Letter to the editor

Comment on idiopathic granulomatous mastitis: A systematic review of 3060 patients

Dear Editor,

We read the recent article “Idiopathic granulomatous mastitis: A systematic review of 3060 patients” published by Martinez-Ramos et al. [1] with great interest. We would like to commend the authors for this study which is prepared as a detailed literature review. However, we would like to emphasize several points regarding the present study.

Martinez-Ramos et al. [1] stated that they have performed a literature search including studies about idiopathic granulomatous mastitis (IGM) published between May 2013 and May 2018. However, it is seen that the authors have collected all the studies regardless of the study period and the institutions and have reported a total number of 3060 patients. This is an important issue because there are multiple studies from the same center written in different time intervals and all have been included which means there is an overlap in the number of patients and there are some patients that have been included in the analysis of the present study over and over again. Therefore, the actual patient number is less than 3060. Here with this commentary, we include a table of the 70 studies that have been used by Martinez-Ramos et al, and we have sorted these studies according to study period and the institutions. If our table is analyzed, there are least two articles from eight centers and the patient numbers have been included over and over again in the present systematic review. This is important because the number of patients that have been included more than once is significant.

The authors have stated that articles reporting less than 3 cases were excluded from the literature search. However, they have included a article reporting 2 cases [2]. In addition, Kutsuna et al. [2] have reported that these two patients had a specific cause underlying granulomatous mastitis (granulomatous mastitis associated with Corynebacterium spp.) which means these two cases cannot be considered as IGM.

In Table 1 presented by Martinez-Ramos et al, two studies by Alitnoprak et al in 2013 [3] and 2015 [4] are included and stated the number of patients as 28 and 26. Analysis of these two articles showed that reported number of patients was just the opposite. This unintentional mistake should be corrected.

It is seen that Martinez-Ramos et al. [1] have included a study by Oltean et al. [5] which reported 85 patients with chronic mastitis. Martinez-Ramos et al have considered these patients as IGM. Granulomatous mastitis is classified in to two groups as primary (idiopathic) and secondary according to presence of underlying disease that causes chronic mastitis. The majority of granulomatous mastitis is seen due to underlying chronic diseases such as tuberculosis, candida spp., Corynebacterium spp., Weber-Christian disease, Sjogren’s syndrome, SLE, hyperprolactinemia, IgG4 sclerosing disease, local trauma, cat scratch disease and are considered as secondary granulomatous mastitis. Authors have made a similar mistake that is common in literature and have considered these cases such as IGM. Another study from the same center showed that only 18.6% of the patients were IGM [6].

Authors have included a study by Allen et al. [6] which reported 43 patients with chronic mastitis. These patients were considered to be IGM but when the article was analyzed in detail, only 8 patients had the histopathologic characteristics consistent with granulomatous mastitis. Some of the remaining patients had IgG4 related breast disease. The IgG4 related breast disease (diagnostic criteria = dense lymphoplasmacytic infiltrate, storiform fibrosis, obliterative phlebitis, >10 IgG4 cells/hpf and >40% IgG4/IgG ratio) is significantly different from the IGM (diagnostic criteria = epithelioid histiocytes, vague or well-formed non-necrotizing granulomas, giant cells) in terms of pathologic characteristics.

Authors have also included the study by Troxell et al. [7] which reported 19 cases with cystic neutrophilic granulomatous mastitis. The histopathologic characteristics of cystic neutrophilic granulomatous mastitis are lobular centric inflammation, cystic spaces rimmed by neutrophils and lipo-granulomas and is usually associated with certain bacterial strains such as Corynebacterium spp. Similar criticism can be made for the inclusion of the study of Johnstone et al. [8]. In addition, the patients reported by Johnstone et al have been stated as 21 in Table 1 but in fact, they reported 15 patients with cystic neutrophilic granulomatous mastitis. In summary, these two studies report a secondary granulomatous mastitis and it is not appropriate to be included in the present study analyzing IGMS.

Pathologic features, biochemical analysis and clinical examination is used in combination for the diagnosis of IGM. Above mentioned criteria are pathognomonic for the pathologic evaluation of IGM. The study that belonged to Chu et al. [9] was also included for analysis which reported 12 patients with only 9 patients with a pathologic confirmation of IGM. Similarly, in the study of Wong et al [10] only 11 of the 37 patients had pathologic conformation of IGM. All the patients in these two studies were included in the present systematic review with the assumption that all the patients had IGM. This issue should be clarified by the authors.

