Chronic Idiopathic Granulomatous Mastitis in a Male Patient

Tapanutt Likhitmaskul, MD[1] ; Suphatcharee Aroonsiriwattana, MD[2]

Tapanutt Likhitmaskul, MD

Abstract

Idiopathic granulomatous mastitis is a rare inflammatory disease of the breast, typically occurs in reproductive age women with a history of pregnancy and breastfeeding. Furthermore, it is exceedingly rare in males. As the clinical signs and symptoms with radiological imaging of idiopathic granulomatous mastitis can imitate two common breast diseases, mastitis with abscess and inflammatory breast cancer, a tissue biopsy is required for clear diagnosis. Given the rarity and unknown etiology of the disease, an ideal treatment method has not been established. Conservative treatment with immunosuppressive therapy has proven good efficacy. However, in recurrent cases and cases unresponsive to conservative or medical treatment, the surgical options, such as surgical wide excision and/or mastectomy, should be considered. In this case report, we present a male patient diagnosed with left idiopathic granulomatous mastitis. We review and discuss the clinical presentation, imaging, diagnosis, and management of this patient that eventually was treated successfully with wide local excision.

Keywords: granulomatous mastitis, male, idiopathic

[1] Breast Center, Bangkok Hospital Pattaya, Chonburi, Thailand
[2] Department of Radiology, Hua Hin Hospital, Prachuap Khiri Khan, Thailand

*Address Correspondence to author:
Tapanutt Likhitmaskul, MD
Breast Center, Bangkok Hospital Pattaya,
301 Moo 6 Sukhumvit Road, Km. 143,
Banglamung, Chonburi 20150, Thailand.
email Tapanutt.Li@bph.co.th

Received: February 5, 2024
Revision received: February 8, 2024
Accepted after revision: February 15, 2024
BKK Med J 2024;20(1): 33-38.
DOI: 10.31524/bkkmedj.2024.12.002
www.bangkokmedjournal.com

Idiopathic granulomatous mastitis (IGM) is an uncommon chronic inflammatory breast disease with unknown etiology in females and is exceptionally rare in male patients.[1] IGM is often mistaken for more common breast disorders, breast abscess or infectious mastitis, and can mimic inflammatory breast cancer both clinically and radiological.[2,3] Since Kessler and Wolloch[4] first described it in female patients in 1972, there have been hundreds of female cases reported worldwide.[5] However, there are a limited number of reported cases of IGM in males, still there is not enough data on clinical management of these patients.[6] In both male and female patients, IGM usually presents as a unilateral firm breast mass with inflammation of overlying skin, abscesses and fistulae.[1,5] It usually poses a complex diagnostic and therapeutic dilemma because of the unknown etiology and rarity. The final diagnosis of IGM is confirmed by histopathological tissue examination of biopsy specimens. The characteristic histologic features of IGM includes nonnecrotizing granulomatous lesions and microabscesses confined to breast lobules without evidence of any infection or carcinoma.[7,8]

There is no definitive treatment following IGM diagnosis. Different treatment options are available including expectant management, antibiotics, steroids, other immunosuppressive drugs and surgical excision with uncertain degrees of response and recurrence. Long-term follow-up is routinely suggested in any treatments due to high rate of recurrence.[5]

In this report we present a male patient with inflammatory left breast mass finally diagnosed as IGM. We review and discuss the history, clinical presentation, imaging, diagnosis, and course of treatments for this disease including antibiotics, steroids and surgical therapy in this patient who eventually was treated successfully with wide local excision.

Case Report

A 71-year-old British male patient presented with a painful palpable left breast mass for a few weeks with associated inflammation of overlying skin and nipple. The patient denied any history of recent breast trauma. He had underlying coronary artery disease after successful stent implantation 6 years ago and previous surgical treatment for basal cell carcinoma at his right ear 2 years ago. He used to be a heavy smoker for more than ten years however he had stopped smoking after coronary stent placement. His father and brother were diagnosed with lymphoma, however, his family history was negative for both breast cancer and autoimmune disease along with no recent tuberculosis exposure. Systemic review was unremarkable as well.

