % for the C1 pattern, 33.9 % for the C2 pattern, and 47.1 % for the C3 pattern [14]. Several earlier studies have reported even higher prevalence rates, ranging from 63 % to 78 %; however, these studies have focused on comparing seizures during the perimenstrual phase versus other phases of the cycle [15–17].

One of the United Nations' Sustainable Development Goals for 2022 is good health and well-being (Goal 3); it encompasses a broad spectrum of health-related targets, including reducing maternal mortality, preventing diseases, and ensuring access to healthcare services for everyone. Research in the field of epilepsy—particularly catamenial epilepsy—is crucial for achieving these global health objectives. This study presents a bibliometric analysis of catamenial epilepsy, which to the best of our knowledge, has never been conducted in the literature. The aims of this research are to review the catamenial epilepsy literature by providing a thorough bibliometric analysis to investigate the trend based on the annual number of publications; most contributing authors; contributing subject discipline; thematic maps; gaps and potential future study topics; countries and organizations with the largest contributions and their collaboration; and the literature that has citations. Additionally, this study aims to provide a comprehensive description of the current state of knowledge in the field of catamenial epilepsy.

2. Methods

Bibliometric analysis is a technique for discovering scientific trends and organizing research [18], assuring the information quality and establishment of the findings obtained [19]. It also enables the creation of a distinctive viewpoint from a sufficiently in-depth analysis. This inquiry, the Scopus database is used. Scopus is among the available databases with the broadest worldwide coverage of publications including journals, books, and conferences, as well as articles [20]. Scopus is also user-friendly and supports multiple software packages for obtaining bibliometric information (authors, titles, publication dates, references, abstracts, institutions, and countries) [21].

Fig. 1 depicts the processes of evaluating the collected documents [22]: (1) data collection by establishing search keywords to locate relevant literature according to the criteria in the Scopus database and refining the gathered archives; (2) data visualization by

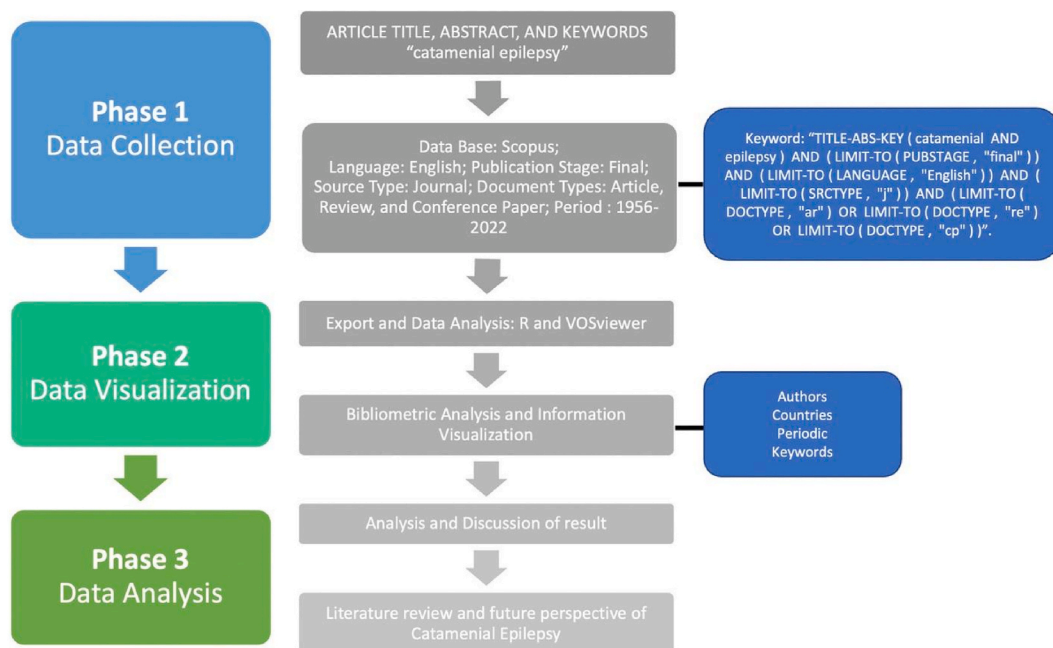


Fig. 1. The methodology phases on constructing bibliometric analysis.

exporting the obtained data to the R package and VOSviewer for bibliometric analysis and displaying the information; and (3) data analysis to identify the primary themes discussed.

The search was conducted on 15 October 2022, using the key phrase “catamenial epilepsy” The initial search revealed a total of 440 publications, which we restricted to those written in English and in which the publication stage was complete, the source type was a journal, and the document types an article, review, or conference paper. Non-English documents, articles in press, other sources (such as book and book series), and other document types (such as book chapter, letter, editorial, short survey, note, book, and erratum) were excluded from this analysis, and 320 publications met all of the requirements (published between 1956 and 2022). Acquired information is stored in RIS files and processed with Mendeley to complete the metadata. Then, data were validated and holes were filled. Afterwards, co-authorship and co-occurrence analysis were conducted and a network map was also developed based on citation examination.

The R package’s bibliometrics utility is designed for quantitative scientometrics and informetrics [23]. In addition, bibliometric technologies allow the categorization and analysis of vast quantities of historical data obtained from research undertaken during a certain period to extract information from the repository. Bibliometric and meta-analyses rely on quantitative techniques and can, therefore, avoid or mitigate bias, in contrast to systematic literature reviews, which typically rely on qualitative techniques and are susceptible to interpretation bias from scholars with diverse academic backgrounds [24]. The Bibliometrix R package was initially installed and loaded with R Studio. The Biblioshiny application was initiated by entering Biblioshiny() into the R console. Biblioshiny is a web application that gives non-programmers access to the R package Bibliometrix, of which numerous tools enable researchers to conduct in-depth bibliometric study [25].

VOSviewer was employed to build bibliometric networks. This software enables data extraction including authorship, journals, organizations, nations, and keywords. To view the existing associations among bibliometric data, outputs are presented as overlapping circles [26]. The distance between each circle represents the strength of the association between the terms displayed. Various hues are used to denote distinct phrase groups; moreover, the circle size is proportional to the phrases’ occurrence frequency [18]. Depending on the number of links, the number of clusters in each map might differ. Taking into account the associations between terms inside every cluster, the pertinent topics were identified and analyzed in depth. Subsequently, a comprehensive description of the current state of knowledge was provided based on the particular relevant topics in scope of catamenial epilepsy.

3. Results

3.1. Historical background

The initial research on catamenial epilepsy in all articles on the Scopus database revealed a 1948 article by the title “*L’épilepsie catameniale* [Catamenial epilepsy]” [27]. Although this article marked an early exploration of catamenial epilepsy, it was not included in the bibliometric analysis due to its non-compliance with the established inclusion criteria. Nevertheless, it is a historical milestone in the study of catamenial epilepsy, representing the early stages of research into this complex condition.

