Conservative Management for Idiopathic Granulomatous Mastitis Mimicking Carcinoma: Case Reports and Literature Review

Vijay Naraynsingh, Seetharaman Hariharan*, Dilip Dan, Patrick Harmarayan and Surujpaul Teelucksingh
Faculty of Medical Sciences, The University of the West Indies, St. Augustine, Trinidad, West Indies

Abstract. Idiopathic granulomatous mastitis (IGM) is a relatively rare condition and may commonly mimic carcinoma. Clinically and radiologically, these lesions could be misdiagnosed as carcinoma and histopathology is the most definitive method of diagnosis and to differentiate it from other granulomatous conditions such as sarcoidosis and Wegener’s granulomatosis. We report two cases of IGM presenting with ill-defined indurated mass, peau d’orange and ulcerative lesions clinically mimicking carcinoma. They were successfully treated with corticosteroids. Even abscesses can be aspirated and avoid surgery. A high index of suspicion is necessary to diagnose this condition to prevent unnecessary mastectomy.

Keywords: Idiopathic granulomatous mastitis, lesions mimicking breast cancer, conservative management

INTRODUCTION

Although IGM has been recently reported in the literature, it is still considered to be a relatively rare condition [3]. These lesions commonly mimic malignancy and if not diagnosed preoperatively, may lead to unnecessary breast surgery [9,10]. IGM most often resolves with medical therapy alone; hence, surgeons should be aware of this differential diagnosis [2]. We report 2 cases of IGM suspected to be malignant early during the course of presentation; however, histopathology revealed the lesions to be benign.

CASE REPORT

Case 1

An otherwise healthy 33-year old pre-menopausal female para-2 presented with a 4-week history of swelling, pain and tenderness in her right breast. She had developed ulcerating lesions, peau d’orange and tethering. Her last breast feeding was four years ago and she never used hormonal contraception. On examination, the breast was grossly enlarged and firm on palpation with an indurated mass measuring 13 × 10 cm. There were three punctuate ulcerations oozing seropurulent material when squeezed. Three firm axillary lymph nodes were palpated. The initial gross appearance of the mass is shown in Fig. 1a. Mantoux testing for tuberculosis (TB) was negative. The patient’s blood count and biochemical parameters were normal. Antineutrophil cytoplasmic antibodies and antinuclear antibodies were also negative. Histopathology of an incisional biopsy showed granulomatous mastitis. Special stains for fungi and Zeil-Neilsen for tuberculosis were negative. There were no naked granulomas of sarcoidosis or the vasculitis of Wegener’s granulomatosis. The typical granulomas with lymphocytic cellular infiltrate of IGM were evident.

She was treated with prednisolone 20 mg q8h for 8 weeks. Her ulcerative lesions healed at 3 months
Fig. 1. (a) Gross appearance of granulomatous mastitis (Case 1); (b) Healed lesions after medical management (Case 1).

Case 2

A 37 year old premenopausal female para-2 who breast fed both children presented with swelling of her left breast and mild pain. The skin had, in some areas, become erythematous and developed ulceration with purulent oozing. There was marked induration throughout the breast with ulceration and peau d’orange. There was moderate axillary lymphadenopathy. She had no history of hormonal contraception and had stopped breast feeding eight [8] years before. Mantoux test for TB was negative. Biopsy showed only inflammatory tissue with no evidence of neoplasia and she was treated empirically with trimethoprim-sulphamethoxazole for 3 weeks. The ulcers healed and the swelling started to regress. Six weeks later, she again presented with indurated mass on the opposite breast. This also had punctuate ulcerating areas oozing seropurulent material; the lesions were grossly similar to what she had in her left breast earlier (Fig. 2a). Ultrasound showed an abscess and aspiration of the cavity yielded 15 ml of thick brownish yellow pus [Fig. 2a (Insert)]; however microbiological culture was negative for bacterial and fungal growth. Biopsy confirmed IGM. Special stains for fungi and TB were negative. She was treated with amoxicillin for five [5] days and prednisolone 20 mg q8h for six [6] weeks (Fig. 2b). The ulcerative lesions healed, the induration settled and she has had no recurrence at three [3] years in either breast.

DISCUSSION

IGM is considered a relatively rare disease [3]. This condition was first termed as granulomatous lobular mastitis by Going et al. in 1987 after a lobule-centered distribution of the breast lesion was noted on histological examination [8]. Tuberculosis is still an aetiology to be considered for any granulomatous lesions of the breast especially in migrant populations from countries where the disease is prevalent [24]. A large survey in Pakistan reported that of 3768 breast cases, granulomatous lesions were found in 0.37% of patients which included tuberculous lesions [1]. Tuberculosis is still prevalent in Trinidad but this aetiology was ruled out in both our patients by Mantoux testing as well as by microbiological culture for tuberculous acid fast bacilli (AFB).