On physical examination, there was an ill-defined border 5-6 cm firm mass in the central part, under the left nipple, of his left breast with tenderness and localized erythematous skin. He did not have nipple discharge. Contralateral breast and bilateral axillary lymph nodes were unremarkable. His temperature and other vital signs were normal.

Digital breast mammography with tomosynthesis showed irregular mass at retroareolar region of left breast with mild nipple retraction without microcalcification (Figure 1,2).

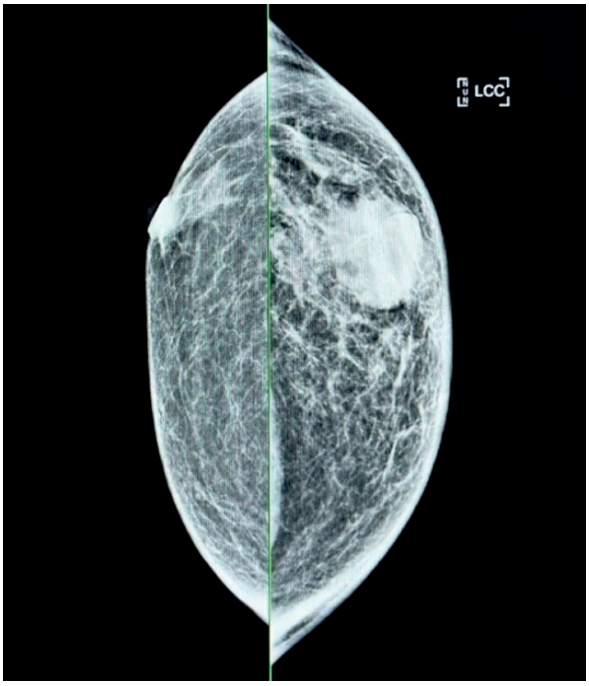

Figure 1: The craniocaudal (CC) view of mammography. Irregular left retroareolar mass with retracted nipple.

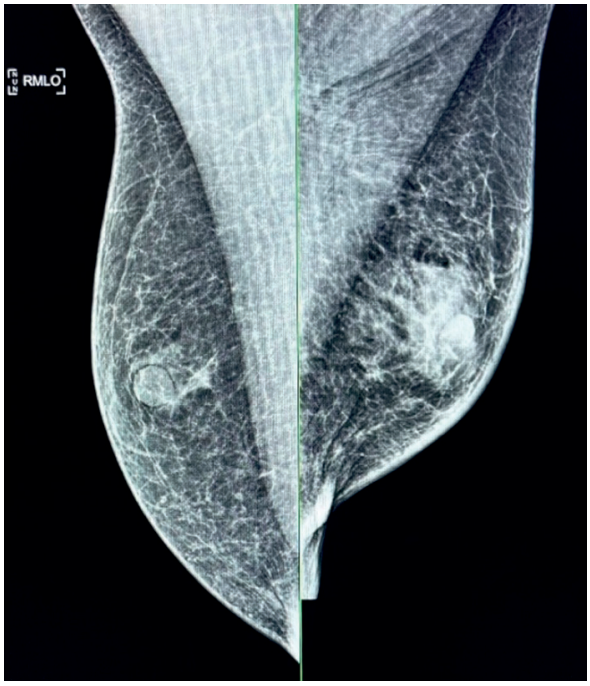

Figure 2: The mediolateral oblique (MLO) view of mammography. Irregular left retroareolar mass with retracted nipple.

Additional breast ultrasound revealed irregular hypoechoic mass with internal cystic space at subareolar region of left breast, measuring about 3.4x2.0x2.3 cm in size with increased internal vascularity (Figure 3).

Abnormal findings on mammography and ultrasound were categorized as Breast Imaging - Reporting and Data System (BI-RADS) 4. Mastitis with evolving breast abscess or inflammatory breast cancer was suspected; therefore, oral antibiotic was prescribed to him. Three days later, the patient's condition had improved and inflammatory symptoms had decreased, however, there was no change in the size of the mass. To confirm the diagnosis of the mass, after stopping aspirin for 3 days, tissue biopsy was done by core needle under ultrasound guidance method. The histopathological evaluation of tissue biopsies showed acute suppurative inflammation with granulation tissue formation and few scattered foreign body giant cells ingesting some material (Figure 4) without identified area of invasive carcinoma. These findings could be found in suppurative lesion associated with ruptured epidermal inclusion cyst, dermal abscess or periphery of inflammatory breast cancer. Further excisional biopsy for definite diagnosis was performed one week later. Tissue was sent for histology, gram stain, acid-fast bacilli (AFB) stain, Grocott's methenamine silver (GMS) stain, aerobic and anaerobic cultures. All stains and cultures were negative. No microorganisms were identified. There was no evidence of malignancy. Postoperative histopathological examination showed chronic abscess lined by severely inflamed granulation tissue containing numerous foamy macrophages and epithelioid histiocytes, Langhans giant cells with small lymphocytes and aggregates of neutrophils in small suppurative foci, which were compatible with IGM. The lesion was present at the closest margin of the specimen.