3.2. Publication trends

A bibliometric analysis was performed on 320 publications, mostly articles (61.4 %), reviews (36.1 %), and conference papers (2.5

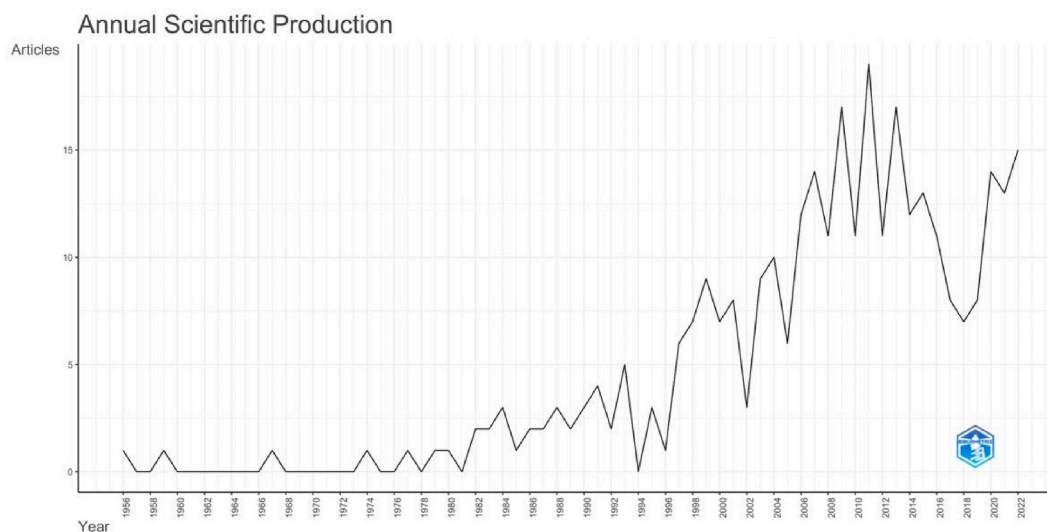


Fig. 2. Catamenial epilepsy research trends on a worldwide scale (1956–2022).

Fig. 2 depicts the research trends based on articles published annually. An upward trend was observed between 1982 and 2010; this period may have been associated with increased awareness, research funding, and advancements in diagnostic and analytical tools, which collectively may have contributed to the surge in publications. Subsequently, a slight decline was observed from 2012 to 2018, followed by the resumption of an upward trend in 2019. The year 2011 had the highest number of publications (19). Of the 320 articles pertaining to catamenial epilepsy, 239 (74.92 %) have been published in the last 20 years. This concentration of research in the recent past may be attributed to advancements in technology, increased understanding of the condition, and growing recognition of catamenial epilepsy as a distinct area of study. (At the time this study was conducted, 2022 was still in progress, and the number of publications for that year may have increased).

3.3. Contributing authors

Based on the most contributing authors, Reddy, D.S. from Department of Neuroscience and Experimental Therapeutics, Texas A&M University, College Station, United States is the author with the most contributions with 30 articles and a total of 1,660 citations. This demonstrates the substantial impact of his work on the scientific community and highlights his position as a leading authority in catamenial epilepsy research. His most influential article is about “Neurosteroid withdrawal model of perimenstrual catamenial epilepsy” [28], with 129 citations and the most recent article is related to “Neurosteroid replacement therapy for catamenial epilepsy, postpartum depression and neuroendocrine disorders in female” [29]. Herzog, A.G. and Harden, C.L. also contributed the most, with 20 and 10 articles, respectively. Their work has added depth to our understanding of catamenial epilepsy and its clinical implications. Fig. 3 shows a visual description of authors’ publications over the years.

3.4. Interdisciplinary areas

Catamenial epilepsy research originates from various subject areas, with “medicine” dominating the field with 258 publications (49.5 %). This predominance reflects the central role of clinical medicine in diagnosing, treating, and understanding the condition. This finding highlights the importance of medical professionals in shaping the trajectory of catamenial epilepsy research. “Neuroscience” follows closely with 158 publications, comprising 30.4 % of the research output; “biochemistry, genetics, and molecular biology,” with 41 publications (7.9 %); and “pharmacology, toxicology, and pharmaceuticals” with 37 publications (7.1 %). In addition, “psychology,” “nursing,” “agricultural and biological sciences,” “chemical engineering,” “chemistry,” “veterinary,” “arts and humanities,” “multidisciplinary,” “engineering,” and “social sciences” have contributed to it, although publications in these fields are still limited. While the contributions from these fields are currently more limited, they may offer fresh perspectives and insights into various aspects of the condition. Interdisciplinary collaboration among these fields can expand the breadth of catamenial epilepsy research, ultimately leading to a more comprehensive understanding of the condition and improved patient care.

An interesting aspect of the subject area is that the approach occurs not only in medicine with human subjects but also in veterinary medicine. Some articles that use the catamenial epilepsy approach to veterinary medicine are related to “Alternative therapeutic options for medical management of epilepsy in apes” published by the Journal of Zoo and Wildlife Medicine [30], “The influence of sex hormones on seizures in dogs and humans” by Veterinary Journal [31], and “Association between estrus and onset of seizures in dogs with idiopathic epilepsy” by the Journal of Veterinary Internal Medicine [32].

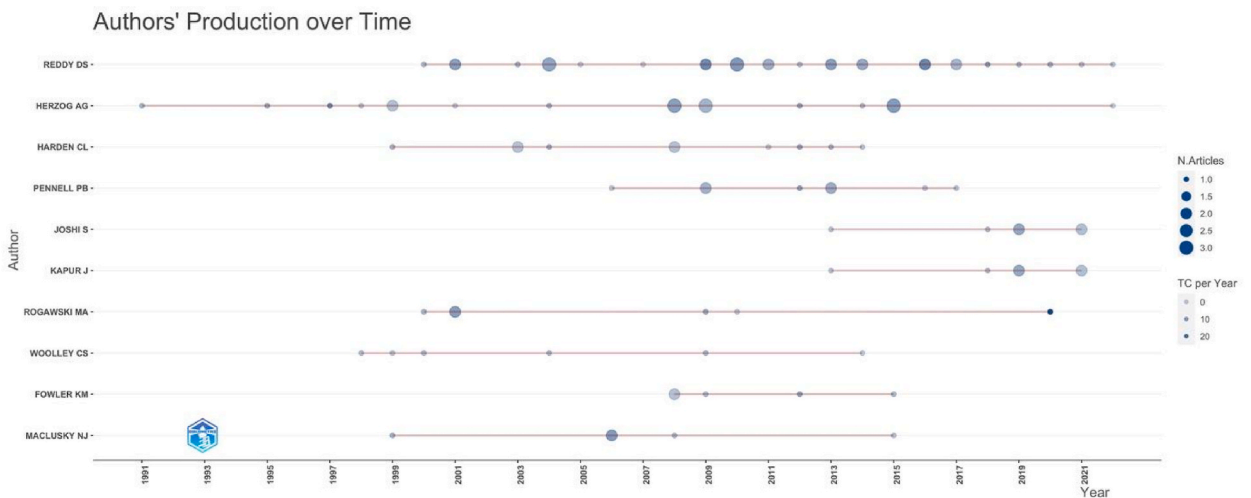


Fig. 3. Authors’ production on catamenial epilepsy research over time.

