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ORIGINAL PAPER

# Delayed breast reconstruction in idiopathic granulomatous mastitis

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## Abstract

**Background** Idiopathic granulomatous mastitis is an exceptionally rare, chronic benign inflammatory disease of the breast, more common in developing countries and infrequently seen bilaterally. The symptoms can mimic many other breast conditions, including inflammatory breast carcinoma, and the diagnosis is one of exclusion. The management includes non-surgical treatments, such as broad spectrum antibiotics, steroids and methotrexate, and surgical options, comprising of wide local excision and even simple mastectomy.

**Methods** The case notes of patients with idiopathic granulomatous mastitis referred to our Breast Unit were reviewed and data was collected on patients demographics, medical and surgical treatments and subsequent outcomes.

**Results** Four patients were treated in our unit, all Caucasian women, three of whom were bilateral cases. Three of the patients were reconstructed using oncoplastic techniques whilst the fourth patient awaits reonstruction.

**Conclusions** Oncoplastic breast reconstructive techniques play a fundamental role in the management of these patients, as they give the surgeon the freedom to perform a wider resection, thus reducing the rate of recurrence, whilst at the same time, knowing that there are reconstructive options to restore any breast aesthetics. As such, the value of a multidisciplinary team cannot be underestimated.

Level of Evidence: Level V, therapeutic study.

**Keywords** Idiopathic granulomatous mastitis · Oncoplastic reconstruction

## Introduction

Idiopathic granulomatous mastitis (IGM), first described by Kessler and Wolloch in 1972 [1], is a rare, benign and chronic granulomatous disease of the breast, of unknown aetiology. It accounts for less than 1% of all the benign pathologies of the breast and is quite distinct from secondary granulomatous mastitis, which can be related to tuberculosis, fungal infections, diabetes mellitus, sarcoidosis, polyangitis and exposure to ruptured breast implants [2]. IGM can mimic inflammatory breast cancer, both clinically and radiologically. Diagnosis is mainly one of exclusion and histopathology forms the basis for the diagnosis of IGM [3].

The optimal treatment strategy for IGM is not well defined. Whilst there are well-prescribed treatment protocols for both breast cancer and infective mastitis, there appears to be no consensus on the management of IGM [4]. The strategies described include conservative management, medical or surgical options or a combination of all the treatments. Serial debridement and wider local excisions, and in occasional cases, mastectomies, have been the mainstay of surgical treatments. However, in recent years, an oncoplastic reconstructive approach has slowly evolved in the management of patients with IGM, yielding disease control as well as maintaining the aesthetics of the patient's breasts [5, 6].

In this paper, we report on the oncoplastic reconstructive techniques used in the management of patients with IGM treated at our breast unit and report on their outcomes. Our early experience of this rare disease in

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four cases has allowed us to recommend a simple treatment protocol.

## Materials and methods

The case notes of patients with IGM referred to the Breast Unit were reviewed, and data was collected on patient demographics, medical and surgical treatments and subsequent outcomes.

## Results

Four female patients were identified, with a mean age of 47.5 years (26–62 years) (Table 1). The patients were all white Caucasian, with three patients being non-smokers. None of the patients were pregnant at the time of disease presentation. Three patients had bilateral disease, and none suffered from erythematous nodosum or arthritis.

The commonest presentation was a painful inflammatory breast mass, with one patient presenting with a history of multiple breast abscesses. The duration of symptoms ranged between 6 months and 20 years, and all four patients underwent a full medical examination and the appropriate imaging and tissue biopsies were performed. These included magnetic resonance imaging (MRI), ultrasound, mammography, microbiology sampling and core biopsy for tissue diagnosis. MRI showed multiple ring enhancing lesions in the breast and reactive hyperaemia (Fig. 1), ultrasound revealed abscesses and inflammation of tissues (Fig. 2), whilst mammography initially revealed lesions suspicious of carcinoma (Fig. 3). However, histology from the core biopsy in all cases confirmed the

