

## Idiopathic Granulomatous Mastitis: A Case Successfully Treated with a Minimum Dose of a Steroid

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Idiopathic granulomatous mastitis (IGM) is a benign, chronic non-caseating breast disease, often mistaken for breast cancer. It usually affects women of child-bearing age. The treatment for IGM is inconclusive. In the past, surgical intervention was suggested due to the possibility of malignancy. However, in recent reports, corticosteroid therapy has been used with a good response. We present a case of a female with IGM who was treated successfully with 0.8 mg/kg/day prednisolone. (*Chang Gung Med J* 2005;28:431-5)

**Key words:** granulomatous mastitis, corticosteroid, breast disease.

Idiopathic granulomatous mastitis (IGM) is a benign breast disease first described by Kessler and Wolloch in 1972.<sup>(1)</sup> This disease usually affects women of child-bearing age with a history of oral contraceptive use. It is characterized by chronic, non-caseating granulomatous lobulitis of unknown etiology. Failure to recognize granulomatous mastitis, which may be consequently mistaken for breast carcinoma, has previously resulted in unnecessary mastectomies or open biopsies.<sup>(2)</sup> Herein, we report on a woman with IGM who was treated successfully with steroid therapy.

### CASE REPORT

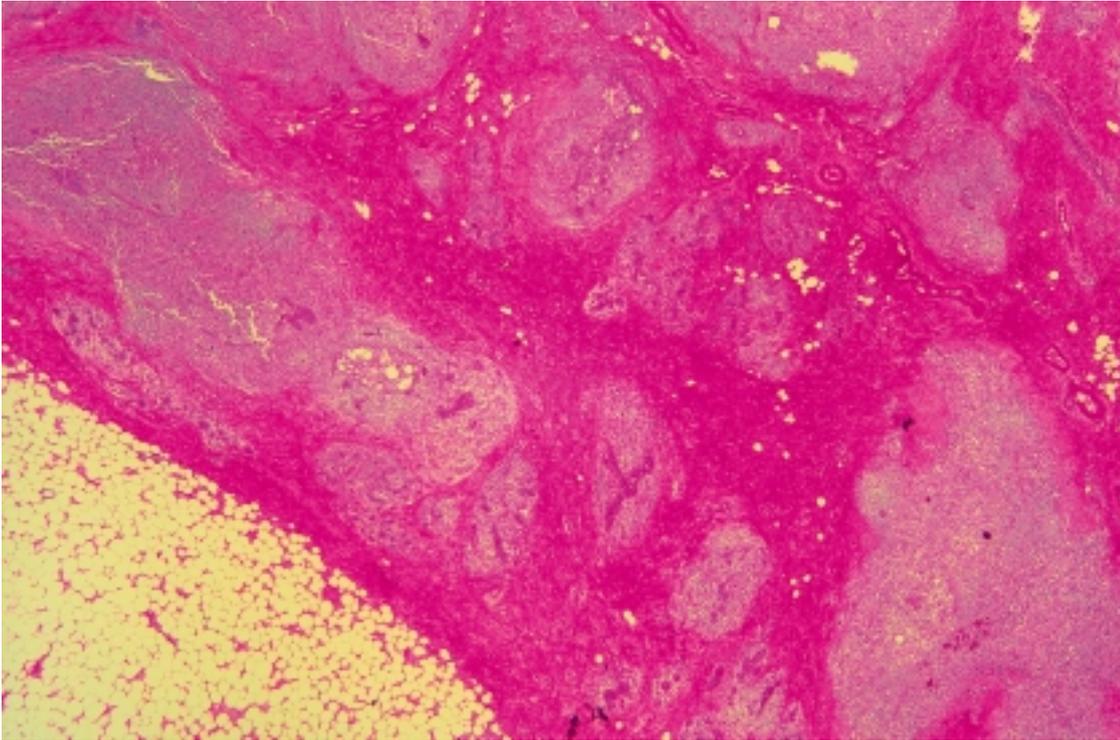
The patient, a 39-year-old woman, first became aware of a right tender breast mass (8 x 7 x 1.5 cm) with localized redness in November 2001 (4 years after the birth of her last child) while living in Australia. She had breastfed both of her children for 10 to 11 months each and had had gestational diabetes during both pregnancies. She had taken oral contraceptive agents for 2.5 years after giving birth

to her second child. The patient had no familial history of breast cancer. She first consulted her family physician in Australia and received a tentative diagnosis of a breast abscess. Open drainage and 6 courses of antibiotics were given, yet her symptoms persisted. In Australia, a right breast core biopsy was first done, followed by open drainage and an excisional biopsy (Figs. 1, 2). Gram stain and bacterial culture, fungal culture, acid-fast bacillus stain, and mycobacterial culture of the tissue specimen were negative. The erythrocyte sedimentation rate (ESR) was 44 mm/h, and the chest roentgenogram was normal. Microscopic examination of the excisional biopsy specimen showed a chronic abscess lined by severely inflamed granulation tissue containing numerous foamy macrophages and epithelioid histiocytes, with small lymphocytes and aggregates of neutrophils in small suppurative foci. In areas of microabscess formation/suppuration, there was a lipid vacuole in the center of the inflammatory process. A diagnosis of IGM was established.

