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Case report

Idiopathic Granulomatous Mastitis: A Case Report

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Abstract
Idiopathic granulomatous mastitis (IGM) is a rare, chronic, benign inflammatory disease of the breast. It is characterized by the development of a painful breast mass that gradually increases in size. Its etiology is unclear. It impacts women of reproductive age who have a history of pregnancy and lactation. Imaging findings were nonspecific; histopathologic examination is the best diagnosis. Its management combines surgery, antibiotics, corticosteroid therapy, and anti-inflammatories. We report a 38-year-old woman with a history of pregnancy and breastfeeding through the right side only presented with severe, increasing breast pain and noticed a small lump with tenderness and warmth; the physical examination revealed a mass with associated redness in the upper inner quadrant of her left breast. Ultrasonography revealed localized duct ectasia with mural thickening associated with an irregular hypoechoic mass, suggesting granulomatous mastitis. The true-cut biopsy confirmed the diagnosis. The abscess was evacuated through a minor incision performed under local anesthesia three times during 6 months, accompanied by antibiotic treatment. A treatment-free follow-up period resulted in significant improvement and complete resolution after 24 months. To validate the diagnosis of IGM, a thorough evaluation of possible etiologies is essential. Ultrasonography is the most common diagnostic modality. Histologically, it is characterized by neutrophils and the lack of caseous necrosis. Treatment is contentious, with surgical excision reserved for complex and refractory cases. Idiopathic granulomatous mastitis is a rare breast condition characterized by poorly understood causes and undefined treatment protocols. This condition warrants consideration in women of reproductive age.
Keywords: Granulomatous, Mastitis, Imaging, Ultrasonography, Histopathology, Management.

Introduction

Granulomatous mastitis, also known as idiopathic granulomatous mastitis (IGM), is an intriguingly rare, chronic, benign inflammatory breast disease. It primarily affects women of childbearing age, often developing within 5 years postpartum or during lactation [1,2]. Despite its rapidly increasing incidence in recent years, the etiopathogenesis of IGM remains unsolved. Researchers have suggested multiple causes, including tuberculosis (TB), sarcoidosis, mycotic and parasitic infections, diabetes mellitus, and foreign body or hypersensitivity. Additionally, autoimmune processes have been suggested as a possible cause; it usually presents with sinus and abscess formation and noncaseating granulomatous inflammation [2,3].

GM is a relatively recently reported condition, initially described by Veyssiere et al. in 1967 [10]. In 1972, Kessler and Wolloch conducted a clinical and histological diagnosis and evaluation of the etiology of GM and treatment modalities [4,5]. Clinically, IGM appears with symptoms including inconstant pain, asthenia, inflammation, erythema, and a palpable mass, the latter being the most common. The clinical and radiological features associated with IGM are frequently similar to those of breast cancer and several benign inflammatory breast diseases, resulting in misdiagnosis and prolonged treatment [5]. Ultrasound (US) and mammography play crucial roles in diagnosing IGM. However, due to the ambiguous nature of imaging results, the accurate diagnosis of IGM necessitates histopathological confirmation and the elimination of alternative diseases [4-6].

The treatment of IGM involves one or a combination of several treatment options, including abscess drainage, antibiotics, corticosteroids, various immunosuppressive therapies, and treatment-free close follow-up. Treatments can sometimes include radical interventions, such as mastectomy [7,8]. Many authors focused on the suitability of conservative versus surgical treatment options in their review articles, which aimed to clarify diagnosis and management decisions [6-8]. The main goal of this study is to share our experience with a case report of idiopathic granulomatous mastitis, which will contribute to a better understanding and improved management of IGM.

Case report

The patient is a 38-year-old woman with no breast trauma and no family history of tuberculosis, breast cancer, or autoimmune diseases. She has six children with cesarean deliveries and has never taken contraceptives. The patient breastfed her children for one year each through the right side only, three years from her last delivery. She reported experiencing severe, increasing breast pain for 10 days and noticed a small lump with tenderness, warmth, and redness in the upper inner quadrant of her left breast. The patient

