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## Uncommon Location of Idiopathic Granulomatous Mastitis: A Case Report

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## ABSTRACT

**Background:** Idiopathic granulomatous mastitis (IGM) is a rare benign disease involving breast parenchyma mostly in the periareolar region. Childbearing women with a recent history of pregnancy and lactation are more at risk of IGM. The common locations of IGM are retro areolar or periareolar of the breast, but involvement of the axillary region in nonpregnant woman has never been reported elsewhere.

**Case presentation:** A 36-year-old female with a history of two times of pregnancy and lactation 8 months prior to presentation, referred with pain and swelling in the right axillary area. The past medical history and habitual history were negative and she did not use oral contraceptives or other medications. Local physical examination showed normal breasts with bilateral accessory breasts. A tender mass with the size of 4x6cm was palpable in the right axillary region accompanied by erythema and a few secretory fistulas without lymphadenopathy. Cell blood count, fasting blood glucose, HbA<sub>1c</sub>, and serum prolactin were normal. Ultra-sonography (US) demonstrated a soft tissue swelling, edema, and a decreased echogenicity area in the right axillary region compatible with IGM, which was further confirmed by biopsy. The patient was prescribed for on prednisolone 50mg per day and the condition did not improved for two months. To exclude other possible etiologies due to the atypical location, the patient underwent a second US and core-needle biopsy which confirmed the diagnosis of the axillary IGM. Prednisolone was tapered off and a non-steroidal anti-inflammatory drug (NSAID) started. All the symptoms improved in a month and fully resolved in 3 months.

**Conclusion:** IGM is not fully known yet, the presentation and the location can be variable. Considering IGM as a probable diagnosis in inflammatory presentation in the axillary region in patients with accessory breasts is suggested.

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## INTRODUCTION

Granulomatous mastitis (GM) is a rare benign non-necrotizing granulomatous condition that involves breast parenchyma, which is grouped into two main categories of primary or idiopathic (IGM) and secondary. Secondary GM can be due to infections like

histoplasmosis, actinomycosis, and autoimmune diseases such as sarcoidosis, granulomatosis with polyangiitis, IgG4-RD mastitis, foreign body reaction, and fat necrosis.<sup>1</sup> IGM was firstly introduced by Kessler and Wolloch in 1972.<sup>2,3</sup> Only 0.44-1.6% of the breast biopsies are diagnosed with IGM.<sup>4</sup> It mostly involves women of childbearing age and occurs a few years after pregnancy and lactation.<sup>5</sup> Although the exact pathology is not well-known, there are observations suggesting that Hispanic origins and people from middle-east are predisposed to developing

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IGM.<sup>3,6,7</sup> The pathogenesis may be due to autoimmunity, infection, hormonal imbalance, smoking, antitrypsin deficiency, and oral contraceptives; thus, the proper exclusion of possible etiologies should be taken into account.<sup>3</sup>

The incidence of IGM has been reported to be 0.37% in the United States, but unfortunately there are no clear reports about the incidence in the Middle east.<sup>8</sup> IGM mostly presents as a tender unilateral mass in the retro areolar of the breast; bilateral masses are rare. Erythema, skin changes, nipple contraction, abscess, fistula, and lymphadenopathy may also be present.<sup>9-12</sup> Early diagnosis of IGM is important because of its high similarity with breast cancer.<sup>11</sup> Because of similar findings of breast cancer and tuberculosis on presentation and radiological assessments, histological investigation is required. Besides, the secondary GM should be excluded via history and paraclinical assessment, making the final diagnosis faster.<sup>1</sup> An international multidisciplinary consensus in 2021 agreed on comprehensive workup by clinicians, radiologists and pathologists and the use of history taking, physical examination, imaging and laboratory modalities to finalize the diagnosis.<sup>13</sup> The delay in diagnosis on average of 4-5 months is common and reported in a systematic review on 3060 cases.<sup>14</sup> In this case report, we present an unusual location of IGM in the accessory breast in the axilla region, which delayed the process of diagnosis.

### CASE PRESENTATION

A 36-year-old female presented with pain and swelling in the right axillary area for 3 months. She had two children and breastfed both of them for two years for 8 months prior the presentation. She denied any other specific disease in the past medical history and did not have a family history of breast cancer. She had never used alcohol and cigarettes and did not use oral contraceptives and other medications.

