REVIEW

Rare and Complicated Granulomatous Lobular Mastitis (2000–2023): A Bibliometrics Study and Visualization Analysis

Mengdi Zhang[1], Dongqing Pu[1], Dandan Feng[1], Guangxi Shi[2], Jingwei Li[2]

[1]First Clinical Medical College, Shandong University of Traditional Chinese Medicine, Jinan City, People's Republic of China; [2]Department of Thyroid and Breast Diagnosis and Treatment Center, Affiliated Hospital of Shandong University of Traditional Chinese Medicine, Jinan City, People's Republic of China

Correspondence: Jingwei Li, Department of Thyroid and Breast Surgery, Affiliated Hospital of Shandong University of Traditional Chinese Medicine, Jinan City, People's Republic of China, Email 71000395@sdutcm.edu.cn

Purpose: Granulomatous mastitis (GLM) is a rare and complex chronic inflammatory disease of the breast with an unknown cause and a tendency to recur. As medical science advances, the cause, treatment strategies, and comprehensive management of GLM have increasingly attracted widespread attention. The aim of this study is to assess the development trends and research focal points in the GLM field over the past 24 years using bibliometric analysis.
Methods: Using GLM, Granulomatous mastitis (GM), Idiopathic granulomatous lobular mastitis (IGLM), and Idiopathic granulomatous mastitis (IGM) as keywords, we retrieved publications related to GLM from 2000 to 2023 from the Web of Science, excluding articles irrelevant to this study. Citespace and VOSviewer were employed for data analysis and visualization.
Results: A total of 347 publications were included in this analysis. Over the past 24 years, the number of publications has steadily increased, with Turkey being the leading contributor in terms of publications and citations. The University of Health Sciences, Istanbul University, and Istanbul University Cerrahpasa were the most influential institutions. The Breast Journal, Breast Care, and Journal of Investigative Surgery were the journals that published the most on this topic. The research primarily focused on the cause, differential diagnosis, treatment, and comprehensive management of GLM. Issues related to recurrence, hyperprolactinemia, and Corynebacterium emerged as current research hotspots.
Conclusion: Our bibliometric study outlines the historical development of the GLM field and identifies recent research focuses and trends, which may aid researchers in identifying research hotspots and directions, thereby advancing the study of GLM.
Keywords: granulomatous lobular mastitis, bibliometric analysis, visual analysis, knowledge graphs, review

Introduction

Granulomatous lobular mastitis (GLM) is a non-lactating breast inflammatory disease that forms lesions in the mammary gland lobules and is primarily characterized by non-caseating necrotizing granulomas.[1] The incidence of GLM represents 0.3%~1.9% of all breast diseases worldwide[2] and has shown a significant increase recently, accounting for 4%~5% of benign breast lesions in developing countries.[3] Women of childbearing age with a history of childbirth are more susceptible to GLM. The causes of GLM and the triggers for its recurrent episodes remain unknown.[4] The main treatment modalities for GLM include antibiotics, steroids, immunosuppressants, and surgery.[5] Steroid shock therapy offers notable short-term benefits but carries significant side effects, often accompanied by complications such as Cushing's syndrome.[6] Surgical interventions can compromise the aesthetic appearance of the breast, impact breastfeeding capabilities, and cause irreversible damage, thereby increasing the economic, psychological, and occupational burdens on women.[7]

Bibliometric analysis is a statistical tool that provides a comprehensive overview of specific scientific research fields through qualitative and quantitative methods, reveals the characteristics and evolving trends of literature, outlines

3709

research trajectories, and forecasts future directions.[8] To date, the field of GLM has not been systematically assessed through bibliometric analysis. In this study, we employ bibliometric analysis to explore the knowledge dimension of the GLM field, quantify research patterns, and address literature gaps. Our bibliometric study on publications in the GLM field from 2000 to 2024 utilizes the national cooperation network, author network, co-citation analysis, and the co-occurrence of keywords. This study evaluates the research trends, contributions from various countries and institutions, the most productive journals, highly cited articles, references, and keywords. It also reveals the current research focal points and potential future research directions.

Materials and Methods
Data Source and Search Strategy
The data were sourced from the Web of Science (WOS) Core Collection, which includes the Science Citation Index-Expanded (SCI-E) and Social Sciences Citation Index (SSCI). WOS is widely recognized as a leading database for bibliometric analysis.[9–11]

The publication years spanned from 2000 to 2023, and only articles and reviews written in English were considered. The relevant literature was identified using the keywords Granulomatous lobular mastitis (GLM), Granulomatous mastitis (GM), Idiopathic granulomatous lobular mastitis (IGLM), and Idiopathic granulomatous mastitis (IGM). The search criteria are detailed in Table S1.

Data Analysis
Four metrics were utilized to assess the productivity and impact of the research publications: total publications (TP), total citations (TC), average citations per publication (ACPP), and H-index. The H-index indicates that an entity has H publications, each of which has been cited at least H times by other articles.[9] These metrics gauge the academic influence of a country, institution, or journal.

VOSviewer (version 1.6.19) was employed for analyzing cooperation among countries and institutions and for conducting keyword co-occurrence analysis. A measurement index clustering technique within the software grouped the keywords into distinct clusters according to their association intensity and direction. Each cluster was visually differentiated by color and represented through time progression in varying shades.[12]

CiteSpace (version 6.2.R4) was used for co-citation analysis of references and for detecting citation bursts in references.[13] The time slicing covered January 2000 to December 2023, with a one-year interval. The selection criteria included a g-index (k=25), link retaining factor (LRF)=3.0, maximum links per node (L/N) =10, look back year (LBY) =5, and e=1.0, with other parameters set to default. In this analysis, a clustering algorithm based on keywords and log-likelihood ratio grouped documents into conceptual clusters, each marked with noun phrases to outline different research characteristics. The clusters' colors corresponded to citation years, and CiteSpace also illustrated the temporal trends of high-frequency keywords in the GLM field.

Results
Timeline Distribution of Publications
A total of 347 publications were retrieved, with all records and references displayed in Figure 1. Figure 2 illustrates the development trend of the GLM field over time. Over the past 24 years, the number of publications has steadily increased, reaching 42 in 2023, which is 14 times the number of publications in 2000 (3 publications). This indicates growing research interest in the GLM field. The total number of citations peaked in 2015 (557 citations) and then gradually decreased from 2021 to 2023. The average number of citations per paper plummeted by 99.0% from 61 in 2000 to 0.31 in 2023.

Analysis of Countries Distribution
In this study, 41 countries were identified, as depicted in Figure 3. The leading contributors were Turkey, China, the USA, Iran, England, India, and Japan. Turkey contributed the most papers (104 papers, 29.97%) and ranked first in total
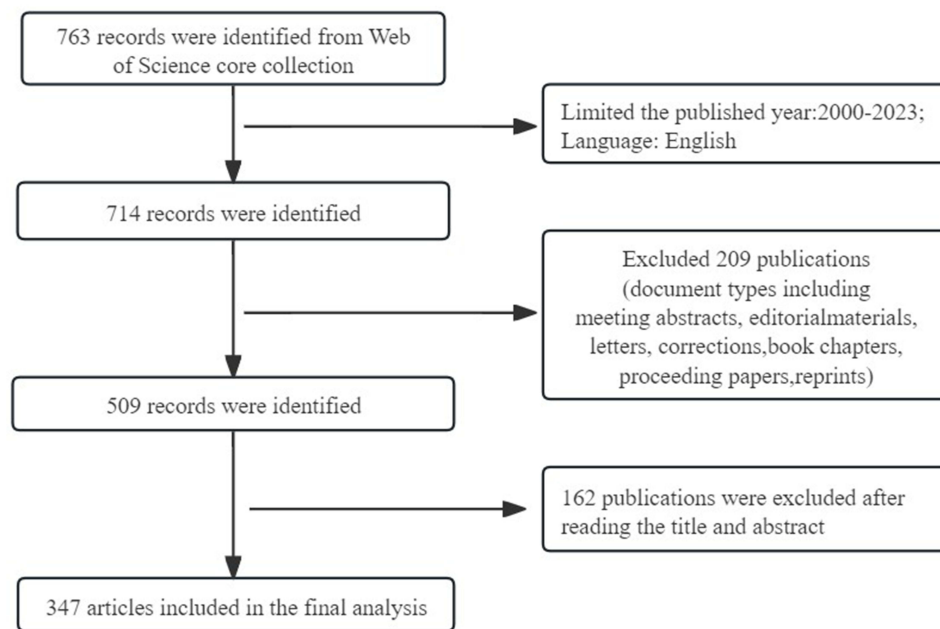
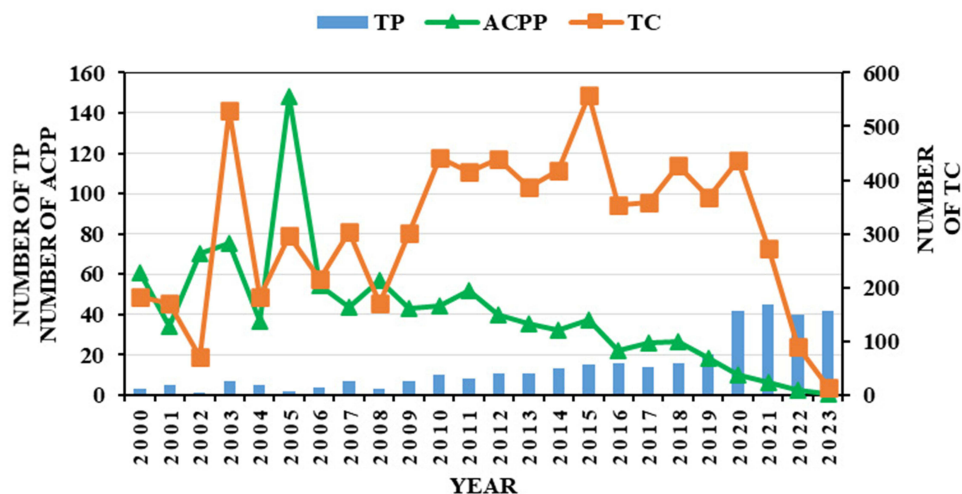**Figure 1** Flow diagram of publications screening.