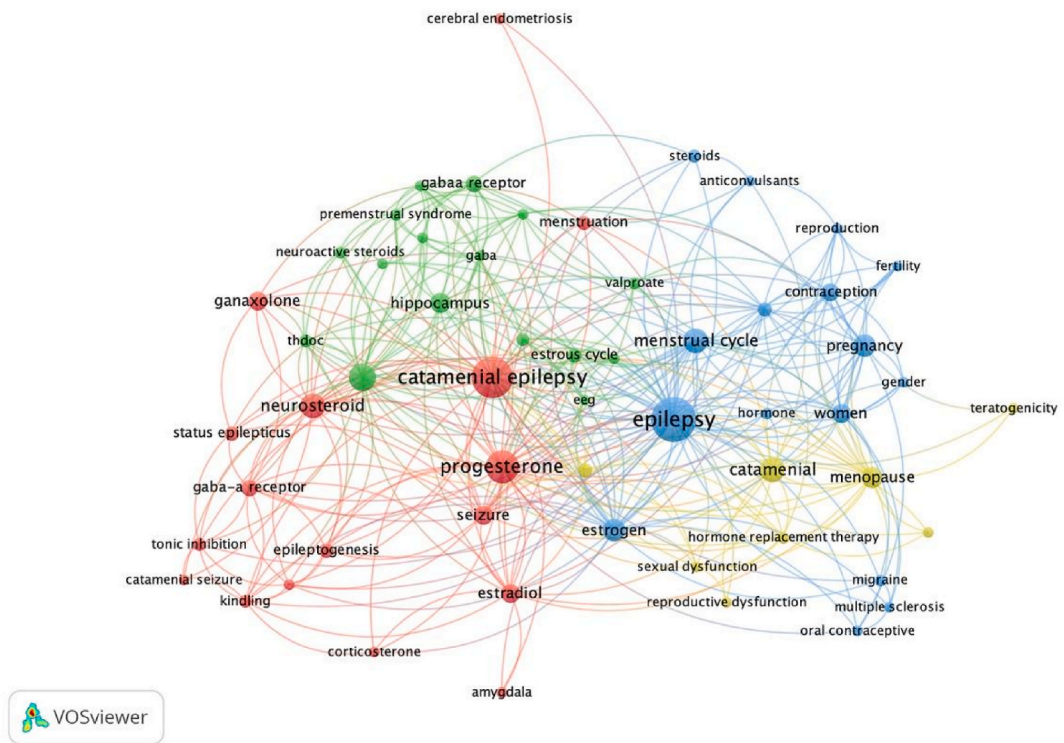


Fig. 5. Network visualizations of catamenial epilepsy-related publications.

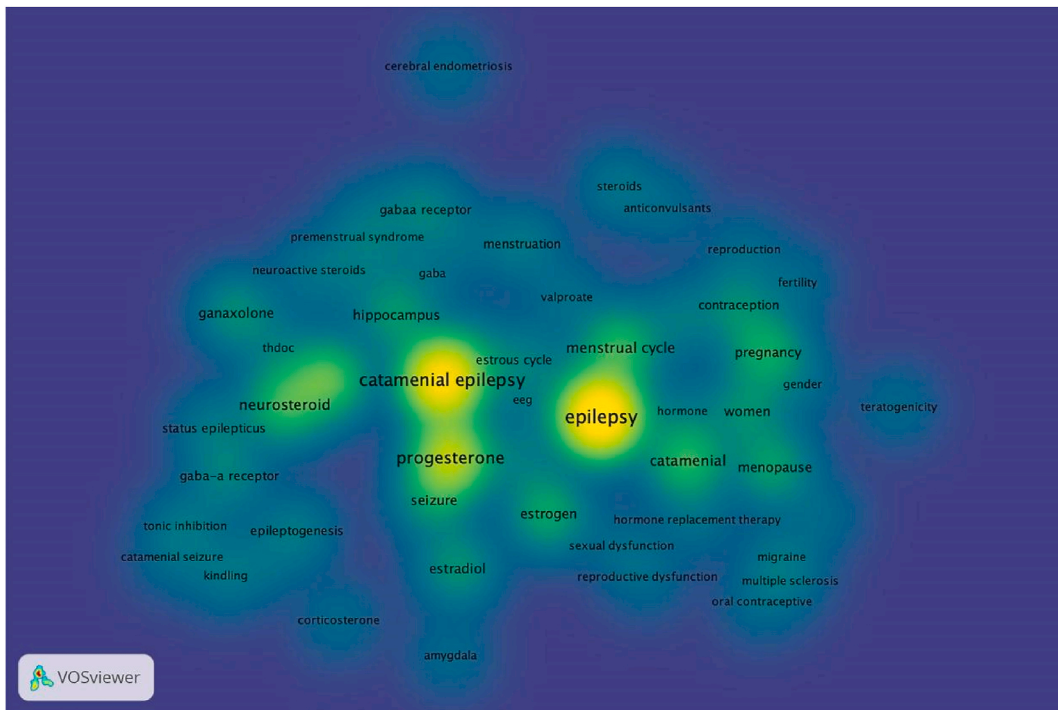


Fig. 6. Density visualizations of catamenial epilepsy-related publications.

Table 1
The result of cluster analysis on catamenial epilepsy publications.

Cluster	Most frequent keywords	Keywords (Number of Occurrence)
1st cluster (17 items)	catamenial epilepsy (76), progesterone (46), neurosteroid (22)	estradiol (12), seizure (12), ganaxolone (11), gaba-a receptor (8), epileptogenesis (6), menstruation (6), status epilepticus (6), kindling (5), tonic inhibition (5), amygdala (3), catamenial seizure (3), cerebral endometriosis (3), corticosterone (3), sex difference (3)
2nd cluster (15 items)	allopregnanolone (26), hippocampus (13), estrous cycle (6)	sex hormone (5), thdoc (5), neuroactive steroid (4), premenstrual syndrome (4), valproate (4), anxiety (3), depression (3), eeg (3), gaba (3), kainic acid (3), pregnanolone (3), puberty (3)
3rd cluster (13 items)	epilepsy (96), menstrual cycle (22), pregnancy (18)	estrogen (17), contraception (10), antiepileptic drug (6), steroid (5), migraine (4), anticonvulsants (3), fertility (3), multiple sclerosis (3), oral contraceptive (3), reproduction (3)
4th cluster (8 items)	catamenial (22), menopause (15), testosterone (6)	Hormone replacement therapy (4), perimenopause (3), reproductive dysfunction (3), sexual dysfunction (3), teratogenicity (3)

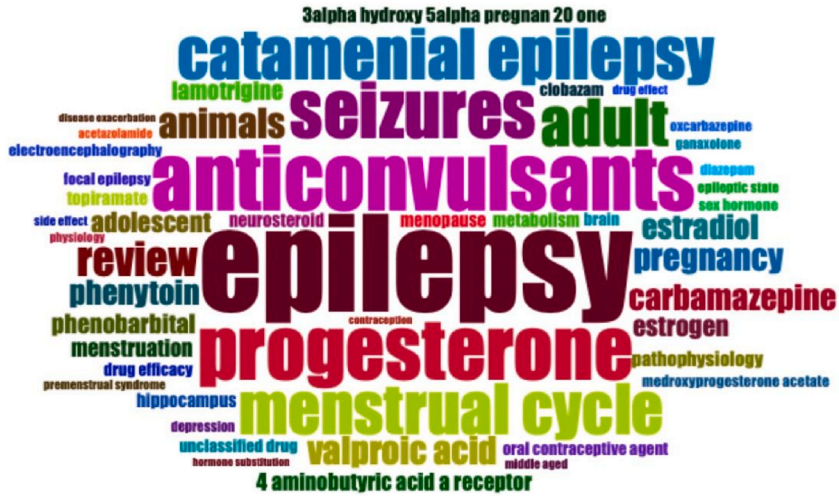


Fig. 7. Word cloud of the most frequent keywords on catamenial epilepsy research.

cloud on keywords with the most significant occurrence via Biblioshiny and obtained results similar to those of the VOSviewer analysis. This reinforces the significance of the identified keywords and their relevance in shaping the research landscape of catamenial epilepsy.