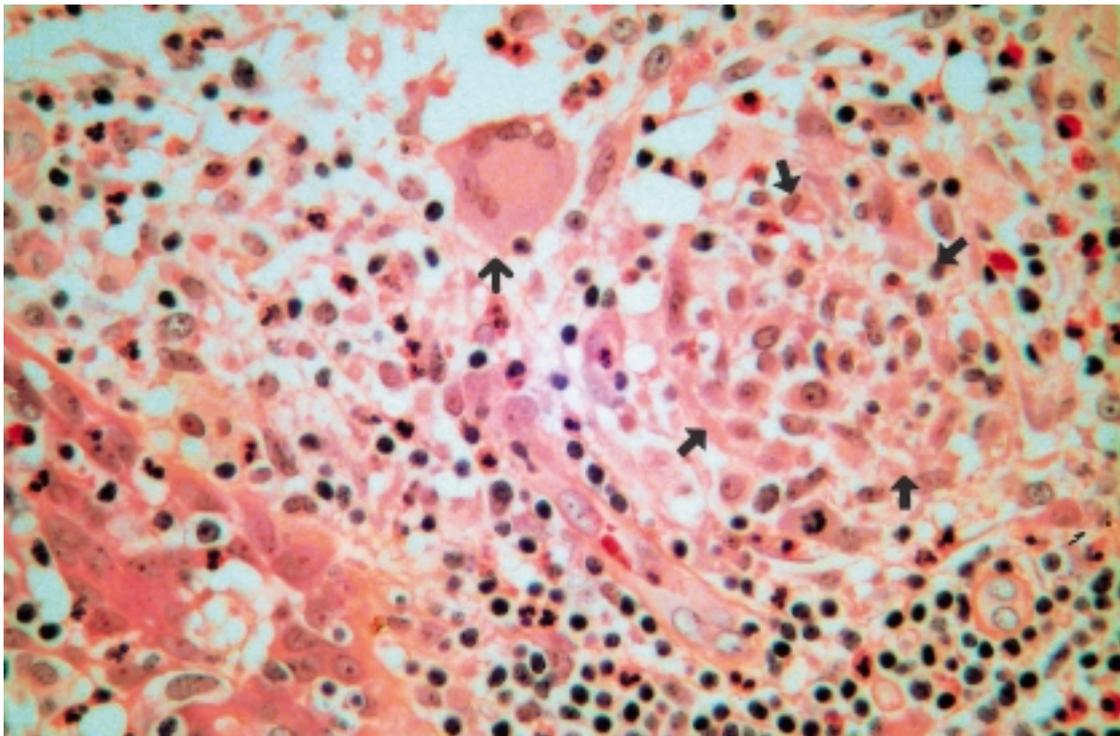
She visited our breast clinic for a second opinion. A physical examination showed indurations,

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**Fig. 1** Granulomatous inflammation involving major lobular areas (H&E, 20x).



**Fig. 2** Granulomatous inflammation, manifested by copious epithelioid histiocytes forming a granuloma (single thin arrow), lymphocytes, Langhan's multinucleated giant cells (multiple thick arrows), and occasionally eosinophils (H&E, 400x).

multiple abscesses, and a fistula with a pus-like discharge in the medial part of the patient's right breast. With the help of the pathological reports from Australia, 30 mg/day prednisolone (0.6 mg/kg/day) was given for 28 days and then was tapered off over the course of 3 weeks. After 1 week of steroid therapy, the fistula had closed, the abscess had subsided, and the induration had decreased, all of which were consistent with the breast echography results. One month after completely tapering off the steroid, local recurrence in the right breast was noted. Prednisolone at 40 mg/day (0.8 mg/kg/day) for 1 month was implemented and was tapered off over the course of 4 weeks. The patient was asymptomatic when last seen at our clinic 12 months later.