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was concerned about her symptoms, representing infectious mastitis. She denied fever, rash, synovitis, myalgias, lymphadenopathy, or other systemic symptoms. She reported no signs of infection.
During the clinical examination, we palpated a tender mass measuring 4 cm in diameter over the superior-internal quadrant of her left breast, along with mild erythema. No nipple discharge was present. She had a typical temperature. The patient exhibited no infectious sites, and the contagious microbiology examination was normal. We decided to order an ultrasound of the left breast and axilla. She had a fever (37.7°C), and her mass had grown to 10 cm over five days, incredibly tender to palpation.
Ultrasound showed signs of ductal mastitis; the left breast has a dense glandular composition and retro and circumareolar duct ectasia with a maximum caliber of 6 mm and mural thickening and turbid mobile contents, which are associated with echogenic fat planes. The ultrasound reveals an interconnected, heavily turbid, partially loculated collection along the 3 o'clock axis, with the largest loculus averaging 3*0.8 cm. The overlying skin connects to this collection, displaying a sinus track measuring 3 mm and associated oedema. BI-RADS 4A classifies the patient as having no pathologically enlarged axillary lymph nodes and an echogenic pattern of chest wall muscles (Figure 1).

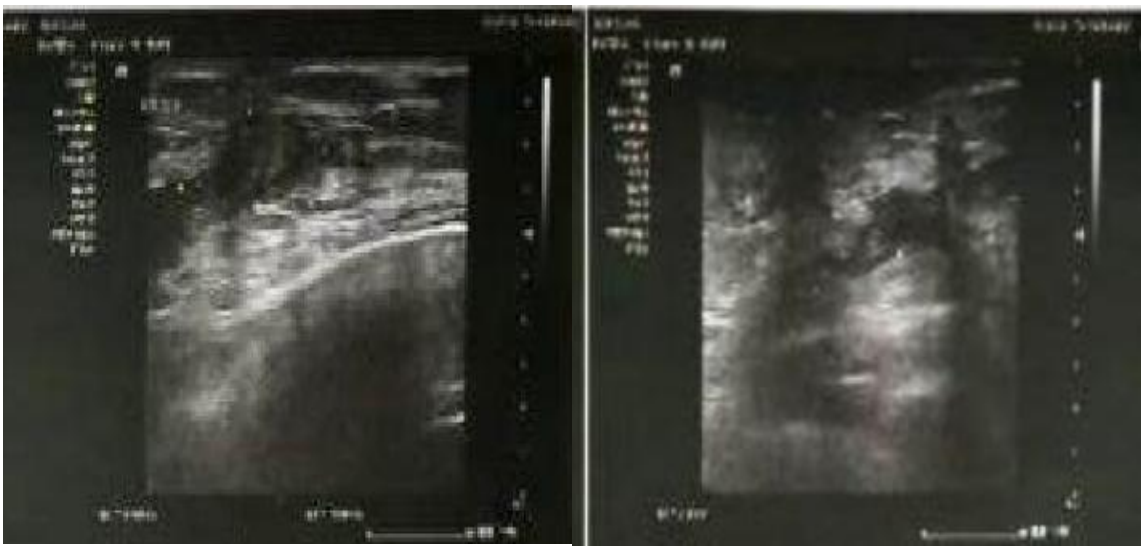

Figure 1. Ultrasonography of the left breast findings were seen with granulomatous mastitis. It shows areolar duct-ectasia, turbid mobile contents with echogenic fat planes, and the largest loculus connected to overlying skin with a sinus track related to oedema and echogenic fat planes.

The patient underwent an ultrasound-guided tru-cut biopsy (TCNB) to obtain a tissue sample. The next day, the biopsy site drained pus and became red, swollen, and painful. We decided to start an intravenous antibiotic therapy course, specifically cephalexin and acetaminophen, twice daily for 8–10 days; after 6 days of treatment, a cutaneous fistulization appeared, and the inspection of the breast found a clinical sign described as “orange peel,” resulting from pus to the skin. Subsequent bacterial, fungal, and tuberculosis cultures and stains yielded negative results. Histology was performed on multiple gray-white tissue cores, showing wide logocentric chronic inflammation with granulomatous reaction. Entangling scattered multinucleated giant cells together with neutrophilic infiltration was noted. The background showed fibrosis and proliferating capillaries. The examined sample showed no signs of atypia or malignancy. The final diagnosis of ultrasound TCNB was granulomatous mastitis with suppuration.
We performed transcutaneous drainage on her, accompanied by an incisional biopsy, which led to some improvement in her symptoms of inflammation, but recurrence occurred in less than one month after surgery. We also performed three rounds of drainage treatment, antibiotic therapy, and wound dressings in the next three months. In the next six months, the patient got pregnant, so we conducted a treatment-free follow-up, and the abscess drained spontaneously. The follow-ups after her delivery occur in one month, three months, and six months, a gradual improvement in healing. Her lesions never returned, and she has been asymptomatic until now.