Figure 2 Timeline distribution of publications in the field of GLM during 2000–2023.

citations (2473 citations) (Table 1). England had the highest average number of citations per paper (36.45), followed by South Korea (30.43) and Canada (28). Collectively, the top ten countries accounted for 90.20% of all publications, with South Korea and Canada tied for 10th place.

The growth trend of the top seven countries in terms of publication numbers over the past 24 years is shown in Figure 4. Turkey, China, and the USA have shown fluctuating growth, with China experiencing the most significant increase. Despite their later entry into this field, Iran and India rapidly increased their publication output. In contrast, the number of publications from Japan has remained low.

To explore the cooperation characteristics among countries in the GLM field, a collaboration network is illustrated in Figure 5. Among the 41 countries, there is 1 from Africa, 10 from Asia, 13 from Europe, 2 from Oceania, 9 from the Middle East, 3 from North America, and 3 from South America. Canada is central in the international cooperation
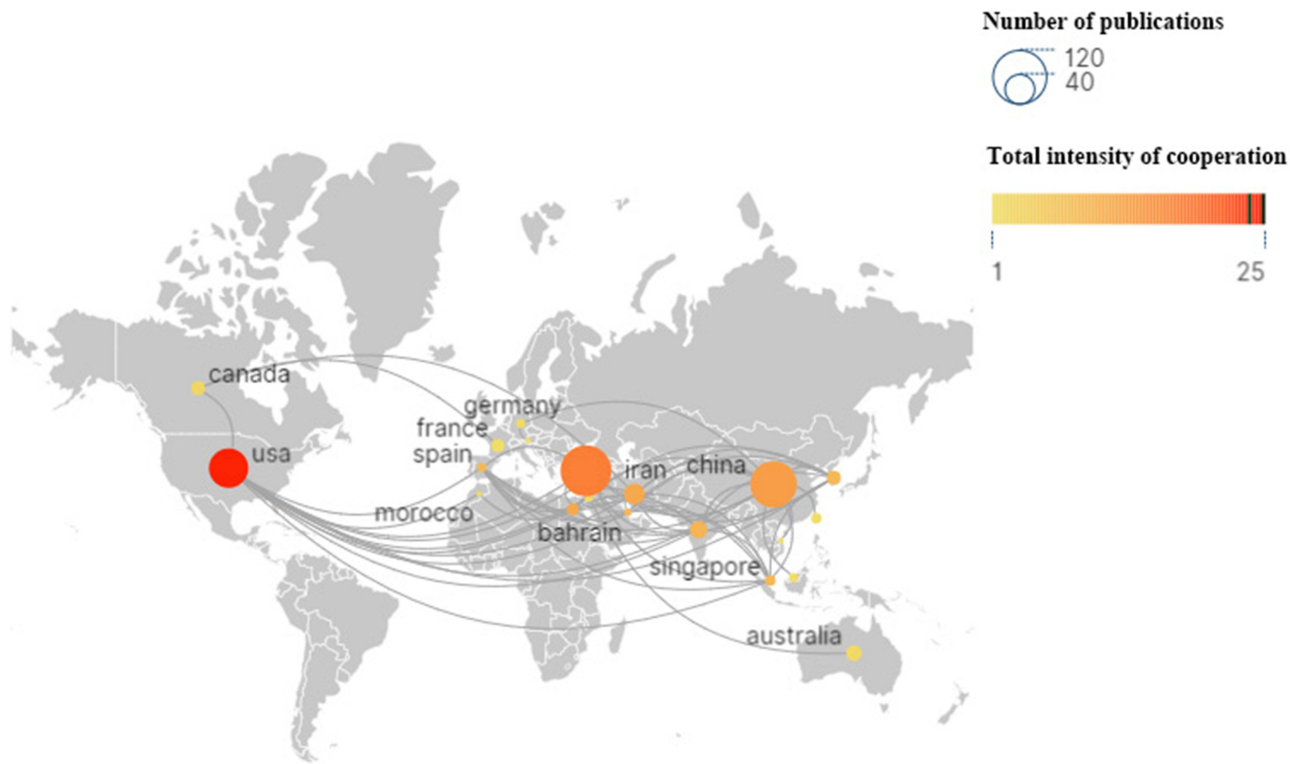
Figure 3 Global collaboration and distribution of publications in the field of GLM. The number of publications and total citations are represented, accordingly, by node sizes and colors.

network, maintaining strong relationships with other countries in this field. However, most countries exhibit unstable and incomplete communication and cooperation.

Analysis of Institutions Distribution

This study identified the 10 most productive and influential institutions in the GLM field (Table 2). The University of Health Sciences Turkey leads with 17 publications, followed by Istanbul University with 15 publications, and Istanbul University Cerrahpasa with 11 publications. In terms of total citations, Istanbul University is at the forefront with 441 citations, while the University of Texas System follows with 244 citations. Notably, five of the top ten institutions are based in the USA and three in China. The collaborative network among these institutions is depicted in Figure 6, which includes 310 institutions that have one or more publications.

Table 1 Top 10 Most Productive Countries in the Field of GLM

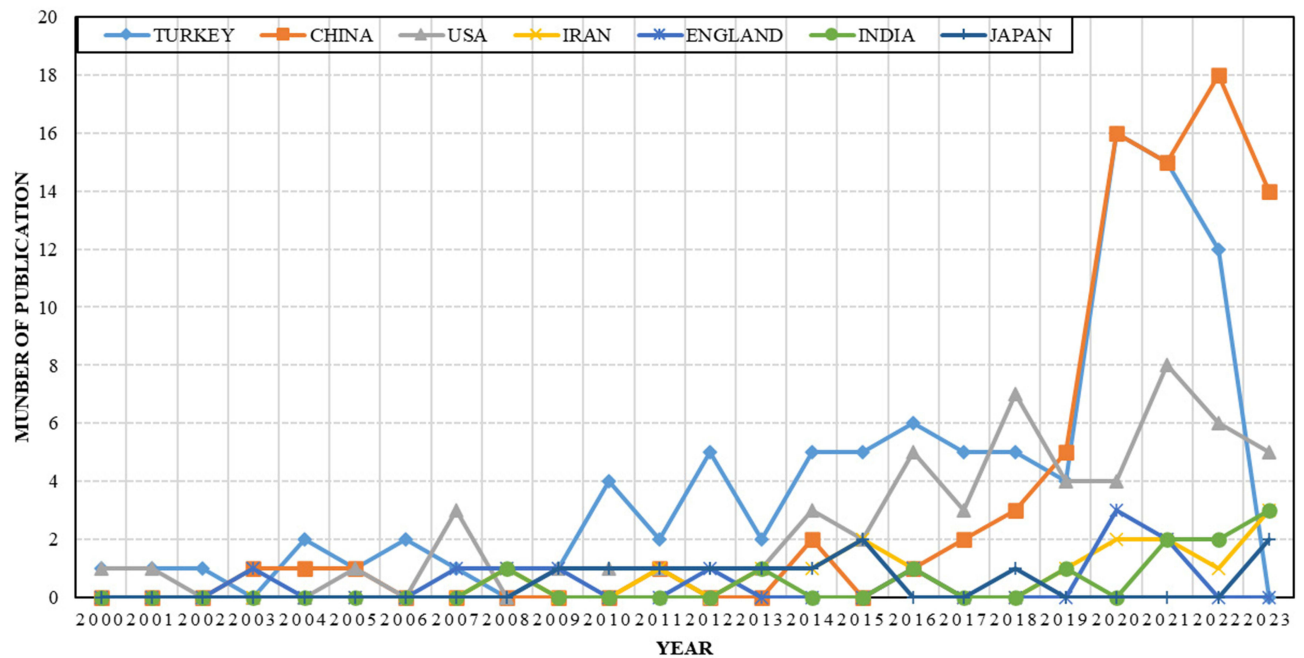| Country	TP	Percentage (%)	TC	ACPP	H-Index
TURKEY	104	29.97	2473	25.99	31
CHINA	80	23.05	855	10.69	15
USA	57	16.43	1463	25.67	22
IRAN	16	4.61	432	27	11
ENGLAND	11	3.17	401	36.45	7
INDIA	11	3.17	69	6.27	4
JAPAN	11	3.17	234	21.27	8
AUSTRALIA	9	2.59	186	20.67	6
CANADA	7	2.02	196	28	5
SOUTH KOREA	7	2.02	213	30.43	5

Figure 4 Top 7 countries distribution trends of publications in the field of GLM.