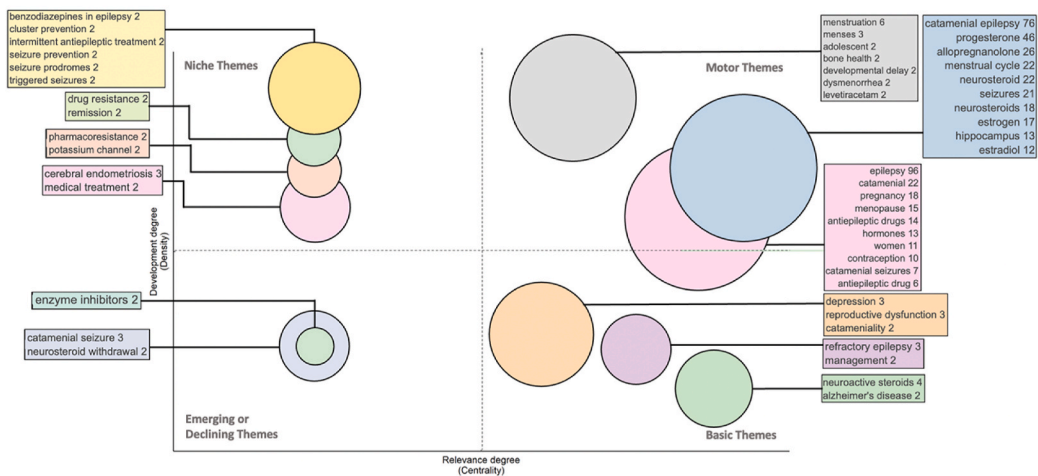


Fig. 8. Thematic map depiction of terms in catamenial epilepsy publications.

3.7. Thematic mapping

This study also included a thematic map depicting the keywords found in the literature on catamenial epilepsy. Themes are keyword clusters whose density and centrality can be used to map them as a two-dimensional image by organizing them into a single circle. The thematic map depicted in Fig. 8 classifies themes based on the quadrant in which they appear, beginning with motor themes in the upper-right quadrant and fundamental themes in the lower-right quadrant. These fundamental themes are the core components that provide the groundwork for further exploration, encompassing topics that are integral to understanding the condition. Emerging or vanishing themes occupy the lower-left quadrant, indicating areas that have either gained or lost prominence in recent years. This insight is crucial for identifying evolving research trends and recognizing themes that are becoming more prominent as well as those that may need further exploration or have become less relevant. Extremely specialized/niche topics occupy the upper-left quadrant, suggesting areas of research that are highly specialized and may be pursued by a select group of scholars. These topics represent cutting-edge areas of study, which—although niche—may uncover groundbreaking insights into catamenial epilepsy. In this study, which is illustrated in Fig. 9, we also presented trending topics or tendencies and the average years of study, offering a time-based perspective on the evolving landscape of catamenial epilepsy research. Analyzing trends and the average years of study helps researchers and practitioners stay current with the latest developments and align their efforts with the most relevant and up-to-date research in the field.

3.8. Global contributions

Additionally, we conducted a bibliometric analysis to assess and depict the research contributors' countries of origin. This assessment revealed important insights into the global landscape of catamenial epilepsy research. The distribution of research activity across different countries and regions sheds light on the international scope and collaboration in this field. Fifty-two countries were identified; however, only 24 matched VOSviewer's requirements (minimum number of documents is 3). The US has undertaken the most research, with 166 publications, followed by the United Kingdom (UK) with 36 publications and Italy with 22. The strong presence of the US in this area signifies the country's commitment to advancing the understanding of this condition and developing effective treatment strategies. This leadership role also positions the US as a valuable collaborator for researchers and institutions worldwide. The UK's contributions reflect its dedication to improving women's health and well-being, with a particular focus on addressing the unique challenges posed by catamenial epilepsy.

Publications on the Asian continent remain extremely scarce, with only 27: six for Turkey; four for China; three for Iran; two for South Korea, Japan, Malaysia, Palestine, and Saudi Arabia; and one for Sri Lanka, Thailand, Pakistan, and Hong Kong. This indicates a gap in research activity in this region, which presents an opportunity for future growth and collaboration. In the Southeast Asia region, only Malaysia and Thailand have published papers, while Indonesia has never published; this underlines the need for increased engagement and research initiatives in countries with lower representation to foster a more inclusive and global research community. Further, regarding the organizations with the most contributions, "Beth Israel Deaconess Medical Center" and "Texas A and M Health Science Center" have 22 and 18 publications, respectively.

