



The effect of corticosteroid treatment on bilateral idiopathic granulomatous mastitis

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ABSTRACT

Idiopathic granulomatous mastitis (IGM) is the commonly encountered form of granulomatous mastitis that may result into repetitive infections and/or abscess formation. Mastitis may develop secondary to a systemic disorder such as tuberculosis, diabetes mellitus, or rheumatoid arthritis, or it may develop as an idiopathic disorder. Idiopathic granulomatous mastitis is the most frequent form of all granulomatous diseases affecting the breast. This disorder frequently presents as painful and fast-growing mass in the breast. Biopsy is required to confirm diagnosis. Surgical excision and immunosuppressive treatment with corticosteroids are employed for therapeutic management. Here we present 3 female cases of bilateral IGM who were followed up and treated successfully with 1 mg/kg/day prednisolone.

Keywords: Idiopathic granulomatous mastitis, bilateral breast, corticosteroid treatment

INTRODUCTION

Mastitis is defined as the inflammation of the breast. The inflammatory process may sometimes be infectious. Mastitis affects women the most, particularly those who are middle aged. Idiopathic granulomatous mastitis (IGM) that mimics malignancy is a type of mastitis that has a chronic progression. Idiopathic granulomatous mastitis is a rare, chronic inflammatory disorder of which the etiology is not well known. It mimics breast cancer clinically and radiologically. It was first described in 1972 by Kessler and Wolloch (1) as noncaseating granulomatous inflammation in histopathological evaluation after ruling out infectious causes (such as tuberculosis and fungal infections) and noninfectious causes (such as sarcoidosis and vasculitis). Clinical presentation may be painful or painless palpable mass or skin fistula with retractions, thereby mimicking breast cancer (2). The disorder mimics breast cancer also in the ultrasonographic and mammographic evaluations (3). This study included three cases of IGM affecting bilateral breasts. The etiopathogenesis of IGM has been postulated to be a local autoimmune reaction or a secondary reaction to parturition, generally affecting young women unilaterally (4). Here we present three cases of granulomatous disease that were treated medically.

CASE PRESENTATIONS

Case 1

A 39-year-old woman who had previously given birth three times complained of redness of the skin, which she first noticed on the skin over the right and later on the left breast, along with pain and a palpable mass. She had a past history of excision of fibroadenoma from the upper lateral quadrant of her breast. The last time she gave birth was 5 years previously. She did not use oral contraceptives. On examination, we found hyperemia of indefinite margins on the skin over the medical parts of both areolas and an immobile palpable mass of 3 × 2 cm in the right breast and another of 4 × 3 cm in the left breast. White blood cell (WBC) count was 11.000 cells/mm³. The masses were tender and hard. There was no nipple discharge, fistula orifice, or axillary lymphadenopathy. Prediagnosis was mastitis and oral sefuroxime axetil 500 mg twice daily was prescribed empirically along with diclofenac sodium. Bilateral breast ultrasonography showed lesions of heterogeneous low density, of which the contours were lobulated and poorly demarcated. There were no fluid collections and bilateral mammography was unremarkable. On pathological evaluation of excisional biopsies from both breast, IGM was diagnosed. The patient developed a continuing discharge from the incision wound of the left breast. Steroid treatment was continued along with weekly examinations. Tapering of steroid treatment depended on weekly clinical findings. She responded clinically well to a 6-week steroid (1 mg/kg/day) regimen. Follow-up period was 24 months. There were no complications or recurrence.

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Case 2

A 31-year-old nulliparous woman who had been taking antidepressant drugs applied to the hospital complaining of redness of the skin over both breasts along with pain and swelling originating from the deeper regions of the breasts and spreading up to the retroareolar regions (Figure 1). WBC count was 9800 cells/mm³. Ultrasonographic diagnosis was mastitis with bilateral retroareolar abscesses that were drained and biopsies were obtained. Steroid treatment was continued along with weekly examinations. Tapering of steroid treatment depended on weekly clinical findings. On pathological evaluation, IGM was diagnosed, and she was administered a 6-week course of steroid (1 mg/kg/day) therapy. Follow-up period was 18 months. There were no complications or recurrence.

Case 3

A 35-year-old woman who had previously given birth twice reported the use of oral contraceptives for 1 year and presented with multiple abscesses in both breasts. The abscesses were drained and from beneath the fistula tract tru-cut biopsies were obtained from the lesion. WBC count was 11800 cells/mm³. Ultrasonographic diagnosis was multiple right breast masses with irregular margins and a highly hypoechoic echostucture with an irregular hyperechoic halo. Histopathological examination showed large nonnecrotic granulomas composed of numerous neutrophils, histiocytes, plasma cells, lymphocytes, and Langerhans-type giant cells. We perceived that granulomatous inflammation rendered the ducts to rupture. Idiopathic granulomatous mastitis was thus diagnosed histopathologically (Figure 2, 3). Steroid treatment was continued along with weekly examinations. Tapering of steroid treatment depended on weekly clinical findings. The patient responded well to a 10-week course of steroid (1 mg/kg/day) therapy. In the sixth week of follow-up, the patient developed dyspeptic complaints and proton pump inhibitor treatment was administered. Follow-up period was 20 months. There were no complications or recurrence.

Written informed consent was obtained from all patients who participated.

