

Granulomatous Mastitis in a Male Breast: A Case Report and Review of Literature

Kei KAWASHIMA¹, Shinya Yamamoto¹, Kazutaka Narui¹, Yoshie Fujiwara¹, Shoko Adachi¹, Mahato Sasamoto², Masanori Oshi², Akimitsu Yamada², Eita Kumagai¹, Masako Otani¹, and Itaru Endo²

¹Yokohama City University Medical Center

²Yokohama City University School of Medicine Graduate School of Medicine

December 23, 2022

Abstract

Granulomatous mastitis (GM) is a rare disease, particularly amongst men. Herein, we present a case of GM diagnosed in a 63-year-old male patient who showed reduction in the tumor size during 3 months of observation.

Introduction:

Granulomatous mastitis (GM), a chronic inflammatory disorder of the breast with unknown etiology, was first described by Kessler et al. in 1972.¹ GM usually affects parous females and is rarely diagnosed in male breasts. We describe a rare case of GM diagnosed in a male patient during the therapeutic course of follicular lymphoma.

Case history:

A 63-year-old male patient with history of chemotherapy for follicular lymphoma presented with a left breast mass detected on a periodic computed tomography (CT) scan. He was diagnosed with follicular lymphoma five years ago and treated with six cycles of a combination of rituximab and bendamustine, followed by treatment with rituximab for two years as maintenance therapy. Recurrence of lymphoma was detected at the cervical and axillary lymph nodes after two years of an uneventful post-therapeutic course. Lenalidomide and rituximab were administered as treatment for recurrence. A CT scan performed at the end of chemotherapy showed disappearance of all target lesions, but appearance of a new mass in his left breast. The patient was referred to our department for further evaluation. On physical examination, a mass measuring approximately 1 cm in diameter was palpable in the subareolar area of his left breast, while no swollen lymph nodes were detected in the ipsilateral axillary region. Laboratory tests including tumor marker analyses were all within normal levels (white blood cell, 2840 / μ l; C-reactive protein, 0.032 mg/dl; carcinoembryonic antigen, 3.8 ng/ml; cancer antigen 15-3, 17 U/ml; sIL-2R, 385 U/ml). CT scan confirmed the presence of a lesion measuring 13.9 \times 12.9 \times 6.1 mm in the subareolar portion of the left breast (Fig. 1). Breast ultrasonography revealed an ill-defined hypoechoic irregular mass with peripheral vascularity in the subareolar portion (Fig. 2). Ultrasound-guided core needle biopsy was performed, and histological examination showed a granulomatous structure comprising mononuclear inflammatory cell infiltration accompanied by foam cells without any evidence of caseous necrosis (Fig. 3). No specific pathogen or foreign bodies were detected. These findings confirmed the diagnosis of GM.

Since there was no suspicion of malignancy and the patient was asymptomatic, he was admitted for observation. Ultrasonography performed after three months revealed that the size of the tumor was 7.9 \times 5.5 \times 5.2

mm (Fig. 4), which was smaller compared to the initial diagnosis. The patient is currently under observation.

Discussion:

GM is diagnosed histologically by confirming the presence of non-necrotizing granuloma formation, without any evidence of microorganisms such as *Mycobacterium* or fungi.² Its etiology is uncertain, and imaging findings are not specific. GM occurs in female breasts and accounts for 1.8% of benign breast disorders proven by biopsy.³ It mainly affects females of reproductive age. The mean age at presentation is 33–38 years.^{1,4,5} Hormonal disruption and autoimmune responses have been suggested as the possible etiological factors.⁶

GM is rarely observed in males. To the best of our knowledge, there are only 16 reported cases of GM in male patients.^{1,2,4-15} The reported cases have been summarized in Table 1. We analyzed 12 cases of patients whose characteristics and clinical courses were described.^{2,6-15} The median age of the patients at the time of presentation was 46 years (range: 17–60 years). Eight patients (73%) had GM in the right breast, while two (17%) presented with bilateral involvement. The median size of the masses was 20 mm (range: 5–72 mm). All patients had a breast mass with or without pain, while two (17%) had ulcerations. Nipple discharge was observed in only one patient (8%). A definitive diagnosis of GM was obtained by core needle biopsy in eight patients (67%), by fine needle aspiration in one patient (8%), and by excisional biopsy in three patients (25%). The baseline characteristics of female GM patients have been described as follows: mean age of 36 years, lesion measuring approximately 50 mm, and nipple discharge observed in 30% patients.¹⁶

One possible reason for rarity in males is the absence of mammary lobules, which are usually affected during this disease. It has been reported that estrogen stimulation causes the development of acini and lobules in male breasts² and might be responsible for the development of GM. Of the 12 cases, four patients had gynecomastia^{10,14,15,16} and one was a transgender (male to female transition, receiving estrogen therapy for six years).² This supports the fact that GM in males might be associated with abnormal hormonal conditions such as gynecomastia and estrogen therapy. This may also be responsible for the higher age of affected male patients compared to female patients, since the ratio of androgen to estrogen in males lowers with age.¹⁸ However, hormonal involvement was not detected in the remaining seven cases and our case. Therefore, it can be concluded that the etiology of GM in males remains unclear.