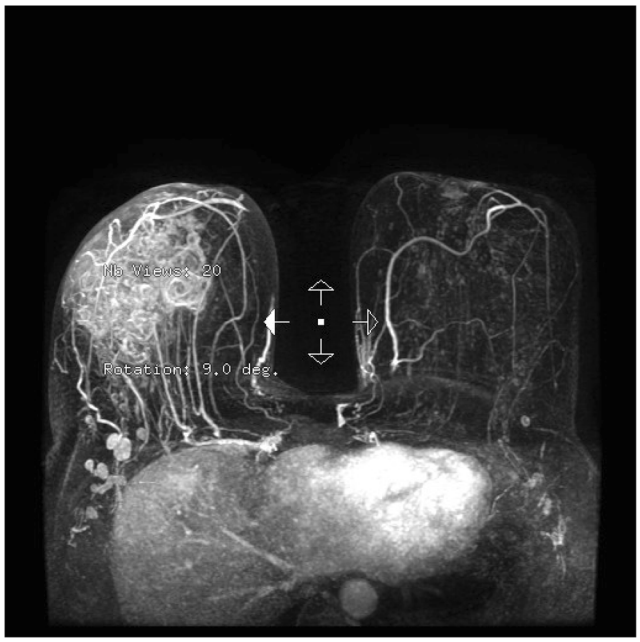



**Fig. 1** MRI images show ring enhancing lesions in breast tissues and inflammatory hyperaemia

**Table 1** Patients with IGM managed at the breast unit

| Patient | Age | Breast affected | Presentation | Medical treatment | Surgical treatment | Delayed reconstruction | Follow-up |
|---|---|---|---|---|---|---|---|
| 1 | 49 | Left | Inflammatory mass | Multiple courses of antibiotics<br>Oral prednisolone | Multiple I&D<br>Left mastectomy | Awaiting weight loss/smoking cessation | 2 years |
| 2 | 62 | Bilateral | Bilateral inflammatory masses | Multiple courses of antibiotics<br>Oral prednisolone | Bilateral central wide local excisions | Retro-glandular implants<br>Nipple reconstruction and areolar tattooing | 2.5 years |
| 3 | 52 | Bilateral | Inflammatory mass on the left, followed by mass on the right | Multiple courses of antibiotics<br>Oral prednisolone | Staged bilateral mastectomies | Initial failed expander/ADM on left side<br>Bilateral LD flaps and implants | 4 years |
| 4 | 26 | Bilateral | Multiple abscesses on the right, followed by same on the left | Multiple courses of antibiotics | Multiple I&Ds<br>Bilateral Hadfield's procedure<br>Bilateral central wide local excision | Sub-pectoral implants, nipple reconstruction, areolar tattooing<br>Larger implants and lateral capsulorraphy | 1.5 years |

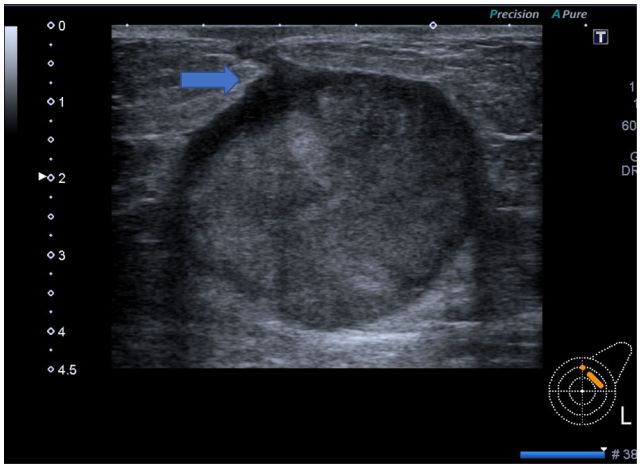



**Fig. 2** Ultrasound scan of breast abscess with sinus to skin

presence of chronic inflammatory cells, associated with a chronic granulomatous reaction, excluding the presence of carcinoma (Fig. 4). The specimens also stained negatively for acid fast bacilli (AAFB) and fungi. All the patients were initially treated with broad spectrum antibiotics, and all required multiple courses of antibiotics. Two of the four patients required multiple incision and drainage (I&D) of breast abscesses. Analysis of all microbiology specimens at presentation failed to yield any growth. In all four cases, their general practitioners had already started them on broad spectrum antibiotics, and it is most