## DISCUSSION

Granulomatous mastitis is a rare, chronic, non-caseating, granulomatous lobulitis of uncertain etiology. Granulomatous mastitis clinically mimics breast cancer and is frequently mistaken for a malignancy, particularly if the regional lymph nodes are enlarged.<sup>(3)</sup> Thus, failure to diagnose it may result in unnecessary mastectomies.<sup>(2)</sup>

The lesions of IGM are usually unilateral and can occur in any of the 4 quadrants. The condition has been known to occur in both breasts.<sup>(4)</sup> The clinical findings and mammographic results of IGM are often similar to those of a carcinoma. As a result, a granulomatous reaction in a carcinoma remains a critically important differential diagnosis for exclusion.<sup>(5)</sup> However, most examples of this unusual complex have areas composed of easily identified intraductal or invasive carcinoma.<sup>(6)</sup> In rare cases, immunohistochemical stains for cytokeratin or smooth muscle actin may be necessary to identify a carcinoma against this background.

Besides mimicking breast carcinoma, other diseases should also be excluded that might cause a granuloma in the breast, such as tuberculosis, syphilis, and histoplasmosis infections, as well as a foreign-body granuloma, vaccination granuloma, mammary duct ectasia, sarcoidosis, Wegener's granulomatosis, giant cell arteritis, and polyarteritis nodosa.<sup>(1,3,7-13)</sup>

Histologically, IGM tissue is predominately composed of inflammatory cells, mainly lymphocytes, associated with epithelioid histiocytes

admixed with Langhan's giant cells. Stains for bacterial identification (such as acid-fast stain, Gram stain, and Warthin-Starry stain) and those for fungal demonstration (for example, PAS and GMS stains) are applied to rule out possible infectious granulomas. Bacterial culture, fungal culture, and mycobacterial culture may be helpful but are much more time-consuming and relatively expensive for exclusion of an infectious granuloma. Hence, the term IGM should be adopted for those lesions which demonstrate no specific etiologic agent.<sup>(3,10,14,15)</sup>

Although its etiology is still unknown, several pathogeneses of granulomatous mastitis have been postulated including autoimmune<sup>(1,5)</sup> and infective processes<sup>(3)</sup> and a local reaction to chemical secretions.<sup>(16)</sup> Associations of IGM with a recent pregnancy, breastfeeding, and oral contraceptives have been reported.<sup>(12,17)</sup>

The treatment of choice for IGM has not yet been established. Prior to 1980, because of the clinical impression of a possible malignancy, surgical excision of the entire lesion was suggested. Complications seem to be related to both the disease process and the surgical procedure and include skin ulceration, abscess formation, fistulae, wound infection, and recurrence.<sup>(1,3,5,13)</sup> Some patients had relapses in the form of chronic mastitis after excisional biopsies.<sup>(5,13,18)</sup> Two decades ago, DeHertogh suggested complete resection or open biopsy with 60 mg/kg/day prednisolone therapy for treatment of granulomatous mastitis.<sup>(19)</sup> Several other reports of using 60 mg/kg/day prednisolone to treat IGM have also been published.<sup>(3,12,13,18,20-22)</sup>

With our patient, in view of her history of gestational diabetes, a conservative dosage of 30 mg/day prednisolone (0.6 mg/kg/day) was initiated with periodic blood sugar monitoring. However, her granulomatous mastitis recurred 11 weeks after the initial treatment. Prednisolone at 40 mg/day (0.8 mg/kg/day) for 4 weeks was then administered with a successful result. Her blood sugar was controlled well through her diet.

Herein, we report a case of idiopathic granulomatous mastitis successfully treated with 40 mg/day (0.8 mg/kg/day) prednisolone. As the administration of a steroid can cause some severe adverse effects, such as opportunistic infections, dermatological manifestations, hypertension, peptic ulcer disease, neuropsychiatric symptoms, hyperglycemia, myopa-

thy, and osteoporosis, we feel that the minimum dose of a steroid should be recommended. The duration and recommended dosage of steroid treatment deserve further clinical research to establish an adequate treatment regimen.

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## 非特異性肉芽腫乳腺炎：使用最小劑量類固醇治療成功之病例

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非特異性肉芽腫乳腺炎為一良性，非酪狀化的乳房疾病，但經常被誤認為乳癌。它常發生於生育年齡的婦女。非特異性肉芽腫乳腺炎的治療方式常有爭議。在過去，因誤認為有乳癌的可能性，致常被建議施行外科手術予以治療。但近期外國文獻報告指出，使用類固醇治療該非特異性肉芽腫乳腺炎是有效的。因此，我們提出一女性病患成功使用每天每公斤0.8毫克劑量的類固醇予以治療非特異性肉芽腫乳腺炎之報告。(長庚醫誌 2005;28:431-5)

**關鍵字：**肉芽腫乳腺炎，類固醇，乳房疾病。

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