The study by Boutfettal and colleagues [11] has been published as a hard copy in 2013 but in fact it was digitally printed in 2012 and therefore, it does not fit the time period stated by the authors.

The 19th reference which belonged to a study by Kaviani et al. [12] which is also included in Table 1 stated that 20 patients with IGM were reported. However, analysis of the article shows that the authors have analyzed 258 breast lesions and only 2 were IGM. Another study published in 2014 by Kaviani et al. [13] reports the management algorithm of 20 patients with IGM.

Table 1 states that Alungal et al. [14] from India reported 25 patients IGM. However, detailed analysis of the databases showed that there is only one case of 25-years old female patients with IGM and erythema nodosum that have been reported by this author. Therefore, the authors should share this particular study by Alungal et al. [14] and give the full citation.

https://doi.org/10.1016/j.ijscr.2021.106250

Received 27 June 2021; Accepted 27 July 2021

Available online 2 August 2021

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The credentials (tag) of three studies could not be evaluated because they could not be reached.

| Authors          | Year | Country       | City                  | Main center                      | Study period | Study type         | Study design        | Language  |
|------------------|------|---------------|-----------------------|----------------------------------|--------------|--------------------|---------------------|-----------|
| Akahane          | 2013 | Japan         | Aichi                 | Aichi Cancer Center              | 2005–2012    | Single Retrospective | English             |           |
| Al-Jarrah        | 2013 | Oman          | Muscat                | Sultan Qaboos University         | 2009–2012    | Single Prospective  | English             |           |
| Altintoprak      | 2013 | Turkey        | Sakarya               | Sakarya University               | 2007–2013    | Single Retrospective | English             |           |
| Boufetal         | 2012 | Morocco       | Casablanca            | Ibn Roch University              | 2000–2009    | Single Retrospective | English             |           |
| Cabrera          | 2013 | Spain         | Valencia              | Doctor Peert University          | 2010–2011    | Single Retrospective | Spanish             |           |
| Gautier          | 2013 | Canada        | Montreal              | Montreal University              | 2008–2011    | Single Retrospective | Spanish             |           |
| Hur              | 2013 | S. Korea      | Seoul                 | Soonchunhyang University         | 2005–2009    | Single Retrospective | English             |           |
| Mohammed         | 2013 | USA           | Houston               | Baylor College of Medicine       | 2000–2010    | Single Retrospective | English             |           |
| Olteran          | 2013 | Egypt         | Cairo                 | Egypt (3) Morocco (2)            | 2006–2012    | Single Retrospective | Spanish             |           |
| Omranipour       | 2013 | Iran          | Tehran                | Tehran University of Med Sci     | 2006–2012    | Single Retrospective | English             |           |
| Oran             | 2013 | Turkey        | Istanbul              | Haseki Research Hospital         | 1999–2011    | Multicentric Retrospective | English             |           |
| Zaragoza         | 2013 | Spain         | Alicante             | Vega Baja Hospital               | NS           | Single Case Report  | Spanish             |           |

In the Materials and methods section, it is stated that only the studies in English language are cited, however, there are 3 studies in Table 1 that are written in Spanish language [15–17]. This issue should also be clarified by the authors.

One very important point that is related with the mentioned study is the reference list that is given by the by Martinez-Ramos et al. [1]. Figure 1 in the mentioned article shows that 70 articles are in accord with the inclusion criteria of the present study. The mentioned authors have cited only 20 of these articles in the reference list. We understand the obligation that there may be some limitation to the number of
references required by the journal however, these references could have been cited in a separate Appendix. This would have provided a citation for the remaining 50 articles that were used in the present study. This is important for the efforts of the authors of those studies. We have prepared an Appendix including all the studies used by Martínez-Ramos et al. [1] and we hope that it will be supplemented to their article.

Ethical approval

None. Our paper is in the format of letter to editor.

Funding

This letter to editor did not receive any specific grant from funding agencies in the public, commercial, or not-for-profit sectors.

CRediT authorship contribution statement

Akbulut S and Sahin TT: Reviewed the literature and wrote the manuscript. Akbulut S and Sahin TT: Supervised the writing process and revised the manuscript.

Guarantor

Akbulut S and Sahin TT are the guarantors for the present commentary and they take full responsibility for the comments and the auxiliary data presented in the commentary article.

Research registration

Not applicable.

Consent

None. Our paper is in the format of letter to editor.

Declaration of competing interest

No conflict of interest about this letter to the editor.

Appendix A. Idiopathic granulomatous mastitis: a systematic review of 3060 patients

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