Although the exact aetiology of IGM is unclear, the disease has been shown to be correlated with breast-feeding and the use of oral contraceptives [6]. There may also be an association of erythema nodosum in these patients [8]. Both our patients did breast feed, but ‘use of pill’ and ‘erythema nodosum’ were excluded in both of them.

IGM manifests with a variety of presentations [5]. These include an ill-defined mass mimicking carcinoma, which is considered to be the commonest presentation [5]. Signs of inflammation such as pain, redness, and peau d’orange, may more likely point to an inflammatory process [5]. Axillary lymph node enlargement may also be seen infrequently. Our patients presented with all the aforementioned clinical signs which mimicked carcinoma. There have been many published reports of this lesion masquerading as malignancy and even resulting in mastectomy [16,17,26].

The other major clinical feature associated with IGM is that it has been very difficult to clinch the diagnosis during the initial presentation [12]. Although many imaging techniques such as mammography, ultrasound, color Doppler ultrasound, non-enhanced MRI, and dynamic MRI have been suggested for the diagnosis, the features are quite non-specific for the lesion [14,20].
On mammography, asymmetrical focal densities with no distinct margins, ill-defined masses with spiculated contours, and bilateral multiple ill-defined nodules could be seen [20]. On ultrasound, a discrete heterogeneous, hypoechoic mass, or multiple abscesses, or multiple central hypo peripheral hyperechoic lesions, heterogeneous hypo- and hyperechoic areas together with parenchymal distortion, and irregular hypoechoic masses with tubular extensions and abscess cavities could be seen [20]. Colour Doppler ultrasound could be used for vascular lesions. On MR mammography, the most frequent finding may be focal or diffuse asymmetrical signal intensity changes that may be hypointense on T1W images and hyperintense on T2W images, without significant mass effect with or without nodular lesions [20]. On dynamic contrast-enhanced mammography, mass-like enhancement, ring-like enhancement, and nodular enhancement could be seen [20]. In summary, the spectrum of findings is very wide and hence clinicians cannot rely upon imaging as a definitive diagnostic test.

Both clinically and radiologically, there could often be misdiagnosis of this condition as breast carcinoma [15]. Most clinicians are likely to consider malignancy for such presentations. IGM is usually confirmed only by histology, which is the cornerstone of diagnosis [19]. In our situation, although biopsy confirmed the diagnosis in both patients, the initial lesion in the second patient was reported as a non-specific inflammatory lesion.

The management of IGM is also highly variable and there is still no generally accepted optimal treatment [27]. Some authors have suggested primary management strategies included observation, steroids, partial mastectomy, and mastectomy [28]. Wilson’s report also quoted success rates with each treatment [28]. Thus, for observation it was 56% and for steroids it was 42%. The success rates for partial mastectomy and mastectomy were as high as 79 and 100% respectively [28].

It has been suggested that treatment for this condition should be undertaken as part of a multidisciplinary team including surgeons and physicians with an interest in inflammatory breast disease [25]. However, uncomplicated IGM should not be a clear indication for mastectomy especially due to the psychological trauma associated with this procedure. In fact, a previous report has advised that it is time to avoid unnecessary mastectomies for these patients [4]. Although some authors have recommended surgery as the definitive treatment, our patients had complete recovery with conservative medical management with corticosteroids. Even if an abscess is encountered (as in our second case) aspiration and antibiotics combined with steroids may be adequate. It is likely that the ‘pus’ produced from the granulomatous inflammatory reaction may be sterile, as in our patient; and not need open surgical drainage.

Many other authors have reported good outcomes with corticosteroid therapy [7,11,21]. However, the doses of prednisolone recommended are varying and sometimes rather high for this condition. There has been a report where the patient developed Cushingoid features and mood disturbances due to steroids [27]. We were able to get early improvement and complete resolution using 1–1.2 mg /kg/day and we suggest that this dose may be tried initially; and a higher dose may be used only when there is no satisfactory response. There have been reports of using antimetabolites such as methotrexate and azathioprine sometimes as monotherapy and in some refractory cases [13,22,23].

In summary, IGM is a relatively rare condition and may commonly mimic carcinoma of the breast. Because it can evade preoperative diagnosis by routine imaging studies, a very high index of suspicion is crucial to contemplate this diagnosis which must be confirmed histologically to avoid unnecessary surgery.
References