CiteSpace, v. 6.2.R4 (64-bit) Advanced
January 22, 2024 at 6:56:42 PM CST
WoS: E:\0 CITESPACE\72\output
Timespan: 2000-2023 (Slice Length=1)
Selection Criteria: g-index (k=25), LRF=3.0, L/N=10, LBY=5, e=1.0
Network: N=44, E=58 (Density=0.0613)
Largest 30 CCs: 44 (100%)
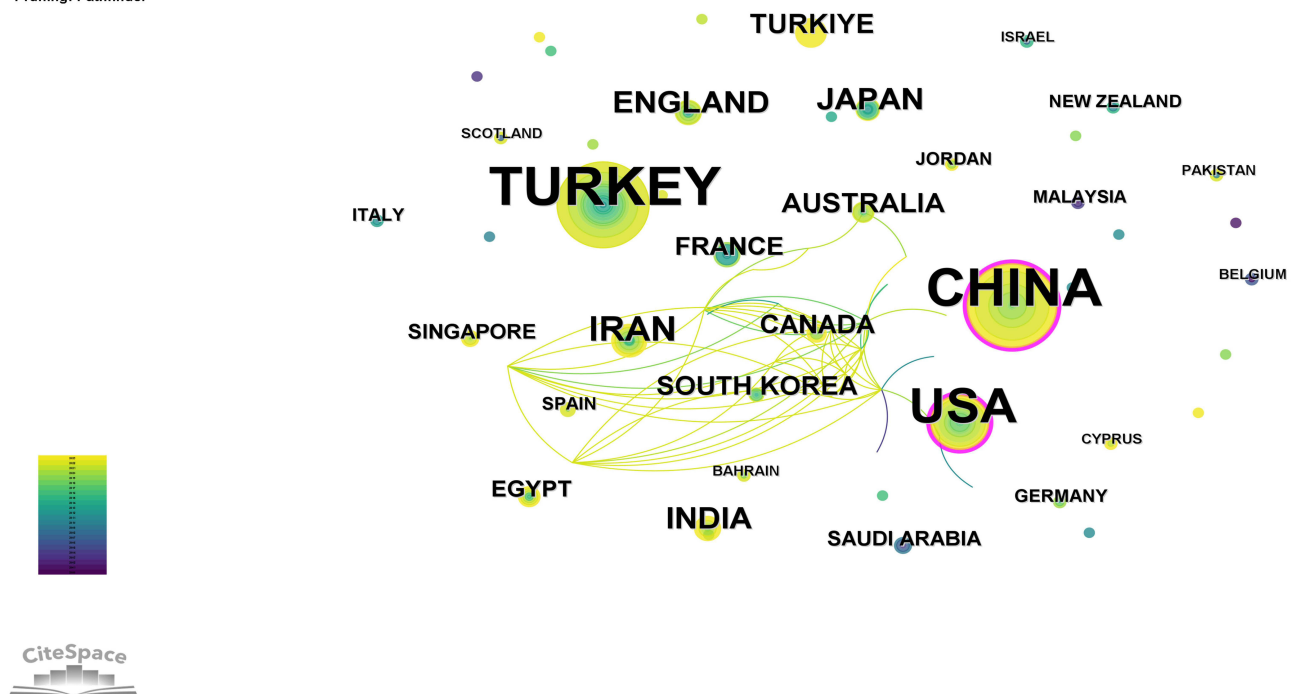Nodes Labeled: 1.0%
Pruning: Pathfinder

Figure 5 Collaboration networks of countries with at least one publication. Nodes represent countries, sized by publication count. Curves illustrate the links between countries, sized by cooperation intensity, with colors denoting regional distribution.

Table 2 Top 10 Most Productive Institutions in the Field of GLM

Institution	Country	TP	TC	ACPP	H-Index
University of Health Sciences Turkey	Turkey	17	191	11.24	8
Istanbul University	Turkey	15	441	29.4	10
Istanbul University Cerrahpasa	Turkey	11	174	15.82	7
Tehran University of Medical Sciences	Iran	8	221	27.63	6
Beijing University of Chinese Medicine	China	7	27	3.86	2
University of Texas System	USA	7	244	34.86	4
Istanbul Medipol University	Turkey	6	33	5.5	3
Konya Research and Training Hospital	Turkey	6	85	14.17	5
University of Electronic Science Technology of China	China	6	14	2.33	2
Wuhan University	China	6	52	8.67	4

Analysis of Influential Journals

A total of 163 journals have published articles on GLM. As indicated in Table 3, the top fifteen journals account for 33.72% of total publications in this area (117 out of 347). The Breast Journal stands out in all metrics, including TP, TC, ACPP and H-Index, highlighting its prominent role in the field. Following in output are Breast Care and Journal of Investigative Surgery. The total citation counts for the Breast Journal and Breast Care exceed 1000. Half of the top ten journals boast an impact factor greater than 2, and these journals predominantly focus on breast diseases.
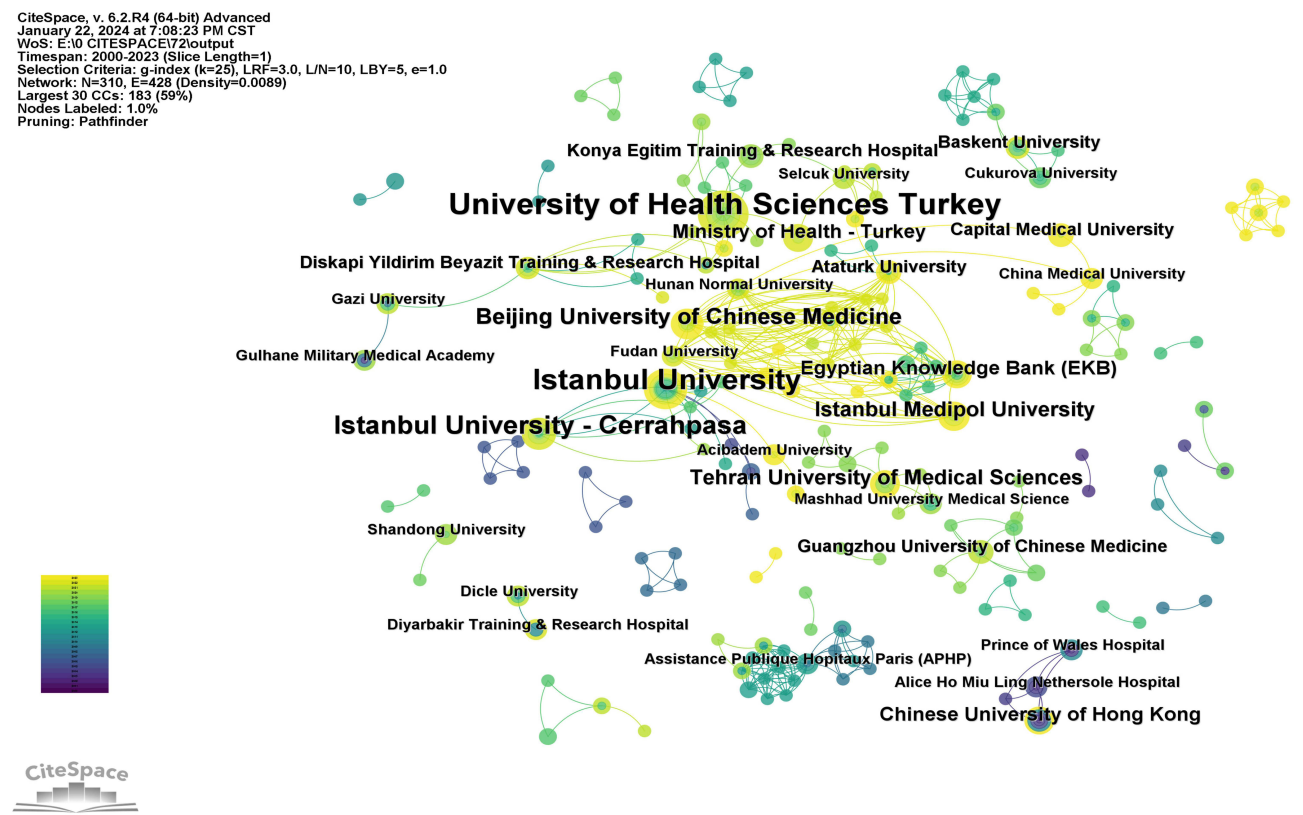

Figure 6 Collaboration networks of institutions with at least one publication. Nodes represent institutions, sized by publication count. Lines indicate institutional linkages, sized by cooperation intensity, with colors representing different countries.

Table 3 Top 14 Most Productive Journals in the Field of GLM

Journal	TP	TC	ACPP	H-Index	IF (2023)
Breast Journal	24	997	41.54	16	2.1
Breast Care	18	327	18.17	11	2.1
Journal of Investigative Surgery	13	160	12.31	8	1.9
World Journal of Surgery	8	466	58.25	7	2.6
Surgery Today	7	233	33.29	6	2.5
India Journal of Surgery	6	4	0.67	1	0.4
Medicine	6	9	1.5	2	1.6
BMC Women's Health	5	49	9.8	2	2.5
Gland Surgery	5	28	5.6	3	1.8
Iranian Journal of Radiology	5	47	9.4	2	0.3
JCPSP-Journal of The College of Physicians and Surgeons Pakistan	5	17	3.4	3	1
Pathology	5	350	70	4	4.5
Rheumatology International	5	81	16.2	3	4
Turkish Journal of Medical Sciences	5	59	11.8	4	2.3

Analysis of Highly Cited Articles

Table 4 presents the top 10 most frequently cited articles in the GLM field. Three articles, each attracting over 150 citations, were published in Pathology, American Journal of Roentgenology, and the Breast Journal, respectively. The most cited article, "A clinicopathological review of 34 cases of inflammatory breast disease showing an association between corynebacteria infection and granulomatous mastitis", authored by Taylor GB in 2003, has received 171 citations. The most recent influential publication, "Aetiology of idiopathic granulomatous mastitis" by Altintoprak F, appeared in the 2014 World Journal of Clinical Cases and has been cited 109 times.