ISSN 2287-0237 (online)/ 2228-9674 (print)

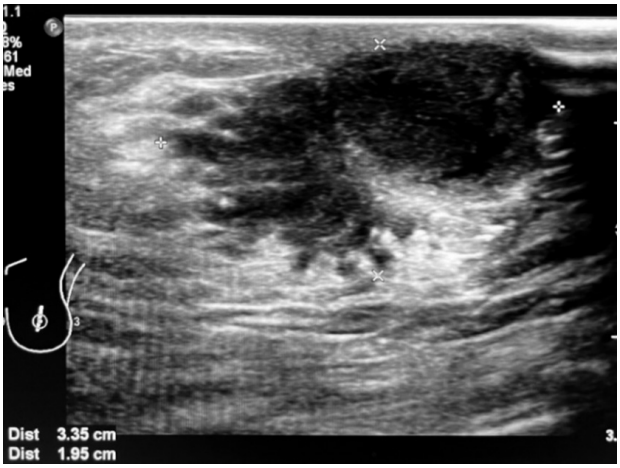

Figure 3: Ultrasonography of the patient's left breast mass. Irregular retroareolar lesion.

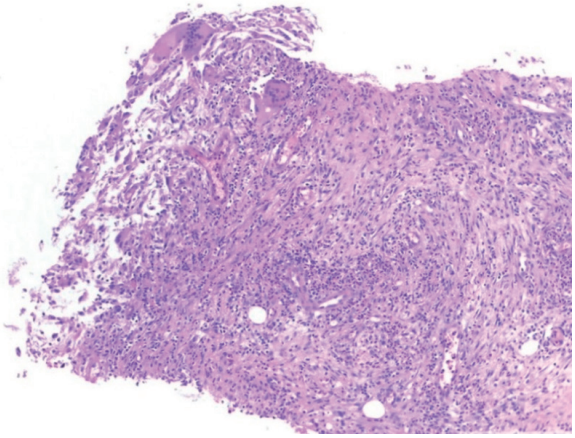

Figure 4: Histological findings of the core needle biopsy specimen from patient's left breast mass. Granulation tissue with foreign body giant cells.

At one month postoperative follow up, the surgical wound was completely healed without any signs of inflammation. However, a few weeks later he developed a new inflamed mass at lower part of left breast, near old surgical scar at left nipple (Figure 5). Although he was given oral antibiotic, there was no improvement. As the pain increased at his new left breast mass with an area of induration and erythema, he subsequently underwent surgical incision and drainage. About 5 ml of purulent discharge drained from the lesion. Both discharge and inflamed tissue were sent for histology and bacterial cultures. The histological diagnosis of granulomatous mastitis was confirmed again; nevertheless, at this time numerous growth of coagulase-negative staphylococci was found in tissue cultures without resistance to any antibiotics. He received postoperative oral antibiotic (clindamycin 900 mg daily), oral steroid (prednisolone 30 mg daily) and wound dressing change (once daily) for 2 weeks. As a result, the surgical wound was filled with good granulation tissue, then spontaneously closed and all inflammatory symptoms disappeared. The patient maintained an initial dose of prednisolone for one month, then reduced and stopped in three months.

Three weeks later he developed a new painful firm mass under the surgical scar. Initial treatment with oral steroid (prednisolone 20 mg daily) had a good response. The inflammation had decreased; moreover, the mass became about half the size. After six weeks of treatment with oral steroid, prednisolone was tapered off completely and the palpable mass had disappeared (Figure 6).