The optimal treatment strategy for GM remains controversial. Treatment approaches include observation, oral corticosteroid administration, and surgical excision. Incision and drainage may be a treatment option for bacterial infection cases. Oral corticosteroids have been used as the first-line treatment in some studies and reported to be effective in decreasing the size of GM.⁴ However, observation without any therapeutic intervention could be considered for asymptomatic cases. Some studies have reported that GM patients who underwent observation without any medication achieved resolution in 7–14.5 months.^{19,20}

Of the 11 reported male cases in which treatment was mentioned, four patients underwent surgical excision. Core needle biopsy was not attempted in these four cases, and the lesion was surgically excised in toto as an excisional biopsy. Of the remaining seven patients, four who received oral corticosteroid therapy were symptomatic, whereas two who underwent observation did not have any symptoms other than the breast mass. In one patient who was treated with bromocriptine for pituitary tumor, the breast tumor disappeared after the treatment. None of the patients experienced recurrence during their clinical course, but the follow-up periods were not sufficient to determine the long-term outcomes. In our case, since the patient was asymptomatic and diagnosis of GM was confirmed histologically, we decided to observe the lesion.

In contrast to other reported cases, the tumor in our case appeared during the clinical course of follicular lymphoma treatment. Although not histologically proven, one possible explanation for the occurrence of GM in our case may be the autoimmune response toward the extranodal lesion of lymphoma in the breast.

To summarize, we presented a rare case of GM in a male patient. Observation may be a viable option in the asymptomatic cases. However, further studies including a larger number of patients and longer observation period are needed for reliable long-term outcomes.

Author contributions:

KK: acquired the data, analyzed and interpreted the data, wrote the manuscript, approved the final manuscript

KK, SY, KN, YF, SA, SM, MO, AY, EK, MO, IE: analyzed and interpreted the data, approved the final manuscript.

Acknowledgments: None

References:

1. Prasad, S., Jaiprakash, P., Dave, A., Pai, D. 2017. Idiopathic granulomatous mastitis: an institutional experience. *Turkish Journal of Surgery* 33:100-103.
2. Sam, K. Q., Severs, F. J., Ebuoma, L. O., Chandandeep, N. S., Sedgwick, E. L. 2017. Granulomatous mastitis in a transgender patient. *Journal of Radiology Case Reports* 11:16-22.
3. Pluguez-Turull, C. W., Nanyes, J. E., Quintero, C. J., Alizai, H., Mais, D. D., Kist, K. A., Dornbluth, N. C. 2018. Idiopathic granulomatous mastitis: manifestations at multimodality imaging and pitfalls. *Radiographics* 38:330-356.
4. Barreto, D. S., Sedgwick, E. L., Nagi, C. S., Benveniste, A. P. 2018. Granulomatous mastitis: etiology, imaging, pathology, treatment, and clinical findings. *Breast Cancer Research and Treatment* 171:527-534.
5. Korkut, E., Akcay, M. N., Karadeniz, E., Subasi, I. D., Gursan, N. 2015. Granulomatous mastitis: A ten-year experience at a University Hospital. *Eurasian Journal of Medicine* 47:165-173.
6. Moris, D., Damaskos, C., Davakis, S., Vailas, M., Garmpis, N., Spartalis, E., Kontos, M., Kontzoglou, K. 2017. Is idiopathic granulomatous mastitis a surgical disease? The jury is still out. *Annals of Translational Medicine* 5:309.
7. Heer, R., Shrimankar, J., Griffith, C. D. M. 2003. Granulomatous mastitis can mimic breast cancer on clinical, radiological or cytological examination: a cautionary tale. *Breast* 12:283-286.
8. Reddy, K. M., Meyer, C. E. R., Nakdjevani, A., Shrotria, S. 2005. Idiopathic granulomatous mastitis in the male breast. *Breast Journal* 11:73-73.
9. Al Manasra, A. R., Al-Hurani, M. F. 2016. Granulomatous mastitis: A rare cause of male breast lump. *Case Reports in Oncology* 9:516-519.
10. Joo, L., Hyun Yeo, S., Young Kwon, S. 2018. Idiopathic granulomatous lobular mastitis in a male breast: A case report. *Iranian Journal of Radiology*, In Press.
11. Farrokh, D., Alamdaran, A., Haddad, A. S., Kheirollahi, M., Abbasi, B. 2019. Male granulomatous mastitis, a rarely encountered disorder. *Breast Journal* 25:517-518.
12. Dinç Elibol, F., Elibol, C., Tekin, L., Dönmez, C. 2020. A very rare cause of male breast lump: idiopathic granulomatous mastitis. *Breast Journal* 26:778-779.
13. Sahin, C., and B. Agridag Ucpinar. 2020. Idiopathic granulomatous mastitis in a male breast following intravesical bacille calmette-guerin treatment. *Journal of Infection and Public Health* 13:830-832.
14. Yin, Y., Cheng, Y., Wang, B., Chen, L., Chen, H. 2020. Male granulomatous mastitis with hidradenitis suppurativa, a rarely encountered disorder. *Breast Journal* 26:2058-2059.
15. Madzima, T. R., Yuen, V., Lu, F.-I., Curpen, B., Skarpathiotakis, M. 2022. Bilateral male idiopathic granulomatous mastitis of the breast: a case report. *Annals of Breast Surgery* 6:9-9.
16. Yin L, Agyekum EA, Zhang Q, Wu T, Qian X. 2022. Gynecomastia with rare granulomatous lobular mastitis: a case report and literature review. *Journal of International Medical Research* 50:3000605221075815.