Table 4 Top 10 Articles with the Highest Citations in the Field of GLM

Year	First Author	Title	Journal	Citations
2003	Taylor, GB[14]	A clinicopathological review of 34 cases of inflammatory breast disease showing an association between corynebacteria infection and granulomatous mastitis	Pathology	171
2009	Larsen, LJH[15]	Granulomatous Lobular Mastitis: Imaging, Diagnosis, and Treatment	American Journal of Roentgenology	166
2005	Lai, ECH[16]	The role of conservative treatment in idiopathic granulomatous mastitis	Breast Journal	158
2008	Al-Khaffaf, B[17]	Idiopathic granulomatous mastitis: A 25-year experience	Journal of the American College of Surgeons	143
2005	Asoglu, O[18]	Feasibility of surgical management in patients with granulomatous mastitis	Breast Journal	139
2007	Baslaim, MM[19]	Idiopathic granulomatous mastitis: A heterogeneous disease with variable clinical presentation	World Journal of Surgery	116
2006	Akcan, A[20]	Granulomatous lobular mastitis: A complex diagnostic and therapeutic problem	World Journal of Surgery	115
2000	Erhan, Y[21]	A clinicopathologic study of a rare clinical entity mimicking breast carcinoma: idiopathic granulomatous mastitis	Breast	112
2003	Azlina, AF[22]	Chronic granulomatous mastitis: Diagnostic and therapeutic considerations	World Journal of Surgery	111
2014	Altintoprak, F[23]	Aetiology of idiopathic granulomatous mastitis	World Journal of Clinical Cases	109

Analysis of Co-Cited References

Co-citation analysis of references reflects the structure and dynamics of the knowledge field, revealing both the research frontier and the knowledge base.[24] A total of 540 references related to GLM have been co-cited over the past 24 years. Figure 7 displays the 540 co-cited references derived from the literature spanning from 2000 to 2023. The most frequently cited reference, authored by David Barreto in 2018, centers on the "Factors related to recurrence of idiopathic granulomatous mastitis: what do we learn from a multicentre study". Another important work, "Treatments for Idiopathic Granulomatous Mastitis: Systematic Review and Meta-Analysis", was published in the ANZ Journal of Surgery and Brewing Medicine in 2018 and 2017, respectively. Table 5 provides more detailed information about the first 10 cited references, each of which has been cited at least 25 times. The most cited reference, appearing in Breast Cancer Research and Treatment, is titled "Granulomatous mastitis: etiology, imaging, pathology, treatment, and clinical findings".

As depicted in Figure 7, we identified 12 distinct research clusters, differentiated by various colors and sizes, representing different research topics over the past 24 years. Cluster #0 "Erythema nodosum" and Cluster #1 "Corticosteroid" are larger clusters, with #0 comprising 123 papers mainly focused on various diagnostic methods, clinical manifestations, and treatment options for GLM, and #1 consisting of 67 papers addressing clinical manifestations and recurrence. Earlier research topics in this field include Cluster #6 "breast tumor", #8 and #10 "breast mass", #11 "breast abstraction", #4 "steroid therapy", and #5 "granulomatous infection". The most recent research hotspots are Clusters #0 "erythema nodosum", #3 "cystic neutrophilic granulomatous mastitis", and #2 "meta-analysis".

Figure 8 illustrates the top 20 references with the strongest citation impact. The citation bursts began as early as 2004, with all references having a burst intensity greater than 7. The reference with the most significant burst (Strength=10.94), titled "Granulomatous mastitis: Presentations, diagnosis, treatment, and outcome in 206 patients from the north of Iran", was published in 2016. Table S2 summarizes the detailed information of these top 20 references in the sequence depicted in Figure 8.

Analysis of Keywords Co-Occurrence and Hotspots

A total of 1549 keywords were identified in the dataset of this study, with the 30 most common keywords listed in Table S3. The keywords management, diagnosis, lobular mastitis, diagnosis and therapy emerged as the top five, indicating significant attention in the GLM field (Figure 9).

The keyword co-occurrence network, constructed using 78 keywords that appeared at least five times, was segmented into six clusters (Figure 10). Cluster 1 (red) focused on the etiology of GLM, featuring keywords such as Corynebacterium, infection, and prolactin. Cluster 2 (green) also addressed some causes of GLM, like lesions and breast lesions, but concentrated more on diagnosis and differential diagnosis. This cluster included differential diagnosis
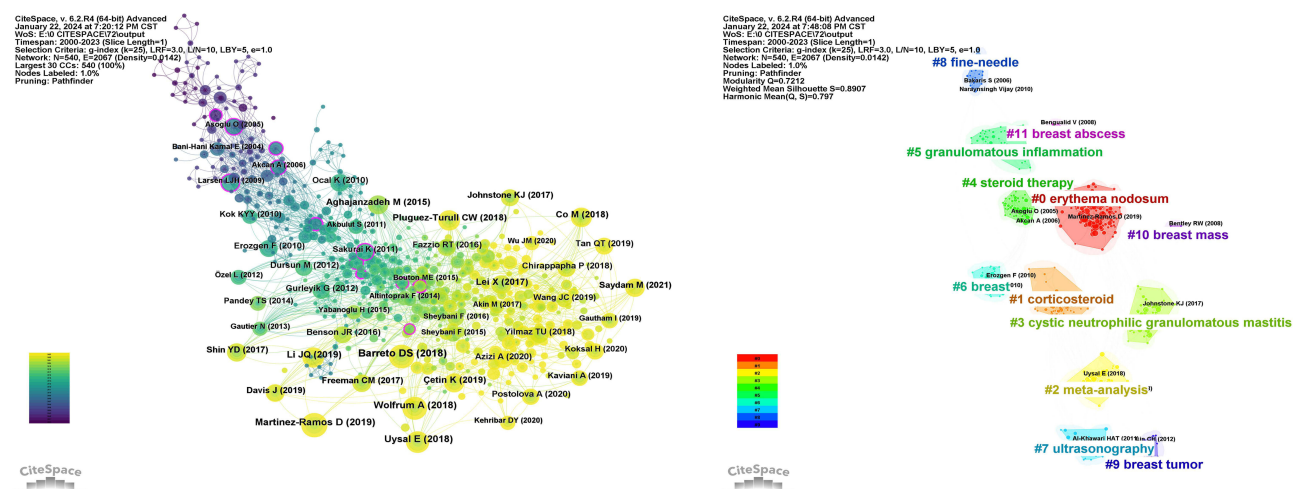

Figure 7 Visualization map of co-cited references and clustering network. The left image maps each reference by citation frequency, with purple nodes for older and yellow for more recent references; purple circles indicate betweenness centrality. The right image scales each circle by betweenness centrality.

Table 5 Top 10 Co-Cited References in the Field of GLM

Title	First Author	Year	Journal	Co-Citation	Centrality
Granulomatous mastitis: etiology, imaging, pathology, treatment, and clinical findings[25]	David S Barreto	2018	Breast Cancer Research and Treatment	40	0.02
Factors related to recurrence of idiopathic granulomatous mastitis: what do we learn from a multicentre study[26]	Erdal Uysal	2018	ANZ Journal of Surgery	37	0.07
Idiopathic granulomatous mastitis: A systematic review of 3060 patients[27]	David Martinez-Ramos	2019	Breast Journal	36	0.01
Granulomatous Mastitis: A Therapeutic and Diagnostic Challenge[28]	Angelika Wolfrum	2018	Breast Care	34	0.01
Diagnosis and Treatment of 75 Patients with Idiopathic Lobular Granulomatous Mastitis[29]	Jieqing Li	2018	Journal of Investigative Surgery	30	0.06
Idiopathic Granulomatous Mastitis: Manifestations at Multimodality Imaging and Pitfalls[30]	Cedric W Pluguez-Turull	2018	Radiographics	27	0.02
Idiopathic granulomatous mastitis: a 10-year study from a multicentre clinical database[31]	Michael Co	2018	Pathology	26	0.04
Comparison of Topical, Systemic, and Combined Therapy with Steroids on Idiopathic Granulomatous Mastitis: A Prospective Randomized Study[32]	Kenan Çetin	2019	World Journal of Surgery	25	0.03
Granulomatous mastitis: Presentations, diagnosis, treatment and outcome in 206 patients from the north of Iran[33]	Manouchehr Aghajanzadeh	2015	Breast	25	0.05
Treatments for Idiopathic Granulomatous Mastitis: Systematic Review and Meta-Analysis[7]	Xin Lei	2017	Breastfeeding Medicine	25	0.07

methods such as fine-needle aspiration, ultrasonography, MRI, and mammography, along with diseases requiring differentiation such as breast tuberculosis, breast cancer, and other cancers. Clusters 3 (blue) and 4 (yellow) discussed GLM treatment options, including surgical treatment, steroid, corticosteroid, recurrence, management, and more. Clusters 5 (purple) and 6 (bright blue) both referenced erythema nodosum, a complication of GLM, and conditions requiring differential diagnosis, such as breast tuberculosis and other types of mastitis.

VOSviewer utilized distinct colors to represent keywords based on their average appearance year, as depicted in Figure 11. Here, keywords like recurrence, hyperprolactinemia, corynebacterium, women, and infection are highlighted as recent research hotspots. Additionally, the time trend analysis of the top 26 keywords is shown in Figure 12, indicating that management, diagnosis, lobular mastitis, disease, and therapy have been the most frequently studied topics.