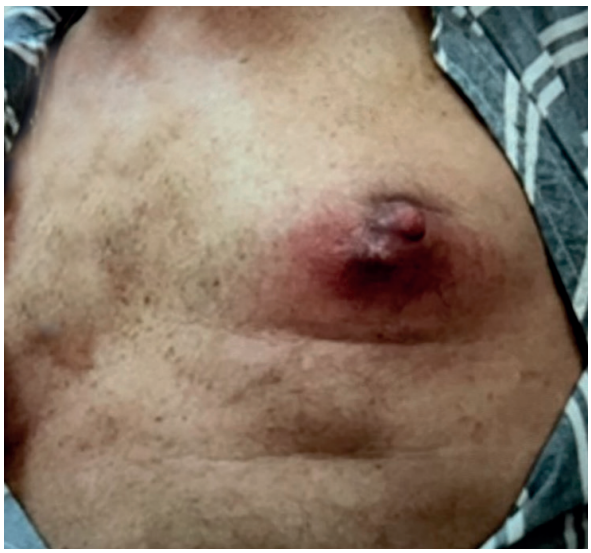

Figure 5: The patient's left breast. Recurrent inflammatory mass, almost two months after first surgical excision

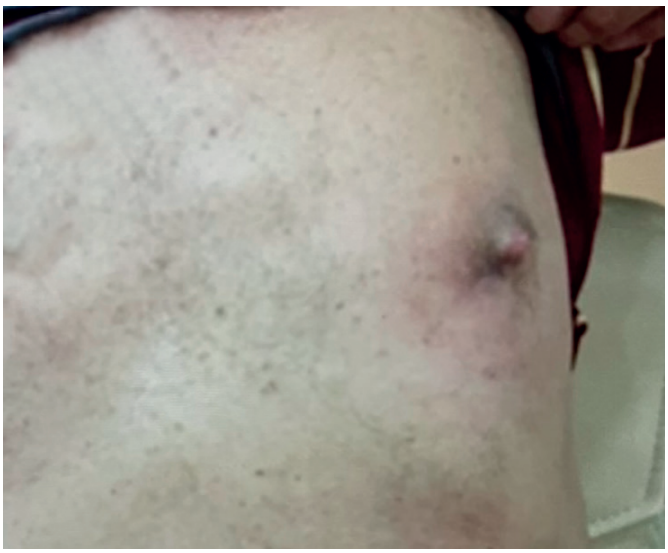

Figure 6: The patient's left breast. Complete resolution after steroid therapy.

Nonetheless, three weeks later the inflamed palpable mass came back again in the same location in the left breast. The patient started on 20 mg of prednisolone daily and some improvement of inflammatory symptoms was seen. However, the size of left breast mass was slightly smaller after four weeks of steroid treatment. So, he eventually underwent wide local excision of recurring left breast mass including left nipple. Two weeks later his symptoms had been totally resolved. The surgical wound healed as well.

At the last follow-up visit, nine years after surgery, the patient was asymptomatic and no local recurrence was noted.

Discussion

IGM is defined as granulomatous mastitis without any evidence of infection and no specific causes. It is a rare, noncancerous, chronic inflammatory breast disease that is seen more commonly in young women of fertile age and the usual age range is 22 to 42 years.[9,10] Women with a history of pregnancy and breast lactation within the preceding five years are the most frequently affected.[11] It is exceptionally rare during pregnancy and in men.[5,6,12]

IGM was first described in 2005 and only 17 reported cases were reported between the ages of 17-60, the median age of the patients at the time of presentation was 46 years.[1] Clinical presentation of IGM is usually unilateral, variable size, firm and ill-defined breast mass. It is often associated with local tenderness and skin inflammation like our patient with inflammatory retroareolar mass in this report. In more chronic presentations, skin ulcerations, nipple inversion, fistulae and abscesses may eventually develop.[5,13] IGM usually is a diagnosis of exclusion because it's commonly mistaken as common acute breast infection, infective mastitis or breast abscess and can clinically and radiologically imitate the symptoms of breast cancer, especially in elderly patients, particularly if the regional lymph nodes are palpable.[2,3,5] If symptoms of breast infection are not quickly improved or if there are repeated relapses after regular treatment, this disease should be included in the differential diagnosis. Thus a prompt tissue biopsy should be performed for definite diagnosis.[14,15]

