



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 Altintoprak 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 cases 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 confirmation 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 Bouffettal 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.

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**Table 1**

Basic characteristics of 67 of the 70 articles used in the study of Martinez-Ramos et al. Some patient numbers were corrected. 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 Rochd University	2000–2009	Single	Retrospective	English
Cabrera	2013	Spain	Valencia	Doctor Peset University	2010–2011	Single	Retrospective	Spanish
Gautier	2013	Canada	Montreal	Montreal University	2008–2011	Single	Retrospective	English
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)	2008–2011	Multicentric	Retrospective	English
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
Akcan	2014	Turkey	Kayseri	Erciyes University	1995–2012	Single	Retrospective	English
Cornejo-Juar	2014	Mexico	Mexico City	National Cancer Institute	2007–2011	Single	Retrospective	English
Gunduz	2014	Turkey	Sakarya	Sakarya University	2008–2013	Single	Retrospective	English
Handa	2014	USA	New York	Jacobi Medical Center	2006–2012	Single	Retrospective	English
Joseph	2014	USA	New York	Bellevue Hospital Center	2012–2014	Single	Retrospective	English
Karanlik	2014	Turkey	Istanbul	Istanbul University (Oncology)	2007–2011	Single	Prospective	English
Kaviani	2014	Iran	Tehran	Tehran University of Med Sci	2007–2011	Multicentric	Retrospective	English
Kiyat	2014	Turkey	Ankara	Ataturk Training Res Hospital	2006–2011	Single	Retrospective	English
Pandey	2014	USA	Chicago	John H. Stroger Jr. Hospital	2006–2010	Single	Prospective	English
Salehi	2014	Iran	Isfahan	Isfahan University Med Sci	2011–2013	Single	NS	English
Altintoprak	2015	Turkey	Sakarya	Sakarya University	2007–2014	Single	Retrospective	English
Atak	2015	Turkey	Istanbul	Goztepe Res Training Hospital	2008–2013	Single	Retrospective	English
Bouton	2015	USA	Phoenix	Maricopa Medical Center	2002–2013	Single	Retrospective	English
Ciftci	2015	Turkey	Istanbul	Gelisim University	NS	Single	Case Report	English
Durur	2015	Turkey	Erzurum	Ataturk University	2014–2014	Single	Retrospective	English
Gudimani	2015	India	Kannur	Kannur Medical Hospital	NS	Single	Case Report	English
Korkut	2015	Turkey	Erzurum	Ataturk University	2001–2010	Single	Retrospective	English
Kutsuna	2015	Japan	Tokyo	National Center Global Health	NS	Single	Case Report	English
Mahlab Guri	2015	Israel	Rehovot	Kaplan Medical Center	1999–2012	Single	Retrospective	English
Mizrakli	2015	Turkey	Istanbul	Istanbul University (Cerrahpasa)	2009–2011	Single	Retrospective	English
Sanint	2015	Spain	Barcelona	Germans Trias i Pujol University	2008–2014	Single	Retrospective	Spanish
Sheybani	2015	Iran	Mashhad	Imam Reza Teaching Hospital	2012–2014	Single	Prospective	English
Skandarajah	2015	Australia	Victoria	Melbourne University	2000–2013	Multicentric	Retrospective	English
Yabanoglu	2015	Turkey	Adana	Baskent University	1999–2013	Single	Retrospective	English
Yukawa	2015	Japan	Nara	Kinki University	2001–2013	Single	Retrospective	English
Ahmed	2016	Egypt	Alexandria	Alexandria University	2010–2013	Single	Prospective	English
Allen	2016	Egypt *	Cairo	Egypt (3) Morocco (2)	2008–2011	Multicentric	Retrospective	English
Bilal	2016	S. Arabia	Riyadh	King Khalid University	NS	Single	Retrospective	English
Elzahaby	2016	Egypt	Mansoura	Mansoura University	2013–2016	Single	NS	English
Fazzio	2016	USA	Rochester	Mayo Clinic	2000–2015	Single	Retrospective	English
Helal	2016	Egypt	Cairo	Ain Shams University	2008–2013	Single	Retrospective	English
Nikolaev	2016	USA	Florida	Florida Atlantic University	NS	Single	Case Report	English
Oztekin	2016	Turkey	Ankara	Ankara Training Res Hospital	2009–2013	Single	Retrospective	English
Troxell	2016	USA	Portland	Oregon Helath Sci University	2005–2015	Single	Retrospective	English
Velidedeoglu	2016	Turkey	Istanbul	Istanbul University (Cerrahpasa)	2010–2013	Single	Retrospective	English
Yilmaz	2018	Turkey	Kocaeli	Kocaeli University	2008–2014	Single	Retrospective	English
Aghajanzadeh	2015	Iran	Rasht	Guilan University Med Sci	2006–2013	Multicentric	Retrospective	English
Ail	2017	India	Maharashtra	Smt. Kashibai Navale Med Coll.	2009–2016	Single	Retrospective	English
Akin	2017	Turkey	Ankara	Gazi University	2006–2015	Single	Retrospective	English
Bashir	2017	USA	New York	Harlem Hospital Center	2010–2016	Single	Retrospective	English
Calis	2017	Turkey	Kirsehir	Ahi Evran University	2014–2016	Single	Retrospective	English
Chu	2017	USA	Dallas	Texas University	2007–2014	Single	Retrospective	English
Deng	2017	China	Yunnan	Kunming Medical University	2012–2016	Single	Retrospective	English
Farouk	2017	Egypt	Mansoura	Mansoura University	2012–2016	Single	Prospective	English
Freeman	2017	USA	Cincinnati	Cincinnati Medical Center	2004–2016	Single	Retrospective	English
Fruchter	2017	USA	New York	New York University	NS	Single	Case Report	English
Jeon	2017	S.Korea	Incheon	Gachon University	2005–2016	Single	Retrospective	English
Johnstone	2017	Australia	Brisbane	Queensland University	2010–2015	Single	Retrospective	English
Mahmodlou	2017	Iran	Urmia	Imam Khomeini University	2010–2015	Single	Retrospective	English
Prasad	2017	India	Manipal	Kasturba Medical College	2006–2014	Single	Retrospective	English
Salehi	2017	Iran	Isfahan	Isfahan University Med Sci	NS	Single	NS	English
Shin	2017	S.Korea	Chungbuk	Chungbuk National University	2008–2014	Single	Retrospective	English
Uysal	2018	Turkey	Gaziantep	Sanko University	2011–2016	Multicentric	Retrospective	English
Wong	2017	Hong Kong	Pokfulam	Hong Kong University	2009–2015	Single	Retrospective	English
Li	2019	China	Tianjin	Tianjin Central Hospital	2010–2015	Single	Retrospective	English

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 Martinez-Ramos et al. [1] and we hope that it will be supplemented to their article.

### Ethical approval

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### 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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