DISCUSSION

As a rare chronic inflammatory disorder of the breast that may be confused with carcinoma IGM more frequently affects women of the middle age (the third and fourth decades) and is generally encountered within few years of parturition (5). The etiopathogenesis is not well known. Proposed predisposing factors include autoimmunity, oral contraceptive use, infectious agents, tuberculosis, hormonal disorders, pregnancy, hyperprolactinemia, and alpha-1 antitrypsin deficiency (6, 7). The higher incidence of IGM during the postpartum and lactation periods and that among women using oral contraceptives draws attention to hormonal factors. Approximately one-third (33%) of all IGM patients have reported the use of oral contraceptives and cases not related to pregnancy (8, 9). One of our cases reported a history of use of oral contraceptives. Histopathological findings including inflammatory cells in a lobular pattern suggest cellular autoimmune reaction against some histological elements of the breasts. Some cases have been reported to be affected by other autoimmune disorders (10). However, in contrast to other autoimmune disorders, the absence of vasculitis or prominent plasma cell infiltration



Figure 1. A case with bilateral mastitis: Arrows show external opening

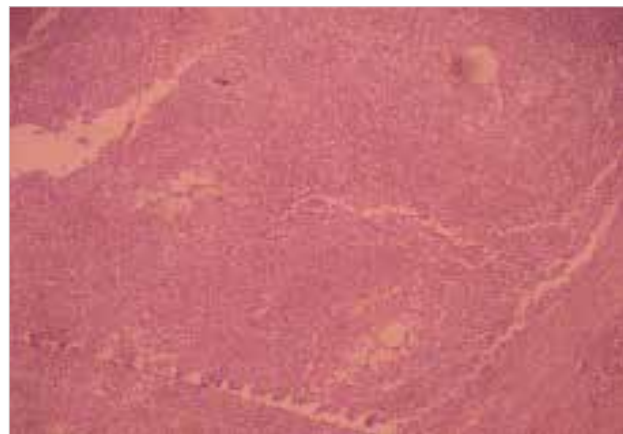


Figure 2. Histopathological features: At a smaller magnification, area depicting three separate small noncaseating granuloma structures can be observed

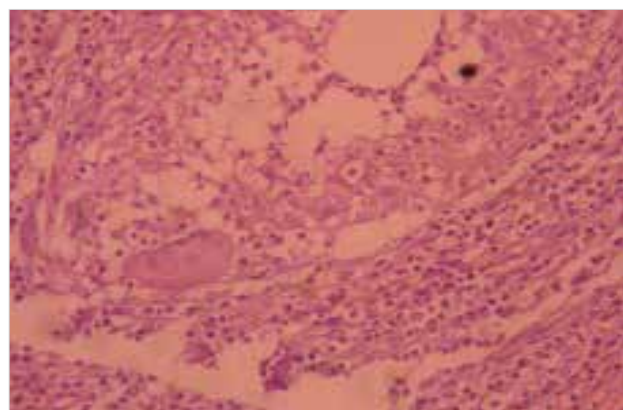


Figure 3. At a larger magnification, granulomas appear to be composed of the epithelioid histiocytes and multinuclear giant cells

disfavors the possibility of autoimmune reaction. Thus far, no microorganism has been isolated from the lesions, thus ruling out infectious factors.

In addition to mimicking breast carcinoma, other diseases that may cause a granuloma in the breast, such as tuberculosis, syphilis, histoplasmosis infections, foreign-body granuloma, vaccination granuloma, mammary duct ectasia, sarcoidosis, Wegener's granulomatosis, giant cell arteritis, and polyarteritis nodosa, should also be excluded. Here we reported three

cases of IGM successfully treated with 40 mg/day (1 mg/kg/day) prednisolone. Because steroid administration can cause some severe adverse effects, such as opportunistic infections, dermatological manifestations, hypertension, peptic ulcer disease, neuropsychiatric symptoms, hyperglycemia, myopathy, and osteoporosis, we believe that the minimum dosage of a steroid should be recommended. The duration and recommended dosage of steroid treatment require further clinical research to establish an adequate treatment regimen. Several other reports of using 60 mg/kg/day prednisolone to treat IGM have also been published. However, in the literature the recommended dose ranges between 0.5 and 2 mg/kg/day (11-15).

One of our cases had the history of use of oral contraceptives before and after parturition. She had no diagnosis for other autoimmunities and the histopathological evaluation did not show vasculitis. Sarcoidosis is a granulomatous disorder of unknown cause. The granulomas in sarcoidosis are discrete and perivascular and include no necrosis or microabscesses. In our cases, there were no radiological findings attributable to sarcoidosis, and the presence of microabscesses on histological examination rules out sarcoidosis.

Idiopathic granulomatous mastitis affects both breasts at the same frequency and can be bilateral in 5%–15% of cases. Involvement of upper lateral quadrant is most commonly reported (2, 10, 14). Our study included bilateral cases of granulomatous mastitis. Clinical presentation of the disorder is mostly painful or painless palpable mass in the breast. It may present as acute inflammation with erythema, tenderness, and warmth or may exhibit a chronic progression with fistula, abscesses, and ulceration of the skin over the breast, retraction and leaks from the nipple thereby mimicking breast cancer (11). Although very rare, axillary lymph nodes may be palpable.

CONCLUSION

This study included three cases of IGM affecting bilateral breasts. Here we described the clinical presentation, diagnosis, evaluation, and treatment of this disorder. Its clinical presentation may mimic breast cancer or abscess.

Thus far, no consensus regarding the treatment modality has been reached. Current studies suggest preventing aggressive intervention as the initial approach in cases of IGM with minimal clinical findings. However, surgery is unavoidable in cases that do not respond to medical treatment or present with grave clinical findings (such as recurrent abscesses and fistula).

Nonoperative approaches and medical treatment with steroids are preferred for patients with bilateral IGM.

Informed Consent: Written informed consent was obtained from patients who participated in this case.

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