Veyssiere[16] was the first person to introduce the term granulomatous mastitis in 1967. The certain etiology of this disease is currently undetermined. Kessler and Wolloch[4] who first described this disease in 1972 supported an autoimmune process corresponding to the microanatomy. An association has been suggested with recent pregnancy and breastfeeding has led to the foreign body reaction to extravasated secretions from the breast lobules.[17] Associations with underlying lymphatic cancer, abnormal hormonal conditions such as gynecomastia and estrogen therapy have also been reported. The autoimmune response toward the extranodal lesion of lymphoma in the breast was reported in a male patient with history of previous treatment for follicular lymphoma that might be one of the probable causes of his IGM.[1] The absence of breast lobules in males may be one possible reason for rarity of male IGM.[18] Gynecomastia and estrogen stimulation can cause the development of mammary lobules in male breasts that might be associated with IGM in males.[19] However, there are limited cases of male IGM, and there is still not enough data on these associations in males.

IGM and breast cancer frequently have similar appearances in breast imaging. Mammographic pictures of IGM can be variable from normal findings in dense breast patients to masses with benign or malignant appearance and, most commonly focal asymmetrically increased density that look like mammographic features of breast cancer. Sonographic findings are also varied and relate to histologic patterns. The most common finding on breast ultrasonography is an irregular or ill-defined hypoechoic mass as demonstrated in our patient. Although magnetic resonance imaging of the breast is better at detecting cancer than mammography and ultrasound, it cannot differentiate between a granulomatous process and other inflammatory disorders.[5,8]

Tissue histopathological examination remains the gold standard for the diagnosis of IGM. This is because the clinical signs and radiologic findings of IGM are indistinguishable from breast cancer especially inflammatory type. The characteristic histologic features includes non-necrotizing granulomatous lesions and micro-abscesses confined to breast lobules without evidence of specific etiologic microorganism or carcinoma favor diagnosis of this disease.[5,7,8,10] The differential diagnoses include breast cancer, other granulomatous breast infections such as tuberculous and mycotic infections especially when the fistulae to the skin occur, autoimmune diseases (sarcoidosis, Wegener's granulomatosis), foreign body reaction and granulomatous reaction in cancer must be ruled out before diagnosing IGM.[20,21] So a tissue sample is needed to avoid misdiagnosing as breast cancer. The definitive diagnosis can be only made by tissue histopathological confirmation, so core needle tissue biopsy is preferred over fine needle aspiration cytology because a core biopsy is more accurate as it shows the tissue architecture.[5]

Given the rarity and unknown etiology of the disease, an ideal treatment method has not been established. Treatment options depend on the severity of the disease and may include close observation, antibiotics, steroids, other immunosuppressive drugs or surgical excision.[5,22] In females, IGM is generally treated with conservative and medical treatment first, especially in patients with mild symptoms or uncomplicated disease. However, for male IGM the treatment options were limited to surgical excision and mastectomy in the literature.[6] Due to limited cases, the effects of each treatment options in the treatment of male IGM remain controversial.

The different rates of recurrence reported for each treatment method have been shown only in female patients.[5,23] Some studies reported that asymptomatic IGM patients who underwent observation without any therapeutic intervention, spontaneous resolution seen in 7-14.5 months.[10,23,24] In most patients with IGM, the initial clinical presentation as infective

ISSN 2287-0237 (online)/ 2228-9674 (print)

mastitis leads to antibiotic usage. Antibiotics may be useful in the treatment of other granulomatous breast infection. Although an association of IGM with local infection with Corynebacterium species has recently been proposed, there is no role of antibiotics in the management of true IGM cases.[25] This is because the condition of patients usually worsens or remains unresponsive to antibiotics and can lead to different antibiotic combinations and antibiotic overuse.[26,27] For female patients with more severe symptoms, steroids are administered. Although a low-dose is preferred, steroids should be started at a dose of 1 mg/kg per day and higher doses continually administered until the lesions completely resolve; then taper the steroids slowly according to clinical response.[15] Responses often present within weeks of treatment as it appears in the treatment course of our male patient. Treatment with steroids requires several months (usually up to six months) to achieve a complete response.[9,15] Complete resolution of the disease is seen in 80% of those who received steroids. However, about half of cases relapse after decreasing or stopping the dose of steroids as seen in this male patient.[15,28]