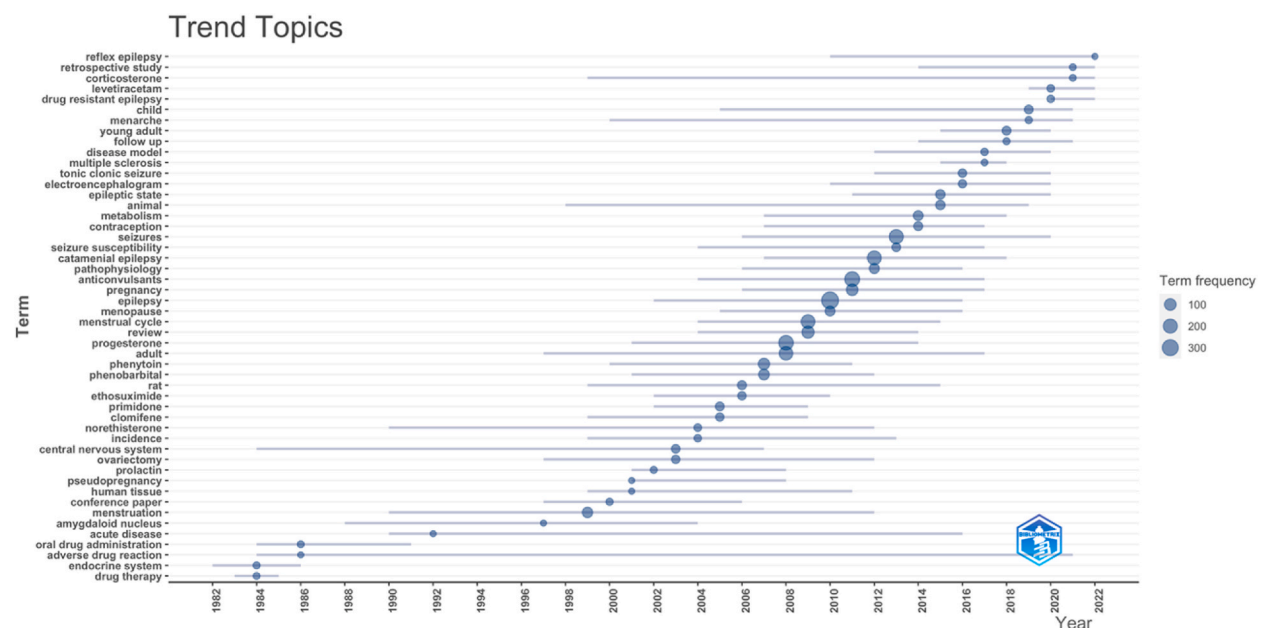


Fig. 9. Trend topics distribution on catamenial epilepsy studies.

3.9. Influential articles

Furthermore, Hosie et al.'s article by the title "Endogenous neurosteroids regulate GABA_A receptors through two discrete transmembrane sites," which was published in 2006 [33], has been cited 582 times, which makes it the most cited in its field. It is also considered the most influential and well-read publications due to its high average number of citations per year (36.375), which indicates that this article has maintained its relevance and continues to be a valuable resource for researchers and scholars in the field. The prominence of this publication suggests that it has served as a foundational reference, shaping the direction of subsequent research and contributing to a better understanding of the neurobiological mechanisms underlying catamenial epilepsy. Researchers and clinicians have turned to this article as a cornerstone in their exploration of potential therapeutic interventions and treatment strategies. Table 2 presents the most influential articles from the Scopus database on catamenial epilepsy-related publications, along with their author, source, year of publication, number of citations and number of citations per year.

4. Discussion

Various studies have discussed catamenial epilepsy, although the literature remains scarce. This is evident from the fact only 440 related publications have been found overall since the first related publication in 1948 (an average of 5.95 articles per year and only 0.19 % [440/229 521] of all publications on epilepsy as a whole). However, the publication trend has increased over the past two decades and across various academic fields. According to the analysis above, a number of disciplines still present gaps. To be more specific, publications remain particularly scarce in the areas of practice guidelines, risk assessment, and medication.

The etiology and risk factors of catamenial epilepsy have not been entirely understood. This retrospective case-control clarified the relative risk for a positive family history of alcohol use disorder among women with catamenial epilepsy relative to women with non-catamenial epilepsy, with a risk ratio of 3.46 for those with (C1) perimenstrual and (C3) luteal phase patterns. The results of this study also revealed that a history of alcohol use disorder was a highly specific (91.7 %) indicator of catamenial epilepsy [37]. Further investigations could reveal other biomarkers that are more sensitive than seizure-menses calendar. Future studies on genetics and other etiologies are also being conducted, based on which proactive predictive and preventive measures can be implemented. To develop effective, sex-specific therapeutics for epilepsy, greater knowledge of the molecular and neural network foundation of sex differences in seizure patterns and reactions to antiepileptic medicines is essential.

The researched therapy choices necessitate a higher standard of evidence, and no specific treatment is available yet. Optimization of conventional anticonvulsant therapy is indicated as the first line of therapy; however, other therapy choices, including non-hormonal and hormonal medications, may be advantageous if the initial therapeutic option becomes ineffective [38]. Nevertheless, these various therapeutic possibilities are still the subject of controversy, and research yields contradictory findings. A review of treatments for seizures in catamenial epilepsy provides very low-certainty evidence that no difference in treatment effectiveness exists between norethisterone and placebo and moderate to low-certainty evidence that no difference in treatment effectiveness exists between progesterone and placebo. Research about the efficacy of hormonal and non-hormonal treatments, particularly in women who do not have regular menstrual cycles, is scarce, and additional clinical trials are necessary in this field. Regardless of the clinical significance of seizures in catamenial epilepsy and prevalence of this phenomenon among women with epilepsy, the literature lacks high-quality randomized controlled studies [3].

Practice guidelines from countries worldwide based on Scopus data are only found from Hong Kong. In accordance with the Update to the Hong Kong Epilepsy Guideline [39], Catamenial epilepsy is treated with intermittent benzodiazepines (for example, clobazam) throughout the menstrual cycle's vulnerable phase [40,41], during which anticonvulsant dosage may be increased. In general, the efficacy of hormone replacement treatment has not been demonstrated [42]; this could be useful for women experiencing more frequent seizures during the perimenstrual phase, particularly those with regular menstrual cycles [43], and additional research is likely necessary before a clinical recommendation may be made [42]. Daily or perimenstrual administration of acetazolamide may be beneficial for the treatment of catamenial epilepsy, despite the little data supporting its use [44].

In Indonesia, its practice management guideline follows the epilepsy management guidelines from the Indonesian Neurologist Association [45]. This refers to the studies [40,46] regarding medical management by which no specific therapy is available and several therapies may be given to help reduce epilepsy—for instance, addition to fast-acting anti-epileptic drugs such as clobazam (daily dosage of 20–30 mg) given 10 days during the menstrual period; acetazolamide (daily dosage of 250–500 mg) given 5–7 days before and during menstruation; and administration of hormones therapy use progesterone, progesterone metabolites, and progesterone antagonists.

Recent research on neurosteroid replacement therapy has shown that neurosteroid replacement therapy is a feasible method for enhancing control, which has a significant effect on decreasing the prevalence of premenstrual dysphoric disorder, postpartum depression, and catamenial seizures in women. Catamenial epilepsy, which is related to perimenstrual endogenous neurosteroids withdrawal or loss, is a distinct kind of pharmaco-resistant epilepsy in women. Neuro-steroids play a predominant role in the pathophysiology of catamenial seizures through plasticity in the expression and function of extrasynaptic GABA-A receptors and other mechanisms. In accordance with this cyclical process, a perimenstrual decrease in neurosteroid levels induces a selective increase in GABA-A receptor expression in the hippocampus. This compensatory increase in GABA-A receptors may function as a Trojan horse for exogenous neurosteroids to suppress excessive excitation, seizures, and behavioral dysfunction. Consequently, extra synaptic targeting provides a biological foundation for neuro-steroid therapy for catamenial epilepsy [29]. In addition, herbal preparations including *Asparagus racemosus* root extracts can be used as monotherapy or adjuvant therapy in conjunction with current anti epileptic drugs for the improved and safe management of catamenial epilepsy and depression [47].

Table 2
Most influential articles on catamenial epilepsy.