Discussion

Analysis of Research Hotspots in GLM Field

The keyword group depicted in Figure 10 illustrates the evolution of research topics in the GLM field over time, while Figure 11 highlights shifts in research focus. Initial topics such as mammography, diagnosis, ultrasonography, MRI, needle-aspiration-cytology, cytology, and abscess dominated the early stages of GLM research. As time progressed, themes like management, breast cancer, lesions, erythema nodosum, steroids, treatment, and surgery have emerged into the research spotlight. Recent hotspots and focal areas now include infection, Corynebacterium, prolactin, recurrence, meta-analysis, and association. This trajectory not only mirrors the typical progression in our understanding of a novel disease—from clinical manifestations and diagnostic approaches to differential diagnosis, treatment options, and complications—but also toward deeper etiological investigations and diverse research methodologies. Concurrently, this progression demonstrates the evolving nature of treatment strategies as clinicians and scientists search for the most effective interventions. With advances in the field, the comprehensive understanding of GLM has deepened, leading to

Top 20 References with the Strongest Citation Bursts

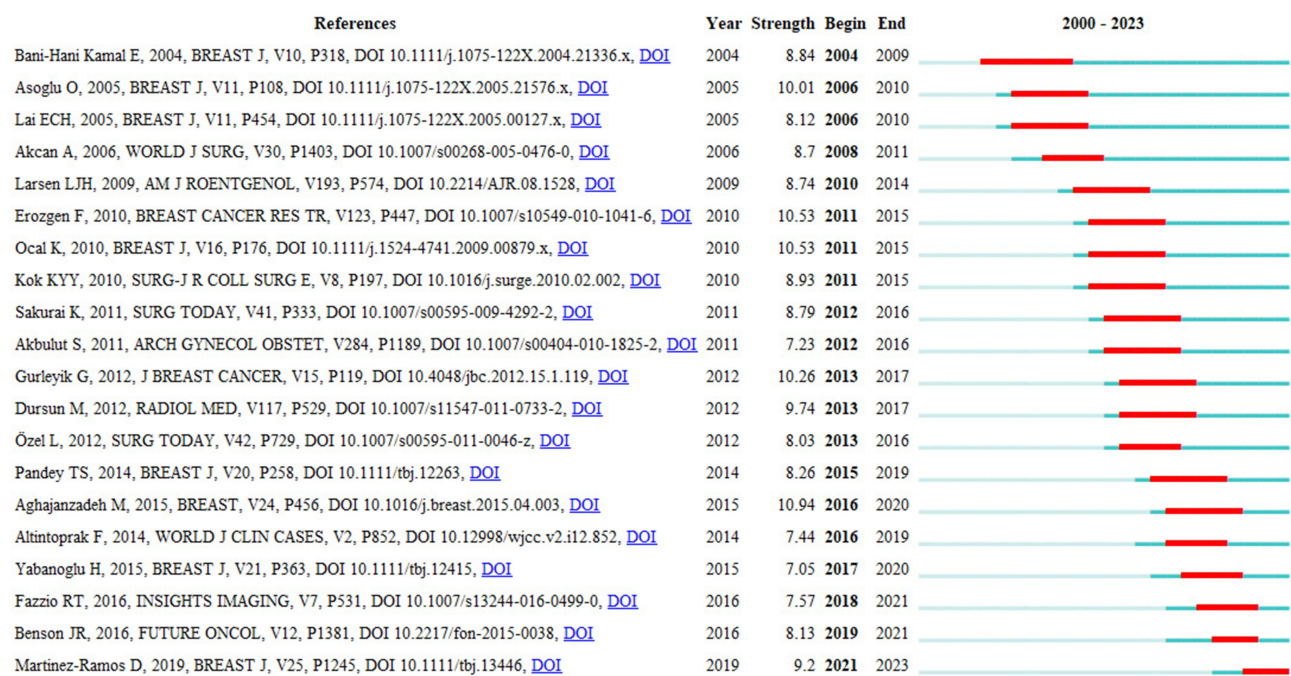| References	Year	Strength	Begin	End	2000 - 2023
Bani-Hani Kamal E, 2004, BREAST J, V10, P318, DOI 10.1111/j.1075-122X.2004.21336.x, DOI	2004	8.84	2004	2009	
Asoglu O, 2005, BREAST J, V11, P108, DOI 10.1111/j.1075-122X.2005.21576.x, DOI	2005	10.01	2006	2010	
Lai ECH, 2005, BREAST J, V11, P454, DOI 10.1111/j.1075-122X.2005.00127.x, DOI	2005	8.12	2006	2010	
Akcan A, 2006, WORLD J SURG, V30, P1403, DOI 10.1007/s00268-005-0476-0, DOI	2006	8.7	2008	2011	
Larsen LJH, 2009, AM J ROENTGENOL, V193, P574, DOI 10.2214/AJR.08.1528, DOI	2009	8.74	2010	2014	
Erozgen F, 2010, BREAST CANCER RES TR, V123, P447, DOI 10.1007/s10549-010-1041-6, DOI	2010	10.53	2011	2015	
Ocal K, 2010, BREAST J, V16, P176, DOI 10.1111/j.1524-4741.2009.00879.x, DOI	2010	10.53	2011	2015	
Kok KYY, 2010, SURG-J R COLL SURG E, V8, P197, DOI 10.1016/j.surge.2010.02.002, DOI	2010	8.93	2011	2015	
Sakurai K, 2011, SURG TODAY, V41, P333, DOI 10.1007/s00595-009-4292-2, DOI	2011	8.79	2012	2016	
Akbulut S, 2011, ARCH GYNECOL OBSTET, V284, P1189, DOI 10.1007/s00404-010-1825-2, DOI	2011	7.23	2012	2016	
Gurleyik G, 2012, J BREAST CANCER, V15, P119, DOI 10.4048/jbc.2012.15.1.119, DOI	2012	10.26	2013	2017	
Dursun M, 2012, RADIOL MED, V117, P529, DOI 10.1007/s11547-011-0733-2, DOI	2012	9.74	2013	2017	
Özel L, 2012, SURG TODAY, V42, P729, DOI 10.1007/s00595-011-0046-z, DOI	2012	8.03	2013	2016	
Pandey TS, 2014, BREAST J, V20, P258, DOI 10.1111/tbj.12263, DOI	2014	8.26	2015	2019	
Aghajanzadeh M, 2015, BREAST, V24, P456, DOI 10.1016/j.breast.2015.04.003, DOI	2015	10.94	2016	2020	
Altintoprak F, 2014, WORLD J CLIN CASES, V2, P852, DOI 10.12998/wjcc.v2.i12.852, DOI	2014	7.44	2016	2019	
Yabanoglu H, 2015, BREAST J, V21, P363, DOI 10.1111/tbj.12415, DOI	2015	7.05	2017	2020	
Fazzio RT, 2016, INSIGHTS IMAGING, V7, P531, DOI 10.1007/s13244-016-0499-0, DOI	2016	7.57	2018	2021	
Benson JR, 2016, FUTURE ONCOL, V12, P1381, DOI 10.2217/fon-2015-0038, DOI	2016	8.13	2019	2021	
Martinez-Ramos D, 2019, BREAST J, V25, P1245, DOI 10.1111/tbj.13446, DOI	2019	9.2	2021	2023	

Figure 8 Top 20 references with the strongest citation bursts in GLM research.

new insights into its etiology, high-risk factors, subtypes, complications, and recurrence. These elements have increasingly become the focus of contemporary research.

Hotspot 1: GLM and Corynebacterium

Recent advances in microbial identification technologies have increasingly affirmed the association between Corynebacterium infection and GLM.[34] Currently, there are approximately 100 species of Corynebacterium, a diverse group of Gram-positive bacteria (GPB). Most clinical isolates of Corynebacterium are derived from female patients, predominantly from those with breast abscesses and GLM.[35] Often, the breast tissue of GLM patients harbors few bacteria, which might result in negative Results during pathological examinations. A notable method to improve the detection rate and facilitate easier identification involves the use of "thick section" Gram staining. Research by Sangoi[36] has demonstrated that increasing the thickness of the slice can improve the detection rate of GPB. Additional techniques being explored include immunostaining with low specificity antiserum, 16S rRNA gene sequencing, and rpoB gene sequencing to accurately identify Corynebacterium.[37,38] Corynebacterium kroppenstedtii is considered a challenging pathogen associated with GLM.[39] Zeng[40] reported that the recurrence rate of treatment combining steroids with antibacterial drugs was significantly lower than that of steroids alone. Commonly chosen antimicrobial agents include doxycycline, amoxicillin, ciprofloxacin, and cefuroxime.[41] Lipophilic antibacterial drugs are preferred for their effective tissue distribution, thus increasing their antibacterial efficacy.[42,43]

Hotspot 2: GLM and Hyperprolactinemia

Hyperprolactinemia (HPRL) is increasingly recognized as a high-risk factor influencing GLM, particularly notable in women of childbearing age, often post-breastfeeding. Some researchers suggest that GLM might be linked to elevated prolactin (PRL) levels, though the exact mechanism remains undefined.[44] HPRL is also associated with several autoimmune diseases, including lupus erythematosus and rheumatoid arthritis.[45] An autoimmune response mediated by Th1 and Th2 cells is stimulated by high PRL levels, which in turn promotes the production of pro-inflammatory cytokines such as interleukin-1 (IL-1), IL-2, and IL-6; these cytokines further stimulate prolactin secretion.[46]

Tree

Figure 9 Tree map of keywords. The size of the module shows the proportion of keywords.