In recurrent cases and cases unresponsive to conservative or medical treatment, as shown in our patient, surgical therapy as surgical excision and/or mastectomy finally should be considered.[6,13,29] Wide excision with negative margin is recommended because of a higher rate of recurrence after only a limited excision. 5-50% recurrence rates have been reported in surgical treatment group and may be higher than steroid treatment in some studies.[29] In addition to high recurrence, postoperative wound complications and fistula formation have been reported in most patients.[10] As seen in our patient, more conservative surgery can be performed after preoperative steroid therapy that delivered significant regression of the inflammatory mass.[30]

Conclusion

Idiopathic granulomatous mastitis is a rare, benign inflammatory breast disease that is common in young women of fertile age, furthermore, extremely rare in males. It is commonly mistaken for inflammatory breast cancer in elderly male patients, so the definitive diagnosis is made by using histopathological examination of tissue biopsies. Only a high index of suspicion will prevent the morbidity of misdiagnosis or delayed diagnosis and inappropriate therapy. Due to the unclear cause and rarity of the disease, there is no standard treatment at present. Treatment depends on the severity of symptoms and available treatment options include close follow-up, medical and surgical therapy. In the future if we come to know the clear etiology of this disease, a new potential treatment with low recurrence may yet be discovered.

Conflict of interests: The authors have declared no conflicts of interests.

Ethical approval: A written informed consent was obtained from the patient for publication of this case report.

References

1. Kawashima K, Yamamoto S, Narui K, et al. Granulomatous mastitis in a male breast: a case report and review of literature. *Clin Case Rep* 2023;11:e7048. doi: 10.1002/ccr3.7048
2. Tuli R, O'Hara BJ, Hines J, et al. Idiopathic granulomatous mastitis masquerading as carcinoma of the breast: a case report and review of the literature. *Int Semin Surg Oncol* 2007;4:21. doi: 10.1186/1477-7800-4-21.
3. Erhan Y, Veral A, Kara E, et al. A clinicopathologic study of a rare clinical entity mimicking breast carcinoma: idiopathic granulomatous mastitis. *Breast* 2000;9(1):52-6. doi: 10.1054/brst.1999.0072.
4. Kessler E, Wolloch Y. Granulomatous mastitis: a lesion clinically simulating carcinoma. *Am J Clin Pathol* 1972;58(6):642-6. doi: 10.1093/ajcp/58.6.642.
5. Likhitmaskul T, Sukpanich R, Visutdiphat S, et al. Bilateral idiopathic granulomatous mastitis: a case report. *Bangkok Med J* 2014; 8:65-70
6. Al Manasra AR, Al-Hurani MF. Granulomatous mastitis: a rare cause of male breast lump. *Case Rep Oncol* 2016;9(2):516-9. doi: 10.1159/000448990.
7. Baslaim MM, Khayat HA, Al-Amoudi SA. Idiopathic granulomatous mastitis: A heterogeneous disease with variable clinical presentation. *World J Surg* 2007;31(8):1677-81. doi: 10.1007/s00268-007-9116-1.
8. Akcan A, Akyildiz H, Deneme MA, et al. Granulomatous lobular mastitis: A complex diagnostic and therapeutic problem. *World J Surg* 2006;30(8):1403-9. doi: 10.1007/s00268-005-0476-0.
9. Rubin G, Meerkotter DA. Idiopathic granulomatous mastitis. *SA J Radiol* 2011;15:4-5.
10. Lai EC, Chan WC, Ma TK, et al. The role of conservative treatment in idiopathic granulomatous mastitis. *Breast J* 2005;11(6):454-6. doi: 10.1111/j.1075-122X.2005.00127.x.
11. Boufettal H, Hermas S, Noun M, et al. Mastite granulomateuse idiopathique bilatérale. *Imagerie de la Femme* 2009; 19:262-4. doi:10.1016/j.femme.2009.09.004.
12. Reddy KM, Meyer CE, Nakdjevani A, et al. Idiopathic granulomatous mastitis in the male breast. *Breast J* 2005;11(1):73. doi: 10.1111/j.1075-122X.2005.21404.x.
13. Bani-Hani KE, Yaghan RJ, Matalka II, et al. Idiopathic granulomatous mastitis: time to avoid unnecessary mastectomies. *Breast J* 2004;10(4):318-22. doi: 10.1111/j.1075-122X.2004.21336.x.
14. Garcia-Rodiguez JA, Pattullo A. Idiopathic granulomatous mastitis: a mimicking disease in a pregnant woman: a case report. *BMC Res Notes* 2013;6:95. doi: 10.1186/1756-0500-6-95.
15. Olsen ML, Dilaveri CA. Idiopathic granulomatous mastitis: a case report of breast abscess. *BMJ Case Rep* 2011:2011:bcr0520114271. doi: 10.1136/bcr.05.2011.4271.
16. Veyssiere C, Vives M, Smadia A. Difficultés diagnostiques de la tuberculose mammaire. Le problème de la mastite granulomateuse. *Lille Chir* 1967;22:104-9.
17. Raj N, Macmillan RD, Ellis IO, et al. Rheumatologists and breasts: immunosuppressive therapy for granulomatous mastitis. *Rheumatology* 2004;43:1055-6. doi:10.1093/rheumatology/keh246