Discussion

Granulomatous mastitis, also referred to as IGM, is a rare, benign, inflammatory, chronic breast condition. It is marked by noncaseating granulomas in the lobules of breast tissue. It mostly happens to women of childbearing age, most often during pregnancy, within 5 years of giving birth, or while breastfeeding [1, 2]. However, the age of occurrence can vary significantly. Studies have been reported on 11-year-old girls and individuals in their 60s, 70s, and 80s; the patient in our study, 37 years old, had a lactation history of more than one year for each of her babies [6-9]. The pathogenesis and etiology of IGM remain unclear and poorly

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understood. Studies have suggested but not substantiated a correlation between IGM and various agents such as autoimmune disorders, breast trauma, hyperprolactinemia, alpha-1 antitrypsin deficiency, local irritations, parasitic, mycotic, and viral infections, smoking, diabetes mellitus, and the use of oestroprogestogenic contraception [5, 8,]. Several studies point out the potential involvement of the extravasation of galactophoric substances after lesions of the ductal epithelium [8,9]. In our study, the patient had never experienced any of these issues and had no prior history of using oral contraceptives. However, it exclusively used her right breast for breastfeeding, never using the left side where the GM occurred.

GM is a relatively recently reported condition; Kessler and Wolloch first recognized idiopathic granulomatous mastitis (IGM) in 1972 [4, 5]. In the literature, IGM is typically demonstrated as a unilateral, detectable breast mass that ranges from 1 to 20 cm in size with sharp borders [1,2,3,8]. Although it commonly appears in the upper outer quadrants, it can appear in any part of the breast; in our case study, a patient had unilateral involvement in the upper inner quadrants with a 10 cm palpable mass. The solid mass may exhibit cutaneous symptoms such as irritation, the spontaneous appearance of pseudo-tumor forms, and a retraction of the nipple that mimics cancer. In chronic and severe cases, the condition may lead to the formation of microabscesses, ulceration, and fistulization of the skin, resulting in visible scars and pus drainage via sinus tracts extending to the skin. Furthermore, ipsilateral axillary lymphadenopathy can develop in certain cases [8,9]. Generally, there is no evidence of an inflammatory syndrome, and bacteriological and mycological samples are sterile [8]. A mild inflammatory response and superinfection with Staphylococcus aureus can appear in abscess cases. Concerning immunology, autoimmunity markers, including antinuclear antibodies and positive rheumatoid factor, may be detected [9].

The patient's medical history, clinical presentation, radiological studies, and histopathological examinations are used to make the diagnosis and differential diagnosis of IGM. Radiological procedures, including mammography (MM), magnetic resonance imaging (MRI), and ultrasonography (US), are polymorphic and nonspecific in most patients and are used mainly to exclude breast cancer rather than confirm GM [2,4]. In addition, the extent of the lesion and the treatment effectiveness are assessed. Subsequently, it is essential to accurately assess the breast volume that may be excised during surgery and the volume of the fascia flap required to reconstruct the affected area.

Ultrasonography is the most common diagnostic modality for patients showing signs of IMG regardless of age. Due to its non-radiative characteristics, which allow evaluating the lesion's dimensions, ultrasound results may reveal irregular hypoechoic masses with vague borders with internal hypoechoic tubular lesions, sometimes accompanied by heterogeneous galactophore ectasia; image-guided biopsies commonly employ ultrasound (puncture or surgical flattening). In this study, the ultrasound findings of our patient detected abscess formation, increased echogenicity of fat tissue due to inflammation, and irregular hypoechoic lesions connected by tubular extensions [4,5].