The systemic examination was unremarkable. On local physical examination, the left breast was intact and bilateral accessory breasts were prominent. A tender mass with ill-defined borders of an approximate of 4x6cm was palpable in the right axillary region accompanied by erythema and a few secretory fistulas. Lymph nodes were not palpable. Figure 1 demonstrates the lesion.

In the blood test, the cell blood count, fasting blood glucose, HbA<sub>1c</sub>, and serum prolactin were normal. In bilateral full digital mammography, focal asymmetry was seen at the right axillary region which required ultra-sonography (US) for better assessment. The US at the time of presentation found soft tissue swelling, edema, and a decreased echogenicity area in the right axillary region without detectable fluid collection and

abscess, suggesting subacute cellulitis. The core needle biopsy was performed showing a breast tissue in the axilla with an inflammatory process containing mixed infiltrates of inflammatory cells as well as granuloma formation which was suggestive of granulomatous mastitis. The specimens were then sent for antibiogram and culture in growth media and PCR for TB and fungus in another test tube. The growth media did not detect bacterial colonization, and PCR for TB and fungus were also negative. With an impression of IGM, prednisolone 50mg per day was prescribed for the patient.



**Figure 1.** Right axillary region with secretory fistulas

In the 2-month follow-up, the condition did not improved. Since the location of the lesion was atypical for IGM and the patient did not respond to prednisolone, another US and a core-needle biopsy were performed to exclude other possible etiologies. The second US reported a 42x15mm hetero echoic structure thick collection in the right axillary region associated with skin thickening which was consistent with focal granulomatous mastitis with BIRAD III (Figure 2). A core-needle biopsy reported similar findings to the previous biopsy which was suggestive of lobar (idiopathic) granulomatous mastitis (Figures 3&4).

With the diagnosis of corticosteroid resistant IGM, the prednisolone tapered off and naproxen 500mg was prescribed twice per day. After a month, the erythema and secretion decreased and the mass became softer than the first presentation. In the 3-month follow-up, the erythema and mass completely resolved, and the scars of previous fistulas appeared; thus, naproxen was discontinued. Nine months after the full remission, the patient was followed for the possible relapse. The breast was intact with normal shoulder movement and scars of resolved fistulas.



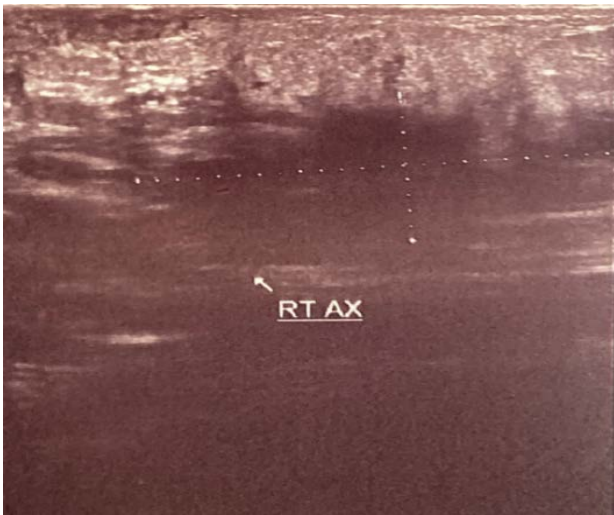


Figure 2. US of the right axillary region

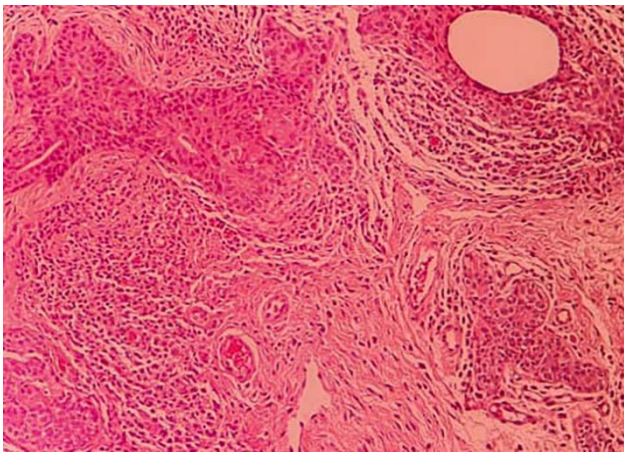


Figure 3. A granulomatous reaction in lobular granulomatous mastitis x400 Hematoxylin Eosin stain