No	Title	Author	Source	Year	Citation	Cit./Year
1	Endogenous neurosteroids regulate GABAA receptors through two discrete transmembrane sites [33]	Hosie, A.M., Wilkins, M.E., Da Silva, H.M.A., Smart, T. G.	Nature 444(7118), pp. 486-489	2006	582	36.375
2	Ovarian cycle-linked changes in GABAA receptors mediating tonic inhibition alter seizure susceptibility and anxiety [34]	Maguire, J.L., Stell, B.M., Rafizadeh, M., Mody, I.	Nature Neuroscience 8(6), pp. 797-804	2005	492	28.941
3	Three patterns of catamenial epilepsy [10]	Herzog, A.G., Klein, P., Ransil, B.J.	Epilepsia 38(10), pp. 1082-1088	1997	378	15.120
4	Neurosteroid metabolism in the human brain [35]	Stoffel-Wagner, B.	European Journal of Endocrinology 145(6), pp. 669-679	2001	274	13.048
5	The role of estrogens in catamenial exacerbation of epilepsy [36]	Logothetis, J., Harner, R., Morrell, F., Torres, F.	Neurology 9(5), pp. 352-360	1959	228	3.619

Several studies have recently been conducted on the development of adjuvant therapies. A study investigated the anticonvulsant activity of cannabidiol in female rats during the proconvulsant hormonal phase, which is similar to catamenial seizures in humans [48]. Another study utilizing ferulic acid treatment in a mouse model demonstrated a significant reduction in seizure susceptibility and depressive-like behavior, potentially as a result of enhanced progesterone, restored oestradiol, corticosterone, monoamines, and glutamate decarboxylase enzyme function [49]. In addition, the anticonvulsant action of *Asparagus racemosus* was examined in a mouse model of cerebral oedema, resulting in a considerable reduction in seizure susceptibility [47]. However, as these investigations are only limited to pre-clinical testing, clinical trial research may be possible in this area.

Another study connected to adjunctive therapy revealed that drug-resistant patients may elect to initiate a ketogenic diet therapy (KDT)—specifically a modified Atkins diet—and that dietary alterations may be developed to target catamenial seizure patterns. Future trials testing KDT-based therapies are feasible given the high rate of adherence and tolerability. The study was limited due to its limited sample size, and future studies could involve a bigger clinical trial to investigate therapies and methods for catamenial seizure patterns in KDT-treated women. Adherence rate and tolerability imply the viability of future research, and it is necessary to discover more effective treatments for cervical cancer in general, but especially for women with a C3 catamenial pattern [50].

Developments related to the operative management of catamenial cases have also begun, although in very few cases. A case report described a patient surgically diagnosed with ovarian endometriosis, who had signs of catamenial epilepsy resistant to pharmacological therapy, and with therapy contraindications for hormone due to cerebral ischemia events, laparoscopic bilateral ovariosalpingectomy was performed. In the year following surgery, the seizure frequency has decreased (from 4 to 5 per week to 1 per month), which led to a gradual reduction in the dosages of valproic acid and phenobarbital [51]. Additional case requiring attention is related to catamenial status epilepticus, the incidence of which remains unknown and has rarely been reported [52]. Catamenial status epilepticus is uncommon but can occur in premenopausal women with repeated episodes of unresponsiveness, seizure clusters, or status epilepticus in both generalized and focal epilepsy. Our case report highlights the importance of monitoring menstrual cycles, establishing a pattern, and diagnosing with perimenstrual video EEG [53].

Catamenial epilepsy is a difficult condition to manage. Globally and throughout Asia—particularly Southeast Asia—related published studies are scarce. They also conform to the keyword analysis of other publications, such as those pertaining to practice recommendations (mainly), risk assessment, and medication. In terms of epidemiology (based on keyword analysis), very few articles address a clear occurrence of catamenial epilepsy, particularly in Asian nations. Other investigations pertaining to anticonvulsants, cerebral endometriosis, corticosterone, EEG, fertility, gaba, kainic acid, oral contraceptive, pregnanolone, teratogenicity are also feasible.

5. Conclusions

This study utilized both quantitative and qualitative data to support and urge more researchers to pay attention to catamenial epilepsy due to its significant prevalence in women and its classification as a female issue. We applied bibliometric analysis to review Scopus articles published on academic topics related to catamenial epilepsy and investigated the current developments in research. According to the findings, the initial result stage provided up to 440 datasets, which decreased to 320 datasets after the inclusion criteria were refined. The findings revealed trends, publication outlets, contributing authors, topic fields or disciplines, contributing countries or institutions, thematic maps, research gaps, potential future topics, and leading articles.

Through bibliometric and visualization methods and a review of the current state of knowledge, we drew the following conclusions: (1) the quantity of publications in catamenial epilepsy area has risen significantly over the past two decades, although publications are still scarce; (2) the majority of relevant research has focused on pathogenesis, diagnosis, and treatment, although publications are still scarce, particularly in the areas of practice guidelines, risk assessment, and medication; (3) collaboration is relatively low at the author, organizational, and national levels, especially in countries in the Asian continent; and (4) as this study used only one database (Scopus), we recommend that future research utilize a variety of databases (e.g., PubMed, Web of Science, and Springer) to obtain additional high-quality scientific contributions.

Funding

This study did not receive any specific grant from funding agencies in the public, commercial, or nonprofit sectors.

Ethical approvals

Review and/or approval by an ethics committee was not needed for this study and informed consent was not required for this study because this study does not involve experiments on animals or human subjects.

Data availability statement

The datasets used and analyzed during this study are available and included in this article or in the supplementary materials or referenced in this article.

CRedit authorship contribution statement

Diah Kurnia Mirawati: Writing – review & editing, Writing – original draft, Visualization, Validation, Supervision, Software, Resources, Project administration, Methodology, Investigation, Funding acquisition, Formal analysis, Data curation, Conceptualization. **Nanang Wiyono:** Writing – review & editing, Supervision, Software, Resources, Project administration, Methodology, Formal analysis. **Muhana Fawwazy Ilyas:** Writing – review & editing, Writing – original draft, Visualization, Validation, Software, Resources, Project administration, Methodology, Investigation, Formal analysis, Data curation, Conceptualization. **Stefanus Erdana Putra:** Writing – review & editing, Writing – original draft, Visualization, Validation, Software, Resources, Project administration, Methodology, Investigation, Formal analysis, Data curation, Conceptualization. **Muhammad Hafizhan:** Writing – review & editing, Writing – original draft, Visualization, Validation, Software, Resources, Project administration, Methodology, Investigation, Formal analysis, Data curation, Conceptualization.

Declaration of competing interest

The authors declare that they have no known competing financial interests or personal relationships that could have appeared to influence the work reported in this paper.

Acknowledgement

Our gratitude goes to Universitas Sebelas Maret, Indonesia, for their willingness to be the study location.

Appendix A. Supplementary data

Supplementary data to this article can be found online at <https://doi.org/10.1016/j.heliyon.2024.e31474>.