Histopathologic evaluation continues to be the sole accurate method for diagnosing IGM. Pathological examination uses various biopsy techniques, such as percutaneous (fine needle, tru-cut, etc.) [12,13], incisional, and excisional biopsy methods. Here, we conducted a tru-cut biopsy before surgery, which showed widespread, long-lasting inflammation with a granulomatous response. Entangling scattered multinucleated giant cells together with neutrophilic infiltration was noted. The background revealed fibrosis and proliferating capillaries with no ductal dilatation had been shown, which effectively confirmed the diagnosis of IGM [4,14]. Furthermore, a surgical incision biopsy performed during drainage confirmed the diagnosis of GM in our case.

The treatment of IGM is controversial and uncodified. Multiple treatment modalities are examined, encompassing surgical resection, antibiotics, methotrexate, bromocriptine, colchicine, immunosuppressive agents, corticosteroids, and conservative monitoring. Treatment primarily relies on the clinician's or surgeon's experience and the severity of the patient's disease. Despite being the primary treatment for IGM, surgical excision is now considered a last resort due to the risks of recurrence, cosmetic complications, and the formation of fistulas postoperatively [15-17].

In our study, the patient underwent three rounds of pus drainage in conjunction with antibiotic therapy before and during pregnancy. Subsequently, surgical drainage and antibiotics ceased, and conservative treatment-free follow-up was opted for. The absence of a standard treatment algorithm, the high recurrence rate despite appropriate treatment, and the potential for IGM to mimic breast cancer in radiologic studies are the most critical points regarding the diagnosis and treatment of IGM. This underscores the urgent need for further research and development of a standard treatment algorithm for medical challenges.

Conclusion

IGM is a rare, chronic, benign inflammatory disease of the breast. It is characterized by the development of a painful breast mass that gradually increases in size. Its etiology is unclear, and it impacts women of reproductive age who have a history of pregnancy and lactation. Imaging findings are nonspecific;

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histopathologic examination is the best diagnosis. Its management combines surgery, antibiotics, corticosteroid therapy, anti-inflammatories, and treatment-free follow-up.

Informed consent
Written informed consent was obtained from the patient for her anonymized information to be published in this article.

Conflict of interest. Nil

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الملخص
التهاب الضرع الحبيبي مجهول السبب هو مرض التهابي حميد نادر ومزمن يصيب الثدي. يتميز بتطور كتلة مؤلمة في الثدي تزداد حجمها تدريجيًا. مسبباته غير واضحة. يصيب النساء في سن الإنجاب اللاتي لديهن تاريخ من الحمل والرضاعة. كانت نتائج التصوير غير محددة؛ الفحص النسيجي هو أفضل تشخيص. يجمع علاجه بين الجراحة والمضادات الحيوية وعلاج الكورتيكوستيرويد ومضادات الالتهاب. نعرض حالة امرأة تبلغ من العمر 38 عامًا ولديها تاريخ من الحمل والرضاعة التي كانت من خلال الجانب الأيمن فقط وجاءت مع ألم شديد ومتزايد في الثدي ولاحظت كتلة صغيرة مع رقة ودفء؛ كشف الفحص البدني عن كتلة مصحوبة باحمرار في الربع الداخلي العلوي من ثديها الأيسر. كشف التصوير بالموجات فوق الصوتية عن توسع موضعي في القناة مع سماكة جدارية مرتبطة بكتلة غير منتظمة ناقصة الصدى، مما يشير إلى التهاب الضرع الحبيبي. أكدت خزعة القطع الحقيقية التشخيص. تم إخلاء الخراج من خلال شق صغير تم إجراؤه تحت التخدير الموضعي ثلاث مرات خلال 6 أشهر، مصحوبًا بعلاج بالمضادات الحيوية. أدت فترة المتابعة الخالية من العلاج إلى تحسن كبير وحل كامل بعد 24 شهرًا. للتحقق من صحة تشخيص التهاب الضرع الحبيبي مجهول السبب، فإن التقييم الشامل للأسباب المحتملة أمر ضروري. الموجات فوق الصوتية هي الطريقة التشخيصية الأكثر شيوعًا من الناحية النسيجية؛ تتميز بالعدلات وغياب النخر الجبني. العلاج مثير للجدال، مع الاستئصال الجراحي المخصص للحالات المعقدة والمقاوم. التهاب الضرع الحبيبي مجهول السبب هو حالة نادرة في الثدي تتميز بأسباب غير مفهومة وبروتوكولات علاج غير محددة. تستحق هذه الحالة النظر فيها عند النساء في سن الإنجاب.

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