

## Granulomatous Lobular Mastitis Manifested Rarely Two Years After Cessation Of Breastfeeding

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### Abstract

Granulomatous lobular mastitis (GLM) is an unusual chronic inflammatory condition of the breast that affects women of childbearing age with a history of breastfeeding. The aetiology of this benign breast condition is still unknown. Hence, it is termed idiopathic granulomatous mastitis. GLM usually emerges with various clinical symptoms, including breast mass, abscess, mastalgia, skin ulceration, nipple retraction, and sinus/fistula formation. In most cases, patients usually present with an ill-defined palpable mass and radiographic features mimicking malignancy. Pathological assessment is crucial to confirm the diagnosis and to rule out differential diagnosis. GLM is diagnosed by pathologic findings of non-caseating granulomas, particularly in the lobules of the breast. Various treatment modalities have come into practice, including wide surgical excision, medical treatment with antibiotics, and corticosteroids. In most cases, the mass forming GLM appears immediately after cessation of breastfeeding. However, we present a case of a patient diagnosed to have GLM after presented to the hospital two years after cessation breastfeeding with rapidly enlarging mass over one month before the consultation.

**Keywords:** Granulomatous lobular mastitis, benign breast disease, breastfeeding.

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### Introduction

Granulomatous lobular mastitis (GLM) is an uncommon, chronic inflammatory condition of the breast initially reported in 1972 by Kessler[1]. Clinical presentation of GLM includes breast mass, breast abscess, skin ulceration, nipple retraction and sinus/fistula formation. The aetiology of GLM is still yet to be elucidated, but several key factors are believed to play an important role in the disease occurrence, including hormonal effects, microbiologic agents, and immunologic disorders [2]. This benign condition commonly affects women in childbearing, while the occurrence of the disease has been reported involving patients in the age range of 11-83 years old. Many pieces of literature have reported a strong association between GLM and breastfeeding [3,4]. Breast lobules tend to change to a secretory phenotype, and the ducts get dilated after birth due to increased breast secretions and subsequent inflammation. During lactation, the lobules remain dilated as the lobules secreted protein-rich fluid under prolactin influence. Continuous long-term breastfeeding leads to long term

distention of the acini and ducts, which eventually ruptures and initiate granulomatous inflammatory response [5]. Most literature reported that the mass forming GLM developed immediately after cessation of breastfeeding and usually occurred among women who have breastfed for a long time[4]. We are presenting a case of rapidly grown mass formed two years after cessation of breastfeeding in a young woman who breastfed her child only for a 1-month duration.

### Case Report

A 26-year-old female presented to our surgical outpatient clinic with a history of a left breast lump for one month before the presentation and described the mass as painless, sudden onset, and rapidly increasing in size. She was not complaining of skin ulceration, nipple retraction, nipple discharge, or bleeding during the presentation. She had no constitutional symptoms such as loss of weight, appetite, fever, or night sweats. She denied a previous history of trauma. She has no known to have any medical illness, past surgical history, and any family history of malignancy. She was never on any form of contraceptives, hormonal supplements, or traditional medication. Allergic history was unremarkable. She achieved her menarche at the age of 13 years old, and she has one child. She breastfed her child two years ago for one month's duration and did not smoke cigarettes or consume alcohol.

On examination, a 5cm x 2cm large round mass was identified on the left inner quadrant of the breast. It had benign features such as a smooth surface and regular margin. The mass was non-tender, mobile upon palpation, without any skin changes, nipple retraction or discharges. There were no axillary or cervical lymph nodes detected. The patient's serum prolactin level was normal. An ultrasound was performed, and it was reported as benign features suggestive of fibroadenosis of the left breast (**Figure 1**). The patient refused a Fine Needle Aspiration Cytology (FNAC) or core biopsy and requested an excisional surgery. Wide local excision of the breast lump was performed under general anaesthesia. The entire mass surgically excised with a margin of 0.5 cm around it and was sent for a histopathological examination. Histopathology report denoted that the breast lobules were surrounded by abundant fibro fatty stroma, and most of the lobules were infiltrated by non-caseating granulomas composed of epithelioid cells, histiocytes and Langhan types of cells admixed with lymphocytes which fit the diagnosis of granulomatous lobular mastitis (**Figure 2**). She was followed up in the clinic for up to 6 months, and the surgical wound healed well without any sign of recurrent or any other complications.