References

- [1] I.A.W. Kotsopoulos, T. Van Merode, F.G.H. Kessels, M.C.T.F.M. De Krom, J.A. Knottnerus, Systematic review and meta-analysis of incidence studies of epilepsy and Unprovoked seizures, *Epilepsia* 43 (2002) 1402–1409, <https://doi.org/10.1046/j.1528-1157.2002.t01-1-26901.x>.
- [2] S. Frank, N.A. Tyson, A clinical approach to catamenial epilepsy: a review, *Perm. J.* 24 (2020) 1–3, <https://doi.org/10.7812/TPP/19.145>.
- [3] M.J. Maguire, S.J. Nevitt, Treatments for seizures in catamenial (menstrual-related) epilepsy, *Cochrane Database Syst. Rev.* 2021 (2021), <https://doi.org/10.1002/14651858.CD013225.pub3>.
- [4] E. Cerulli Irelli, A. Morano, E. Cocchi, S. Casciato, M. Fanella, M. Albini, et al., Doing without valproate in women of childbearing potential with idiopathic generalized epilepsy: implications on seizure outcome, *Epilepsia* 61 (2020) 107–114, <https://doi.org/10.1111/epi.16407>.
- [5] J. Christensen, M.J. Kjeldsen, H. Andersen, M.L. Friis, P. Sidenius, Gender differences in epilepsy, *Epilepsia* 46 (2005) 956–960, <https://doi.org/10.1111/j.1528-1167.2005.51204.x>.
- [6] D. Samanta, PCDH19-Related epilepsy syndrome: a comprehensive clinical review, *Pediatr. Neurol.* 105 (2020) 3–9, <https://doi.org/10.1016/j.pediatrneurol.2019.10.009>.
- [7] S.K. Kessler, E. McGinnis, A practical guide to treatment of childhood absence epilepsy, *Pediatr. Drugs* 21 (2019) 15–24, <https://doi.org/10.1007/s40272-019-00325-x>.
- [8] V. Padmanaban, S. Inati, A. Ksendzovsky, K. Zaghoul, Clinical advances in photosensitive epilepsy, *Brain Res.* 1703 (2019) 18–25, <https://doi.org/10.1016/j.brainres.2018.07.025>.
- [9] R. Keni, B. Mostacci, G. Kiteva-Trenchevska, L. Licchetta, L. Ignjatova, S. Thomas, et al., Women's issues, *Epileptic Disord.* 22 (2020) 355–363, <https://doi.org/10.1684/epd.2020.1173>.
- [10] A.G. Herzog, P. Klein, B.J. Rand, Three patterns of catamenial epilepsy, *Epilepsia* 38 (1997) 1082–1088, <https://doi.org/10.1111/j.1528-1157.1997.tb01197.x>.
- [11] D.S. Reddy, Neurosteroids and their role in sex-specific epilepsies, *Neurobiol. Dis.* 72 (2014) 198–209, <https://doi.org/10.1016/j.nbd.2014.06.010>.
- [12] D.S. Reddy, Neuroendocrine aspects of catamenial epilepsy, *Horm. Behav.* 63 (2013) 254–266, <https://doi.org/10.1016/j.yhbeh.2012.04.016>.
- [13] D.S. Reddy, Catamenial epilepsy: discovery of an extrasynaptic molecular mechanism for targeted therapy, *Front. Cell. Neurosci.* 10 (2016), <https://doi.org/10.3389/fncel.2016.00101>.