Moreover, PRL interferes with immune regulation by inhibiting the negative selection of autoimmune B lymphocytes, thereby promoting autoimmune reactions.[47] Prolactin receptors, widely expressed on immune cells, upon binding to prolactin, activate downstream signaling pathways that control the proliferation, differentiation, secretion, and survival of immune cells.[48] Drug-induced HPRL accounts for a significant portion of cases, with traditional antipsychotics being the most common culprits, associated with an incidence rate of 4.0%-70.0%.[49] A retrospective study indicated that increased prolactin levels in GLM patients were linked to the use of dopamine receptor antagonists.[50] These antipsychotics block dopamine receptors in the hypothalamus, inhibit PRL gene expression, and thus lead to HPRL.[51]

Hotspot 3: Complications of GLM

The extramammary manifestations of GLM encompass inflammatory arthritis, joint pain, episcleritis, and Erythema nodosum (EN), often accompanied by significant increases in systemic reactions, inflammation, and immune-related markers, indicative of an underlying immune process.[52,53] Azizi[54] reported that 4.6% (22/474) of patients with GLM experienced joint pain. The first case of GLM associated with EN was documented in 1987;[55] however, due to GLM's low incidence, EN is even rarer and considered an occasional manifestation.[3] Research suggests that the occurrence of EN may be related to GLM lesions extending over two quadrants and/or the presence of GLM in both breasts. Compared to patients with only GLM, those with EN typically experience a longer disease course.[56,57] A few patients initially present with EN and/or joint pain as the primary symptoms.[58]

Although no definitive evidence of an autoimmune link exists in cases of GLM with EN, the disappearance of EN following surgical removal of breast lesions[59] and the effectiveness of glucocorticoid treatment[60] strongly imply that EN is related to GLM
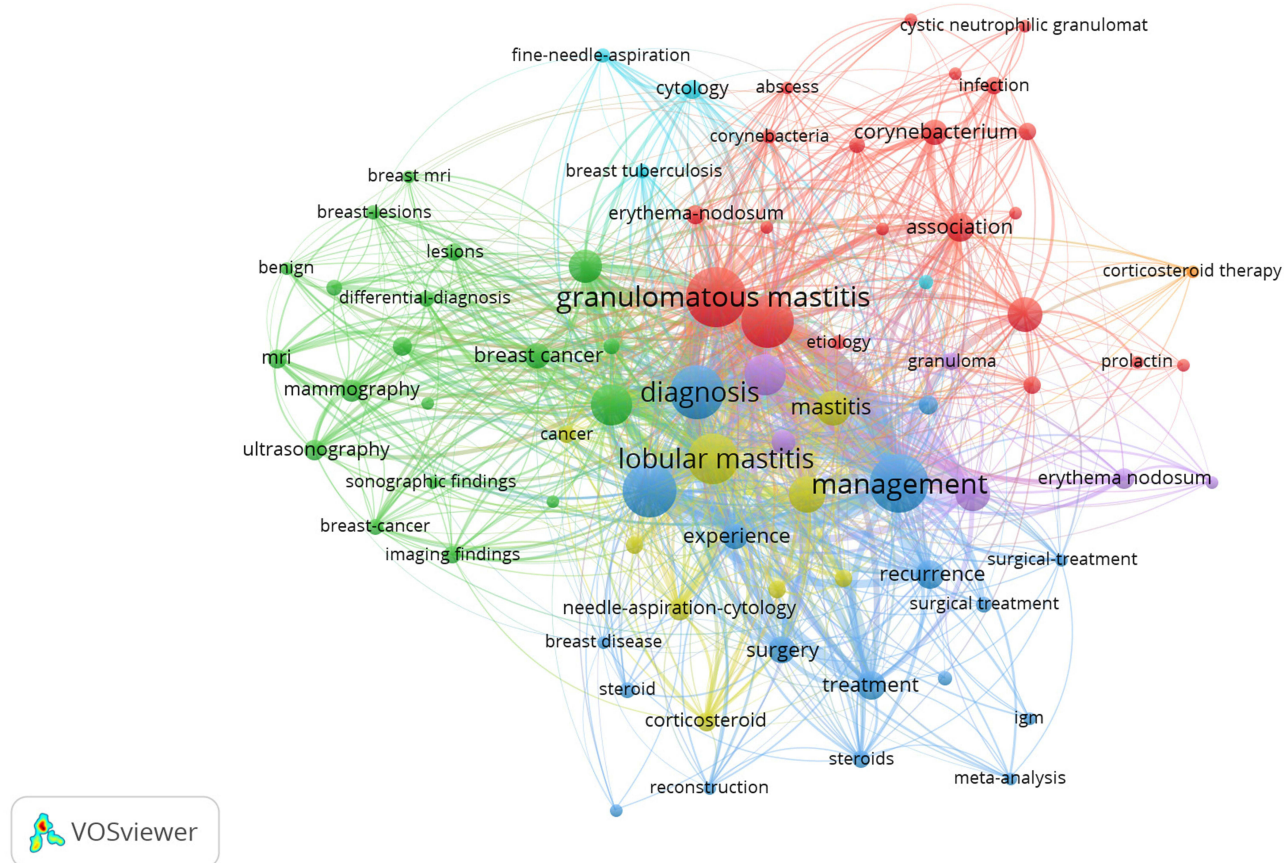
Figure 10 Co-occurrence network of keywords with a minimum frequency of five. Nodes are sized based on keyword occurrence, with lines showing linkages between keywords and colors representing different clusters.
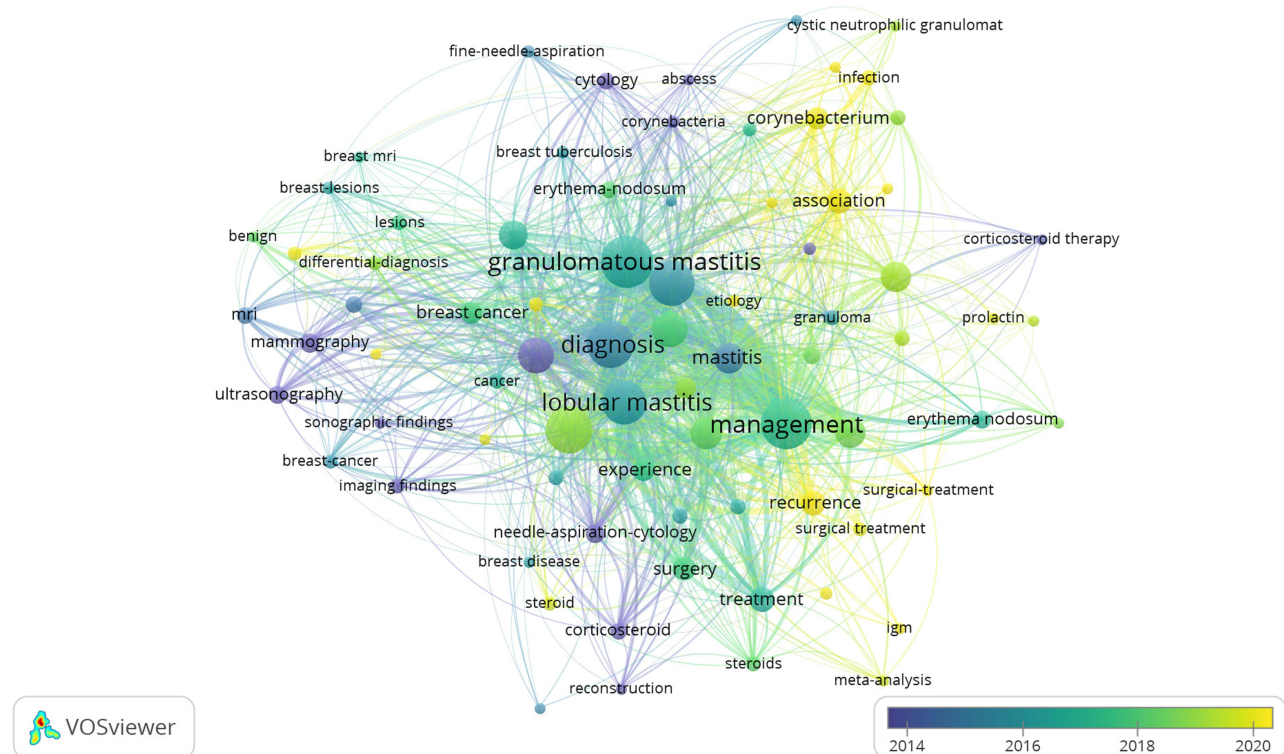

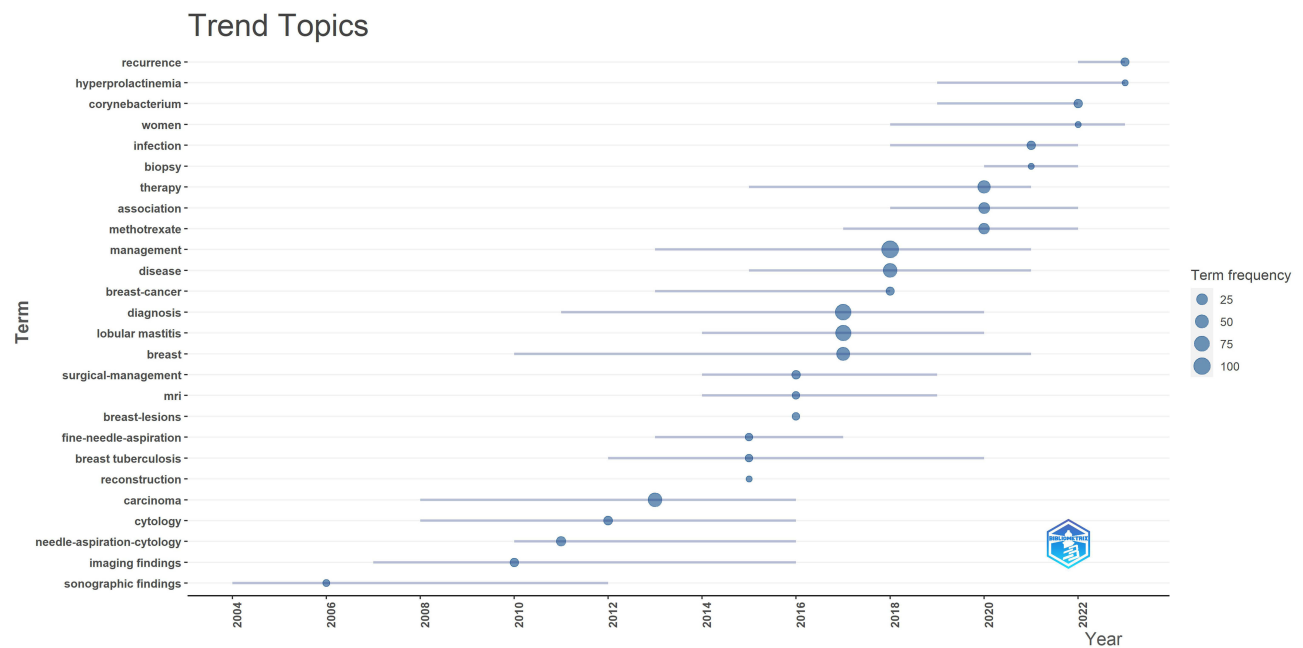**Figure 11** A chronological overview of the co-occurrence map of author keywords based on average appearance year.