17. Sarica O, Kahraman AN, Ozturk E, Teke M. 2018. Efficiency of Imaging Modalities in Male Breast Disease: Can Ultrasound Give Additional Information for Assessment of Gynecomastia Evolution? *Eur J Breast Health*. 14:29-34.
18. Yaghan, R. J., Ayoub, N. M., Shenawi, H. M., Yaghan, L. R. 2020. Idiopathic granulomatous mastitis in the male population: A clinical analysis of 13 reported cases. *Breast Journal* 26:1481-1482.
19. Akbulut, S., Arikanoglu, Z., Senol, A., Sogutcu, N., Basbug, M., Yeniars, E., Yagmur, Y. 2011. Is methotrexate an acceptable treatment in the management of idiopathic granulomatous mastitis? *Archives of Gynecology and Obstetrics*, 284:1189-1195.
20. Bouton, M. E., Jayaram, L., O'Neill, P. J., Hsu, C. H., Komenaka, I. K. 2015. Management of idiopathic granulomatous mastitis with observation. *American Journal of Surgery* 210:258-262.

Figure legends:

Figure 1. Plain computed tomography scan (a: coronal plane, b: axial plane) at the first visit.

A tumor measuring $13.9 \times 12.9 \times 6.1$ mm in the subareolar portion of the left breast (Arrowhead).

Figure 2. Ultrasonogram at the first visit.

An ill-defined hypoechoic irregular mass with peripheral vascularity (Arrow).

Figure 3. Histopathological findings of the core needle biopsy specimen (a: HE $\times 40$ magnification, b: HE $\times 200$ magnification)

Infiltration of mononuclear inflammatory cells accompanied by foam cells (Arrowhead). HE, hematoxylin and eosin stain

Figure 4. Ultrasonogram after three months.

Tumor size reduced to $7.9 \times 5.5 \times 5.2$ mm (Arrow).

Table legend

Table 1. Reported cases of granulomatous mastitis in male patients.

Hosted file

Figure_1.docx available at <https://authorea.com/users/569262/articles/614938-granulomatous-mastitis-in-a-male-breast-a-case-report-and-review-of-literature>

Hosted file

Figure_2.docx available at <https://authorea.com/users/569262/articles/614938-granulomatous-mastitis-in-a-male-breast-a-case-report-and-review-of-literature>

Hosted file

Figure_3.docx available at <https://authorea.com/users/569262/articles/614938-granulomatous-mastitis-in-a-male-breast-a-case-report-and-review-of-literature>

Hosted file

Figure_4.docx available at <https://authorea.com/users/569262/articles/614938-granulomatous-mastitis-in-a-male-breast-a-case-report-and-review-of-literature>

Hosted file

Table1.xlsx available at <https://authorea.com/users/569262/articles/614938-granulomatous-mastitis-in-a-male-breast-a-case-report-and-review-of-literature>