- [14] A.G. Herzog, Catamenial epilepsy: Update on prevalence, pathophysiology and treatment from the findings of the NIH Progesterone Treatment Trial, *Seizure* 28 (2015) 18–25, <https://doi.org/10.1016/j.seizure.2015.02.024>.
- [15] D. Rosciszewska, B. Buntner, I. Guz, L. Zawisza, Ovarian hormones, anticonvulsant drugs, and seizures during the menstrual cycle in women with epilepsy, *J. Neurol. Neurosurg. Psychiatry* 49 (1986) 47–51, <https://doi.org/10.1136/jnnp.49.1.47>.
- [16] D. Rosciszewska, Analysis of seizure dispersion during menstrual cycle in women with epilepsy, *Monogr. Neural Sci.* 5 (1980) 280–284.
- [17] J. Laidlaw, Catamenial epilepsy, *Lancet* 268 (1956) 1235–1237, [https://doi.org/10.1016/S0140-6736\(56\)90003-4](https://doi.org/10.1016/S0140-6736(56)90003-4).
- [18] X. Zhao, S. Wang, X. Wang, Characteristics and trends of research on new energy vehicle reliability based on the web of science, *Sustainability* 10 (2018) 3560, <https://doi.org/10.3390/su10103560>.
- [19] H. Nobanee, F.Y. Al Hamadi, F.A. Abdulaziz, L.S. Abukarsh, A.F. Alqahtani, S.K. AlSubaey, et al., A bibliometric analysis of sustainability and risk management, *Sustainability* 13 (2021) 3277.
- [20] J. Baas, M. Schotten, A. Plume, G. Côté, R. Karimi, Scopus as a curated, high-quality bibliometric data source for academic research in quantitative science studies, *Quantitative Science Studies* 1 (2020) 377–386, https://doi.org/10.1162/qss_a_00019.
- [21] H. Sikandar, Y. Vaicandam, N. Khan, M.I. Qureshi, A. Ullah, Scientific mapping of industry 4.0 research: a bibliometric analysis, *International Journal of Interactive Mobile Technologies (IJIM)* 15 (2021) 129, <https://doi.org/10.3991/ijim.v15i18.25535>.
- [22] W. Barbosa, T. Prado, C. Batista, J.C. Câmara, R. Cerqueira, R. Coelho, et al., Electric vehicles: bibliometric analysis of the current state of the art and perspectives, *Energies* 15 (2022) 395, <https://doi.org/10.3390/en15020395>.
- [23] J. Shi, K. Duan, G. Wu, R. Zhang, X. Feng, Comprehensive metrological and content analysis of the public-private partnerships (PPPs) research field: a new bibliometric journey, *Scientometrics* 124 (2020) 2145–2184, <https://doi.org/10.1007/s11192-020-03607-1>.
- [24] N. Donthu, S. Kumar, D. Mukherjee, N. Pandey, W.M. Lim, How to conduct a bibliometric analysis: an overview and guidelines, *J. Bus. Res.* 133 (2021) 285–296, <https://doi.org/10.1016/j.jbusres.2021.04.070>.
- [25] M. Aria, C. Cuccurullo, Bibliometrix : an R-tool for comprehensive science mapping analysis, *J Informetr* 11 (2017) 959–975, <https://doi.org/10.1016/j.joi.2017.08.007>.
- [26] L. Skaf, E. Buonocore, S. Dumontet, R. Capone, P.P. Franzese, Applying network analysis to explore the global scientific literature on food security, *Ecol. Inf.* 56 (2020) 101062, <https://doi.org/10.1016/j.ecoinf.2020.101062>.
- [27] P.A. Schwob, M. Bonduette, A. Baudet, Catamenial epilepsy, *Sem. Hop.* 24 (1948) 967–970.
- [28] D.S. Reddy, H.-Y. Kim, M.A. Rogawski, Neurosteroid withdrawal model of perimenstrual catamenial epilepsy, *Epilepsia* 42 (2002) 328–336, <https://doi.org/10.1046/j.1528-1157.2001.10100.x>.
- [29] D.S. Reddy, Neurosteroid replacement therapy for catamenial epilepsy, postpartum depression and neuroendocrine disorders in women, *J. Neuroendocrinol.* 34 (2022), <https://doi.org/10.1111/jne.13028>.
- [30] T. Gerlach, V.L. Clyde, G.L. Morris, B. Bell, R.S. Wallace, Alternative therapeutic options for medical management of epilepsy in apes, *J. Zoo Wildl. Med.* 42 (2011) 291–294, <https://doi.org/10.1638/2010-0184.1>.
- [31] S.A.E. Van Meervenne, H.A. Volk, K. Matiassek, L.M.L. Van Ham, The influence of sex hormones on seizures in dogs and humans, *Vet. J.* 201 (2014) 15–20, <https://doi.org/10.1016/j.tvjl.2014.05.008>.
- [32] S.A.E. Van Meervenne, H.A. Volk, L.M.L. Van Ham, Association between estrus and onset of seizures in dogs with idiopathic epilepsy, *J. Vet. Intern. Med.* 29 (2015) 251–253, <https://doi.org/10.1111/jvim.12505>.
- [33] A.M. Hosie, M.E. Wilkins, H.M.A. da Silva, T.G. Smart, Endogenous neurosteroids regulate GABAA receptors through two discrete transmembrane sites, *Nature* 444 (2006) 486–489, <https://doi.org/10.1038/nature05324>.
- [34] J.L. Maguire, B.M. Stell, M. Rafizadeh, I. Mody, Ovarian cycle-linked changes in GABAA receptors mediating tonic inhibition alter seizure susceptibility and anxiety, *Nat. Neurosci.* 8 (2005) 797–804, <https://doi.org/10.1038/nn1469>.
- [35] B. Stoffel-Wagner, Neurosteroid metabolism in the human brain, *Eur. J. Endocrinol.* (2001) 669–679, <https://doi.org/10.1530/eje.0.1450669>.
- [36] J. Logothetis, R. Harner, F. Morrell, F. Torres, The role of estrogens in catamenial exacerbation of epilepsy, *Neurology* 9 (1959) 352, <https://doi.org/10.1212/WNL.9.5.352>.
- [37] A.G. Herzog, Association between family history of alcohol use disorder and catamenial epilepsy, *Epilepsia* 63 (2022), <https://doi.org/10.1111/epi.17261>.
- [38] G. Moscol, P.H. Espino, L.C. Mayor, J.G. Burneo, Epilepsy with catamenial pattern, *Rev. Neurol.* 74 (2022) 303–311, <https://doi.org/10.33588/rn.7409.2022041>.
- [39] R.S. Chang, K.H. Lui, W. Ip, E. Yeung, A.W. Yung, H. Leung, et al., Update to the Hong Kong Epilepsy Guideline: evidence-based recommendations for clinical management of women with epilepsy throughout the reproductive cycle, *Hong Kong Med. J.* 26 (2020) 421–431, <https://doi.org/10.12809/hkmj198367>.
- [40] M. Feely, J. Gibson, Intermittent clobazam for catamenial epilepsy: tolerance avoided, *J. Neurol. Neurosurg. Psychiatry* 47 (1984) 1279–1282, <https://doi.org/10.1136/jnnp.47.12.1279>.
- [41] N. Foldvary-Schaefer, T. Falcone, Catamenial epilepsy: pathophysiology, diagnosis, and management, *Neurology* 61 (2003) S2–S15, https://doi.org/10.1212/WNL.61.6_suppl.2.S2.
- [42] A.G. Herzog, K.M. Fowler, S.D. Smithson, L.A. Kalayjian, C.N. Heck, M.R. Sperling, et al., Progesterone vs placebo therapy for women with epilepsy: a randomized clinical trial, *Neurology* 78 (2012) 1959–1966, <https://doi.org/10.1212/WNL.0b013e318259e1f9>.
- [43] A. Navis, C. Harden, A treatment approach to catamenial epilepsy, *Curr. Treat. Options Neurol.* 18 (2016) 30, <https://doi.org/10.1007/s11940-016-0413-6>.
- [44] B. Mostacci, R. Esposito, S. Lello, F. Bisulli, L. Licchetta, P. Tinuper, Estrogen-related seizure exacerbation following hormone therapy for assisted reproduction in women with epilepsy, *Seizure* 61 (2018) 200–202, <https://doi.org/10.1016/j.seizure.2018.08.024>.
- [45] *Kelompok Studi Epilepsi Perhimpunan Dokter Spesialis Saraf Indonesia (PERDOSSI), Pedoman Tatalaksana Epilepsi, sixth ed., Airlangga University Press, Surabaya, 2019.*
- [46] A. Verrotti Alberto, D'Egidio, Verrotti Agostinelli, P. Pavone, Diagnosis and management of catamenial seizures: a review, *Int J Womens Health* 535 (2012), <https://doi.org/10.2147/IJWH.S28872>.
- [47] P. Pahwa, T. Singh, R.K. Goel, Anticonvulsant effect of *Asparagus racemosus* Willd. in a mouse model of catamenial epilepsy, *Neurochem. Res.* 47 (2022) 422–433, <https://doi.org/10.1007/s11064-021-03455-2>.
- [48] N.R.L.L. Janisset, S.A.A. Romariz, D. Hashiguchi, M.L. Quintella, C. Gimenes, T. Yokoyama, et al., Partial protective effects of cannabidiol against PTZ-induced acute seizures in female rats during the proestrus–estrus transition, *Epilepsy Behav.* 129 (2022) 108615, <https://doi.org/10.1016/j.yebeh.2022.108615>.
- [49] H.K. Dhillon, T. Singh, R.K. Goel, Ferulic acid inhibits catamenial epilepsy through modulation of female hormones, *Metab. Brain Dis.* (2022), <https://doi.org/10.1007/s11011-022-01054-w>.
- [50] E.A. Felton, B.J. Henry-Barron, A.K. Jan, A. Shegelman, K. Faltersack, D. Vizthum, et al., The feasibility and tolerability of medium chain triglycerides in women with a catamenial seizure pattern on the modified Atkins diet, *Nutrients* 13 (2021) 2261, <https://doi.org/10.3390/nu13072261>.
- [51] G.A. Antoniou, G.S. Georgiadis, S.A. Antoniou, P. Pavlidis, D. Maras, G.S. Sfyroeras, et al., Endovascular repair for ruptured abdominal aortic aneurysm confers an early survival benefit over open repair, *J. Vasc. Surg.* 58 (2013) 1091–1105, <https://doi.org/10.1016/j.jvs.2013.07.109>.
- [52] B. Ramanujam, A. Arora, V. Malhotra, D. Dash, S. Mehta, M. Tripathi, A case of recurrent status epilepticus and successful management with progesterone, *Epileptic Disord.* 18 (2016) 101–105, <https://doi.org/10.1684/epd.2016.0792>.
- [53] A.J. Chalissery, E. Murphy, G. Mullins, P. Widdess-Walsh, R. Kilbride, N. Delanty, Recurrent catamenial status epilepticus: is it rare or an under recognized phenomenon in women with epilepsy? *Epilepsy Behav Case Rep* 9 (2018) 19–21, <https://doi.org/10.1016/j.ebcr.2017.10.002>.