Trend Topics

Figure 12 Trend of top 26 the most frequent keywords in GLM research.

and suggest a probable immune mechanism in its pathogenesis. GLM patients generally exhibit a chronic and recurrent course of the disease,[61] with a recurrence rate of 15.4%-24.8%.[62] The risk of recurrence increases 2.16–2.64 times with Corynebacterium infection,[63] and the recurrence rate for breast skin lesions is notably higher.[54] The difference in PRL levels before and after treatment serves as an independent risk factor for recurrence, with patients exhibiting higher PRL levels post-treatment facing a greater risk of recurrence.[64]

Summarizing 24 years of research reveals that the field predominantly consists of single-center case studies,[65,66] and the findings are not widely applicable. Due to GLM's rarity, there is a dearth of extensive, multi-center clinical validation, mechanistic research, or systematic reviews based on a substantial number of published data.

Limitations

Firstly, this study exclusively retrieved publications from the WOS, omitting publications from other databases such as PubMed and Embase, which might introduce selection bias. Secondly, the limitation in the types of literature and languages included in the search strategy may have prevented the identification of all relevant references. Additionally, due to the continuous updates of the database, there is a discrepancy between the search results of this study and the actual number of articles collected. Nevertheless, we anticipate that future research will address these Limitations to the fullest extent possible.

Conclusion

Our bibliometric analysis outlined the global landscape and research evolution in the GLM field over the past 24 years. The study highlighted that research on GLM has gained significant attention in recent years, with a steady increase in publications. We pinpointed the most influential countries, institutions, and journals in this field. Current research predominantly focuses on the etiology, differential diagnosis, treatment, and comprehensive management of GLM. However, numerous challenges and obstacles remain. Additionally, recurrence, hyperprolactinemia, and Corynebacterium have emerged as current research hotspots and potential future research frontiers. Our findings are instrumental in identifying research hotspots and trends, offering insights and new directions for future investigations.

Ethics Approval and Consent to Participate

The data of this study are all from open databases, so it does not involve ethical issues.

Author Contributions

All authors made a significant contribution to the work reported, whether that is in the conception, study design, execution, acquisition of data, analysis and interpretation, or in all these areas; took part in drafting, revising or critically reviewing the article; gave final approval of the version to be published; have agreed on the journal to which the article has been submitted; and agree to be accountable for all aspects of the work.

Funding

Disclosure

The authors report no conflicts of interest in this work.

References

1. Zheng B, Lu M, Chen C, et al. The distinct pattern of granulomatous lobular mastitis with erythema nodosum: more severe conditions and extensive lesions. *J Invest Surg*. 2023;36(1):2257770. doi:10.1080/08941939.2023.2257770

2. Patel RA, Strickland P, Sankara IR, et al. Idiopathic granulomatous mastitis: case reports and review of literature. *J Gen Intern Med*. 2010;25 (3):270–273. doi:10.1007/s11606-009-1207-2

3. Sheybani F, Sarvghad M, Naderi H, et al. Treatment for and clinical characteristics of granulomatous mastitis. *Obstet Gynecol*. 2015;125 (4):801–807. doi:10.1097/AOG.0000000000000734

4. Boldrini C. Granulomatous mastitis (GM) in a young woman with a previous history of prolactin-secreting PitNET and actual normal prolactinemia. *Radiol Case Rep*. 2022;18(2):550–555. doi:10.1016/j.radcr.2022.11.012

5. Yin Y, Liu X, Meng Q, et al. Idiopathic granulomatous mastitis: etiology, clinical manifestation, diagnosis and treatment. *J Invest Surg*. 2022;35 (3):709–720. doi:10.1080/08941939.2021.1894516

6. Karanlik H, Ozgur I, Simsek S, et al. Can steroids plus surgery become a first-line treatment of idiopathic granulomatous mastitis? *Breast Care*. 2014;9(5):338–342. doi:10.1159/000366437

7. Lei X, Chen K, Zhu L, et al. Treatments for idiopathic granulomatous mastitis: systematic review and meta-analysis. *Breastfeed Med*. 2017;12 (7):415–421. doi:10.1089/bfm.2017.0030

8. Hassan W, Yekta BG, Nabavi SM, et al. The progress and research trends of statin medications: advanced epidemiological and bibliometrical assessment. *Curr Probl Cardiol*. 2023;48(6):101638. doi:10.1016/j.cpcardiol.2023.101638

9. Xia DM, Wang XR, Zhou PY, et al. Research progress of heat stroke during 1989–2019: a bibliometric analysis. *Mil Med Res*. 2021;8(1):5. doi:10.1186/s40779-021-00300-z

10. Shen L, Wang S, Dai W, et al. Detecting the interdisciplinary nature and topic hotspots of robotics in surgery: social network analysis and bibliometric study. *J Med Internet Res*. 2019;21(3):e12625. doi:10.2196/12625

11. Wang A, Ju Y, Bi C. Scientometric analysis of researches on Tai Chi and health promotion based on literatures from 1991 to 2021. *Ann Palliat Med*. 2022;11(12):3648–3662. doi:10.21037/apm-22-843

12. van Eck NJ, Waltman L. Software survey: vOSviewer, a computer program for bibliometric mapping. *Scientometrics*. 2010;84(2):523–538. doi:10.1007/s11192-009-0146-3

13. Chen C. Searching for intellectual turning points: progressive knowledge domain visualization. *Proc Natl Acad Sci U S A*. 2004;101(Suppl 1):5303–5310. doi:10.1073/pnas.0307513100

14. Taylor GB, Paviour SD, Musaad S, et al. A clinicopathological review of 34 cases of inflammatory breast disease showing an association between corynebacteria infection and granulomatous mastitis. *Pathology*. 2003;35(2):109–119.

15. Hovanessian Larsen LJ, Peyvandi B, Klipfel N, et al. Granulomatous lobular mastitis: imaging, diagnosis, and treatment. *AJR Am J Roentgenol*. 2009;193(2):574–581. doi:10.2214/AJR.08.1528

16. Lai EC, Chan WC, Ma TK, et al. The role of conservative treatment in idiopathic granulomatous mastitis. *Breast J*. 2005;11(6):454–456. doi:10.1111/j.1075-122X.2005.00127.x

17. Al-Khaffaf B, Knox F, Bundred NJ. Idiopathic granulomatous mastitis: a 25-year experience. *J Am Coll Surg*. 2008;206(2):269–273. doi:10.1016/j. jamcollsurg.2007.07.041

18. Asoglu O, Ozmen V, Karanlik H, et al. Feasibility of surgical management in patients with granulomatous mastitis. *Breast J*. 2005;11(2):108–114. doi:10.1111/j.1075-122X.2005.21576.x

19. Baslaim MM, Khayat HA, Al-Amoudi SA. Idiopathic granulomatous mastitis: a heterogeneous disease with variable clinical presentation. *World J Surg*. 2007;31(8):1677–1681. doi:10.1007/s00268-007-9116-1

20. Akcan A, Akyildiz H, Deneme MA, et al. Granulomatous lobular mastitis: a complex diagnostic and therapeutic problem. *World J Surg*. 2006;30 (8):1403–1409. doi:10.1007/s00268-005-0476-0

21. Erhan Y, Veral A, Kara E, et al. A clinicopthologic study of a rare clinical entity mimicking breast carcinoma: idiopathic granulomatous mastitis. *Breast*. 2000;9(1):52–56. doi:10.1054/brst.1999.0072

22. Azlina AF, Ariza Z, Arni T, et al. Chronic granulomatous mastitis: diagnostic and therapeutic considerations. *World J Surg*. 2003;27(5):515–518. doi:10.1007/s00268-003-6806-1

23. Altintoprak F, Kivilcim T, Ozkan OV. Aetiology of idiopathic granulomatous mastitis. *World J Clin Cases*. 2014;2(12):852–858. doi:10.12998/wjcc.v2.i12.852

24. Leydesdorff L, Comins JA, Sorensen AA, et al. Cited references and Medical Subject Headings (MeSH) as two different knowledge representations: clustering and mappings at the paper level. *Scientometrics*. 2016;109(3):2077–2091.