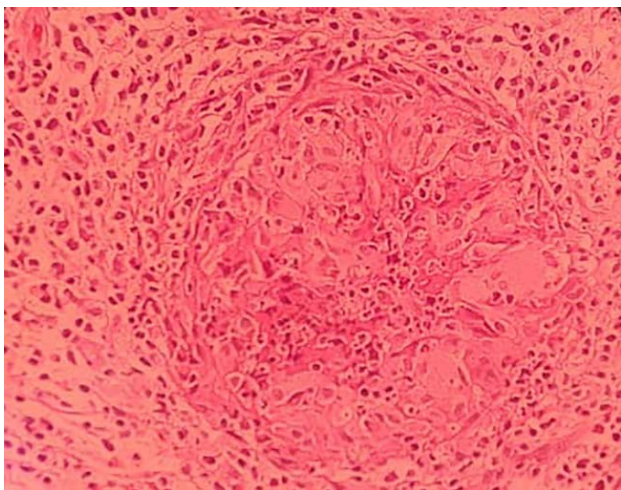


Figure 4. Lobulocentric pattern of inflammation in lobular granulomatous mastitis x200 Hematoxylin Eosin stain

### DISCUSSION

In this case report, we presented an unusual location of IGM, which was not responsive to a two-

month consumption of steroids. Although IGM is mostly present in the periareolar of the breast, the involvement of other locations especially in presence of accessory breasts is not unexpected. Cancer, mastitis, fibroadenoma, phyllodes tumor, and fibrocystic change are the most common diseases that can involve accessory breasts, but IGM should be kept in mind as a possible disease as well.<sup>15</sup> Two case reports have presented IGM in accessory breast tissue in axilla recently, but both cases were in pregnant women.<sup>16,17</sup> Physicians should also consider that suppurative hidradenitis can mimic the same presentation with IGM in the axillary region. Moreover, our patient has a normal prolactin level, which suggests the upregulation of prolactin receptors or their hypersensitivity to prolactin in this case.<sup>13</sup>

IGM is a self-limited disease that resolves within 2 years, but the long period of convalescence is disturbing to many patients.<sup>11,12,18</sup> To date, treatment options have been developed and improved in recent years. Observation is the first approach to treatment in IGM patients. While many clinicians prefer medications for severe and prolonged cases in the first place and start the treatment with oral steroids. The presence of fistula in our patient suggested an advanced disease which required treatment and thus started on corticosteroids.<sup>13</sup> Although steroids can be effective in nearly 80% of the cases, other patients may require alternative treatments. Immunosuppressive agents such as Methotrexate and Azathioprine are the two most common medications in unresponsive cases or patients who have developed complications due to steroids.<sup>19</sup> A study found a combination therapy of methotrexate and steroids with better response in IGM patients than steroids alone.<sup>20</sup> Also, Methotrexate had a high effectiveness methotrexate in patients resistant to corticosteroids.<sup>21</sup> The rate of remission with methotrexate is estimated from 58% to 100% in cases with other autoimmune presentations plus IGM.<sup>22</sup> The response to steroids and immunosuppressor agents is another indication that IGM is an autoimmune disease; however, the potential adverse effects and the need for close monitoring in treatment with immunosuppressive agents discourage some physicians from using them.<sup>5</sup>

To date, non-steroidal anti-inflammatory drugs (NSAIDs) have been prescribed for IGM in recent years and a good response has been observed in a study on 374 patients in Iran.<sup>23</sup> In the presented patient, the undesired complications of immunosuppressors make her refuse to take immunosuppressors. Due to the success of NSAID in the mentioned study on Iranians, we chose to put the patients on NSAID. We found the same result in the mentioned case. In a meta-analysis, surgery as a less favorable treatment option in IGM was not found to be different in relapse rate from non-



surgical treatment. Fistula formation, late wound healing, and disfigurement are also some other possible complications.<sup>24–26</sup> Patients with IGM can experience several relapses for a long time; thus, using medications like NSAIDs with fewer complications than surgery and other non-surgical options should be taken into account, making it a priority especially in patients with limited access to health facilities to monitor the possible complications of medications, and also reluctant to take immunosuppressors.<sup>27</sup>

### CONCLUSION

In conclusion, IGM is a rare inflammatory disease. IGM should be considered as a probable diagnosis in inflammatory presentations in uncommon regions like milk line due to the existence of accessory breasts and can facilitate diagnosis in unusual presentations.

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