**Figure 1: The ultrasound image of the patient's left breast showing a lump with benign features measuring 49.5mm x 23.5 mm**

## HISTOPATHOLOGY REPORT

### MACROSCOPIC DESCRIPTION

An irregular grey, brown, firm tissue mass tagged with short suture superiorly and long suture laterally weighing 33gms. It measures mediolaterally 50mm, superior-inferiorly 40mm and anterior-posteriorly 30mm. There is an irregular grey, soft brown tissue (? detached from the main mass) measuring 35x20x10mm in size. Serial sections reveal irregular grey white and grey brown areas. No obvious tumour could be made out grossly.

Representative sections were submitted into five blocks.

Block keys:-

- A --- Two bits from lateral resected margin painted
- B --- Two bits from medial resected margin painted
- C --- Grey white area with superior resected margin painted
- D --- Grey white area with inferior resected margin painted
- E --- Grey white area with anterior resected margin painted green and

posterior resected margin painted black
<b>MICROSCOPIC DESCRIPTION</b>
Sections show breast lobules surrounded by abundant fibrofatty stroma.
Most of the lobules are infiltrated by non-caseating granulomas composed of epithelioid cells, histiocytes and Langhan type of giant cells admixed with lymphocytes. Tiny areas of suppurative necrosis infiltrated by neutrophils are also present. The surrounding stroma shows dense fibrosis and hyalinisation.No features of malignancy are present.
<b>DIAGNOSIS</b>
Left breast lump --- <b>Lobular granulomatous mastitis.</b>

**Figure 2: Histopathology report of the left breast mass**

### Discussion

Granulomatous lobular mastitis is a rare benign breast condition with an incident estimated at 2.4 per 100000 women and 0.37% of cases in the US [6].The rarity of the disease has caused a lack of data on GLM in Malaysia. There are not many cases reported in Malaysia, especially on very rare casesof GLM breast mass presentedmany years after cessation of a short duration of breastfeeding. Our patient presented two years after stopping her breastfeeding,which is rare as the breast mass in GLM usually appears immediately after cessation of breastfeeding [4]. In the context of clinical manifestation,our patient hadarapidly growing breast masstwo years aftercessation of breastfeeding, mimicking malignancy.

Another debatable factrelated to GLM is the management of the disease,where it is still a dilemma in terms of the best management approach as there is no clinical consensus for anideal treatment [7]. The treatment of choice for GLM includes pharmacological such as antibiotics and corticosteroids or surgical management such as wide local excision and mastectomy [8].

Expectant management with steroids is found to be ameliorable and has high success rates [9]. A study has reported that steroid administration might shrink lesions both pre and postoperatively in persisting masses [10]. However, the recurrence rate in patients managed with expectant management is higher than surgical management, prompting us to offer surgical options to our patients. Studies have also shown that the recurrence rate of GLM in patients treated with corticosteroids can be as high as 50% [11]. Therefore, treatment using steroids should be used cautiously. It has several side effects, such as Cushing's syndrome, weight gain, and hyperglycemia, which led to our patient choosing surgical treatment.

On the other hand, antibiotics do not provide any proven benefit as no studies conducted on antibiotic therapy alone in GLM. A study by Rahim, 2017 mentioned that only 3 of 21 patients recovered with antibiotics usage alone from GLM and the remaining required usage of steroids and surgical intervention. We did not use any form of antibiotic on our patient aligned with another study that reported none of their patients had fully recovered using antibiotics alone in treating GLM [13].

Surgical management is still one of the main options for GLM [8]. Common surgical methods that are being used include incision and drainage (I&D), multidirectional deep drainage (MDD), wide local excision and mastectomy. Surgical treatment appears to be favourable compared to other alternative treatments, especially in recurrence and post-treatment recovery [14]. While wide local excision surgical method has a low recurrence rate, it may lead to scar related complications and cosmetic deformity of the breast. A patient with a large lump like our patient requires complete removal of the mass and complete assurance of the negative margin. However, the patient needs to be alerted on the defacement of the breast structure before the surgery [12]. I&D was not offered to this patient as it may lead to non-healing incision tracts resulting in the formation of sinus tracts [15]. Surgery remains the best treatment option as it is proven to have early recovery and high success rates. In our patient, wide local excision was performed as a diagnostic and therapeutic intervention aligned with reported literature as it proved to be an effective treatment option of GLM.

## **Conclusion**

Granulomatous lobular mastitis is a rare benign inflammatory disease of the breast. It manifests as a rapidly growing lump and usually appears immediately after cessation of breastfeeding, which establishes the rarity of our patient clinical presentation. Histopathological examination of the surgical specimen remains the diagnostic gold standard for GLM. Wide excision surgical treatment of the lump forming GLM is still considered the best therapeutic option with a low recurrence rate and complications.

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