25. Barreto DS, Sedgwick EL, Nagi CS, et al. Granulomatous mastitis: etiology, imaging, pathology, treatment, and clinical findings. *Breast Cancer Res Treat*. 2018;171(3):527–534. doi:10.1007/s10549-018-4870-3

26. Uysal E, Soran A, Sezgin E; Granulomatous Mastitis Study Group. Factors related to recurrence of idiopathic granulomatous mastitis: what do we learn from a multicentre study? *ANZ J Surg*. 2018;88(6):635–639. doi:10.1111/ans.14115

27. Martinez-Ramos D, Simon-Monterde L, Suelves-Piqueres C, et al. Idiopathic granulomatous mastitis: a systematic review of 3060 patients. *Breast J*. 2019;25(6):1245–1250. doi:10.1111/tbj.13446

28. Wolfrum A, Kümmel S, Theuerkauf I, et al. Granulomatous mastitis: a therapeutic and diagnostic challenge. *Breast Care*. 2018;13(6):413–418. doi:10.1159/000495146

29. Li J. Diagnosis and treatment of 75 patients with idiopathic lobular granulomatous mastitis. *J Invest Surg*. 2019;32(5):414–420. doi:10.1080/08941939.2018.1424270

30. Pluguez-Turull CW, Nanyes JE, Quintero CJ, et al. Idiopathic granulomatous mastitis: manifestations at multimodality imaging and pitfalls. *Radiographics*. 2018;38(2):330–356. doi:10.1148/rg.2018170095

31. Co M, Cheng VCC, Wei J, et al. Idiopathic granulomatous mastitis: a 10-year study from a multicentre clinical database. *Pathology*. 2018;50 (7):742–747. doi:10.1016/j.pathol.2018.08.010

32. Çetin K, Sıkar HE, Göret NE, et al. Comparison of topical, systemic, and combined therapy with steroids on idiopathic granulomatous mastitis: a prospective randomized study. *World J Surg*. 2019;43(11):2865–2873. doi:10.1007/s00268-019-05084-x

33. Aghajanzadeh M, Hassanzadeh R, Alizadeh Sefat S, et al. Granulomatous mastitis: presentations, diagnosis, treatment and outcome in 206 patients from the north of Iran. *Breast*. 2015;24(4):456–460. doi:10.1016/j.breast.2015.04.003

34. Saraiya N, Corpuz M. Corynebacterium kroppenstedtii: a challenging culprit in breast abscesses and granulomatous mastitis. *Curr Opin Obstet Gynecol*. 2019;31(5):325–332. doi:10.1097/GCO.0000000000000541

35. Tauch A, Fernández-Natal I, Soriano F. A microbiological and clinical review on Corynebacterium kroppenstedtii. *Int J Infect Dis*. 2016;48:33–39. doi:10.1016/j.ijid.2016.04.023

36. Sangoi AR. "Thick Section" gram stain yields improved detection of organisms in tissue sections of cystic neutrophilic granulomatous mastitis. *Am J Clin Pathol*. 2020;153(5):593–597. doi:10.1093/ajcp/aqz200

37. Fujii M, Mizutani Y, Sakuma T, et al. Corynebacterium kroppenstedtii in granulomatous mastitis: analysis of formalin-fixed, paraffin-embedded biopsy specimens by immunostaining using low-specificity bacterial antisera and real-time polymerase chain reaction. *Pathol Int*. 2018;68 (7):409–418. doi:10.1111/pin.12683

38. Bao R, Gao X, Hu B, et al. Matrix-assisted laser desorption ionization time-of-flight mass spectrometry: a powerful tool for identification of Corynebacterium species. *J Thorac Dis*. 2017;9(9):3239–3245. doi:10.21037/jtd.2017.09.69

39. Collins MD, Falsen E, Akervall E, et al. Corynebacterium kroppenstedtii sp. nov. a novel Corynebacterium that does not contain mycolic acids. *Int J Syst Bacteriol*. 1998;48(Pt 4):1449–1454. doi:10.1099/00207713-48-4-1449

40. Zeng W, Lao S, Jia W, et al. Clinical features and recurrence of Corynebacterium kroppenstedtii infection in patients with mastitis. *BMC Women's Health*. 2022;22(1):276. doi:10.1186/s12905-022-01859-y

41. Le Flèche-Matéos A, Berthet N, Lomprez F, et al. Recurrent breast abscesses due to Corynebacterium kroppenstedtii, a human pathogen uncommon in caucasian women. *Case Rep Infect Dis*. 2012;2012:120968. doi:10.1155/2012/120968

42. Sanchez Eluchans N, Barberis C, Cittadini R, et al. Corynebacterium kroppenstedtii breast infections: report of four cases. *Rev Argent Microbiol*. 2021;53(4):304–308. Spanish. doi:10.1016/j.ram.2021.01.002

43. Dobinson HC, Anderson TP, Chambers ST, et al. Antimicrobial Treatment Options for Granulomatous Mastitis Caused by Corynebacterium Species. *J Clin Microbiol*. 2015;53(9):2895–2899. doi:10.1128/JCM.00760-15

44. Li J, McGregor HP. Idiopathic granulomatous mastitis associated with hyperprolactinemia: a nonoperative approach. *Breast J*. 2017;23(6):742–744. doi:10.1111/tbj.12914

45. Buskila D, Sukenik S, Shoenfeld Y. The possible role of prolactin in autoimmunity. *Am J Reprod Immunol*. 1991;26(3):118–123. doi:10.1111/j.1600-0897.1991.tb00708.x

46. Vieira Borba V, Shoenfeld Y. Prolactin, autoimmunity, and motherhood: when should women avoid breastfeeding? *Clin Rheumatol*. 2019;38 (5):1263–1270. doi:10.1007/s10067-018-04415-y

47. Ewerman L, Landberg E, Hellberg S, et al. Immunomodulating effects depend on prolactin levels in patients with hyperprolactinemia. *Horm Metab Res*. 2020;52(4):228–235. doi:10.1055/a-1126-4272

48. Jeganathan V, Peeva E, Diamond B. Hormonal milieu at time of B cell activation controls duration of autoantibody response. *J Autoimmun*. 2014;53:46–54. doi:10.1016/j.jaut.2014.02.007

49. An FR, Yang R, Wang ZM, et al. Hyperprolactinemia, prolactin-related side effects and quality of life in Chinese psychiatric patients. *Compr Psychiatry*. 2016;71:71–76. doi:10.1016/j.comppsych.2016.08.009

50. Nikolaev A, Blake CN, Carlson DL. Association between hyperprolactinemia and granulomatous mastitis. *Breast J*. 2016;22(2):224–231. doi:10.1111/tbj.12552

51. Tian C, Wang H, Liu Z, et al. Characteristics and management of granulomatous lobular mastitis associated with antipsychotics-induced hyperprolactinemia. *Breastfeed Med*. 2022;17(7):599–604. doi:10.1089/bfm.2021.0341

52. Zabetian S, Friedman BJ, McHargue C. A case of idiopathic granulomatous mastitis associated with erythema nodosum, arthritis, and reactive cough. *JAAD Case Rep*. 2016;2(2):125–127. doi:10.1016/j.jdcr.2016.01.011

53. Jacquin-Porretaz C, Devalland C, Delapparent T, et al. Idiopathic granulomatous mastitis associated with erythema nodosum. *Ann Dermatol Venereol*. 2019;146(8–9):571–576. doi:10.1016/j.annder.2019.04.023

54. Azizi A, Prasath V, Canner J, et al. Idiopathic granulomatous mastitis: management and predictors of recurrence in 474 patients. *Breast J*. 2020;26 (7):1358–1362. doi:10.1111/tbj.13822

55. Adams DH, Hubscher SG, Scott DG. Granulomatous mastitis--a rare cause of erythema nodosum. *Postgrad Med J*. 1987;63(741):581–582. doi:10.1136/pgmj.63.741.581

56. Luo W, Xu B, Wang L, et al. Clinical characteristics and predictive factors of erythema nodosum in granulomatous lobular mastitis. *Australas J Dermatol*. 2021;62(3):342–346. doi:10.1111/ajd.13640

57. Çetin K, Sıkar HE, Güllüoğlu BM. Idiopathic granulomatous mastitis with erythema nodosum: is it a variant of clinical presentation indicating treatment resistance? A retrospective cohort study. *Breast J*. 2020;26(9):1645–1651. doi:10.1111/tbj.13944

58. Salesi M, Karimifar M, Salimi F, et al. A case of granulomatous mastitis with erythema nodosum and arthritis. *Rheumatol Int*. 2011;31 (8):1093–1095. doi:10.1007/s00296-009-1273-0

59. Polat M, Kaya H. Plantar erythema nodosum associated with granulomatous mastitis. *Indian J Dermatol Venereol Leprol*. 2016;82(2):202–204. doi:10.4103/0378-6323.164219

60. Gümüş M, Akkurt ZM, Gümüş H. Is erythema nodosum coexisting with lesions of the breast a suggestive sign for idiopathic granulomatous mastitis? *Turk J Surg*. 2018;34(1):71–73. doi:10.5152/turkjsurg.2017.3161

61. Tasci HI, Turk E, Erinanc OH, et al. Factors affecting recurrence of idiopathic granulomatous mastitis. *J Coll Physicians Surg Pak*. 2022;32 (2):161–165. doi:10.29271/jcpsp.2022.02.161

62. Yuan QQ, Xiao SY, Farouk O, et al. Management of granulomatous lobular mastitis: an international multidisciplinary consensus (2021 edition). *Mil Med Res*. 2022;9(1):20. doi:10.1186/s40779-022-00380-5

63. Tan QT, Tay SP, Gudi MA, et al. Granulomatous mastitis and factors associated with recurrence: an 11-year single-centre study of 113 patients in Singapore. *World J Surg*. 2019;43(7):1737–1745. doi:10.1007/s00268-019-05014-x

64. Huang Y, Wu H. A retrospective analysis of recurrence risk factors for granulomatous lobular mastitis in 130 patients: more attention should be paied to prolactin level. *Ann Palliat Med*. 2021;10(3):2824–2831. doi:10.21037/apm-20-1972

65. Yabanoğlu H, Çolakoğlu T, Belli S, et al. A comparative study of conservative versus surgical treatment protocols for 77 patients with idiopathic granulomatous mastitis. *Breast J*. 2015;21(4):363–369. doi:10.1111/tbj.12415

66. Shin YD, Park SS, Song YJ, et al. Is surgical excision necessary for the treatment of Granulomatous lobular mastitis? *BMC Women's Health*. 2017;17(1):49. doi:10.1186/s12905-017-0412-0