18. Sam KQ, Severs FJ, Ebuoma LO, et al. Granulomatous mastitis in a transgender patient. *J Radiol Case Rep* 2017;11(2):16-22. doi: 10.3941/jrcr.v11i2.2934.
19. Yaghan RJ, Ayoub NM, Shenawi HM, et al. Idiopathic granulomatous mastitis in the male population: a clinical analysis of 13 reported cases. *Breast J* 2020;26(7):1481-2. doi: 10.1111/tbj.13778.
20. Erozgen F, Ersoy YE, Akaydin M, et al. Corticosteroid treatment and timing of surgery in idiopathic granulomatous mastitis confusing with breast carcinoma. *Breast Cancer Res Treat* 2010;123(2):447-52. doi: 10.1007/s10549-010-1041-6.
21. Al-Khaffaf B, Knox F, Bundred NJ. Idiopathic granulomatous mastitis: a 25-year experience. *J Am Coll Surg* 2008;206(2):269-73. doi: 10.1016/j.jamcollsurg.2007.07.041.
22. Patel RA, Rodriguez M. Idiopathic granulomatous mastitis: case report and review of literature. *J Gen Intern Med* 2010;25(3):270-3. doi: 10.1007/s11606-009-1207-2.
23. Akbulut S, Yilmaz D, Bakir S. Methotrexate in the management of idiopathic granulomatous mastitis: review of 108 published cases and report of four cases. *Breast J* 2011;17(6):661-8. doi: 10.1111/j.1524-4741.2011.01162.x.
24. Bouton ME, Jayaram L, O'Neill PJ, Hsu CH, Komenaka IK. Management of idiopathic granulomatous mastitis with observation. *Am J Surg* 2015;210(2):258-62. doi: 10.1016/j.amjsurg.2014.08.044.
25. 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):109-19.
26. Garcia-Rodiguez JA, Pattullo A. Idiopathic granulomatous mastitis: a mimicking disease in a pregnant woman: a case report. *BMC Res Notes* 2013;6:95. doi: 10.1186/1756-0500-6-95.
27. Wilson JP, Massoll N, Marshall J, et al. Idiopathic granulomatous mastitis: In search of a therapeutic paradigm. *Am Surg* 2007;73(8):798-802.
28. Kim J, Tymms KE, Buckingham JM. Methotrexate in the management of granulomatous mastitis. *ANZ J Surg* 2003;73(4):247-9. doi: 10.1046/j.1445-1433.2002.02564.x.
29. Asoglu O, Ozmen V, Karanlik H, et al. Feasibility of surgical management in patients with granulomatous mastitis. *Breast J* 2005;11(2):108-14. doi: 10.1111/j.1075-122X.2005.21576.x.
30. Gurleyik G, Aktekin A, Aker F, et al. Medical and surgical treatment of idiopathic granulomatous lobular mastitis: A benign inflammatory disease mimicking invasive carcinoma. *J Breast Cancer* 2012;15(1):119-23. doi: 10.4048/jbc.2012.15.1.119.