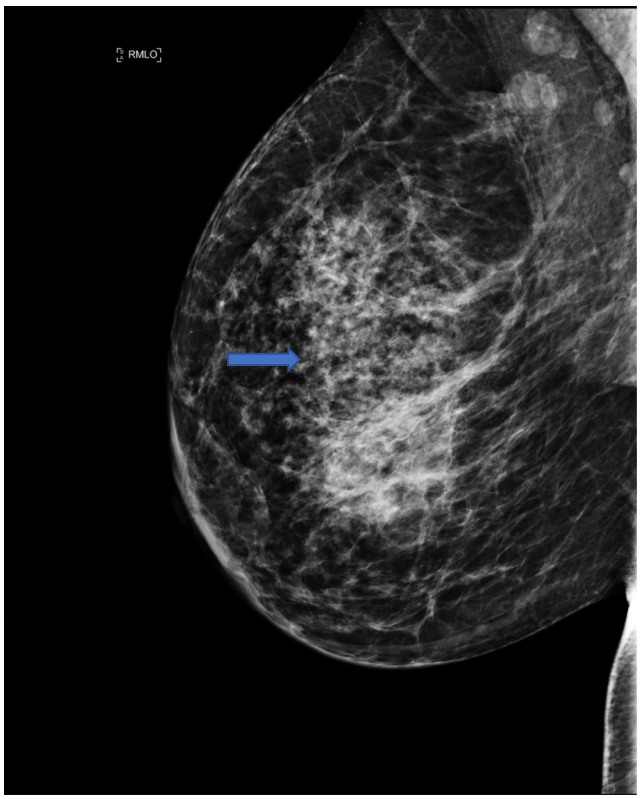



**Fig. 3** Mammography with suspicious areas

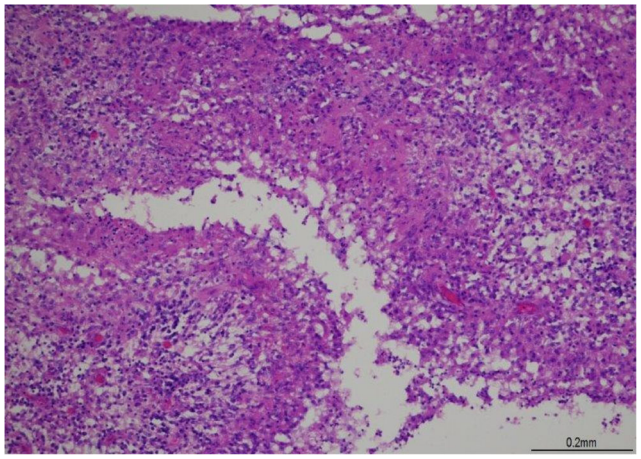



**Fig. 4** Histology slide showing ×10 magnification of granulomatous response with giant cells

likely that we were dealing with sterile abscesses, hence the no growth results. None of the reconstructive procedures were performed until the disease had been eradicated by either wider local excision or mastectomy. Our departmental policy on all reconstructive procedures is IV Augmentin on induction and for 24 h IV and then oral for 7 days; this applies to any breast reconstructions. If allergic to penicillin, then clindamycin is used.

Resolution of symptoms was not achieved with antibiotics and I&D of the breast abscesses, and hence, all four patients were reviewed in a multidisciplinary team setting. A consultant rheumatologist organised chest radiographs and inspected the skin for any rheumatoid nodules and erythema nodosum to exclude the possibility of secondary granulomatous mastitis or other systemic disease. These examinations were negative. All four patients were offered oral prednisolone. Only three patients were started on oral prednisolone for a period of 6 months; the fourth patient refused the course of steroids. The purpose of the steroids was to reduce the degree of inflammation and assist in the surgical excision. However, the disease did not clear, and all four patients progressed to surgical excision of the involved tissues and subsequent reconstruction (Table 2). A total of 22 oncoplastic procedures were performed (Table 3).

Two of the patients required multiple I&D of the breast abscesses, and two patients required wider local excision of affected breast tissues. Two patients needed mastectomies, one unilateral and one bilateral. One patient underwent a Hadfield's procedure, which is a major breast duct excision,

**Table 2** Surgical procedures to eradicate the disease

| Incision and drainage | Multiple |
|---|---|
| Central wide local excision | 4 |
| Simple mastectomy | 3 |
| Hadfield's procedure | 2 |

**Table 3** Oncoplastic reconstructive procedures used for breast reconstruction

| | |
|---|---|
| Breast implants | 8 |
| Nipple reconstruction | 4 |
| Areolar tattooing | 4 |
| Latissimus dorsi flap | 2 |
| Lateral capsulorraphy | 2 |
| Acellular dermal matrix | 1 |
| Tissue expansion | 1 |
| Scar revision | 1 |
| Awaiting reconstruction | 1 |

as she was initially thought to have severe periductal inflammation, before the diagnosis of IGM was made. The patient who had bilateral mastectomies was reconstructed with bilateral latissimus dorsi myocutaneous flaps (Fig. 5). Two patients were reconstructed with breast implants. One patient has undergone nipple reconstruction and tattooing. As all the diseased areas had been resected prior to any delayed reconstruction, no patient was on steroids at the time of reconstruction and the departmental protocol on the use of antibiotics, as previously described, was adhered to. Other procedures included the use of acellular dermal matrix (ADM) and tissue expansion. ADM was not used during the active phase of the disease, but only after all the diseased areas had been excised. In one patient, the planned immediate implant reconstruction following mastectomy was deferred, as multiple pockets of pus were encountered at the time of the mastectomy.

The mean time between surgical resection of the IGM and reconstruction in the three patients was 7.5 months. The mean follow-up time following surgery was 2.5 years, with all patients being clear of IGM at the follow-up appointment.

## Discussion

IGM is a rare chronic inflammatory disease of the breast that affects mainly women of child-bearing age, is more common in the developing world and is often associated with pregnancy and lactation [7, 8]. In our series, all four patients were Caucasian women, which is not common. The prevalence and incidence of the disease have been reported as 2.4 per 100,000 women and 0.37%, respectively [9, 10], although it is thought that these are much higher [11]. IGM can present as a rapidly increasing breast mass, which may be associated with inflammation, nipple retraction, abscesses and chronic draining sinuses [12], and often may not be different from mycobacterial and fungal infections, or deep soft tissue infections. It can also mimic the presentation of a breast carcinoma, especially when it involves the underlying pectoralis muscle with nipple retraction, the overlying skin and axillary lymphadenopathy [12]. There is a tendency for the condition to recur, and patients with recurrent sterile breast abscesses should have IGM excluded [13]. Diagnosis is mainly one of exclusion and can only confirmed by histopathology [14], which shows the features of a non-caseating granuloma, composed of epithelioid histiocytes with giant cells within and around the lobules [15].

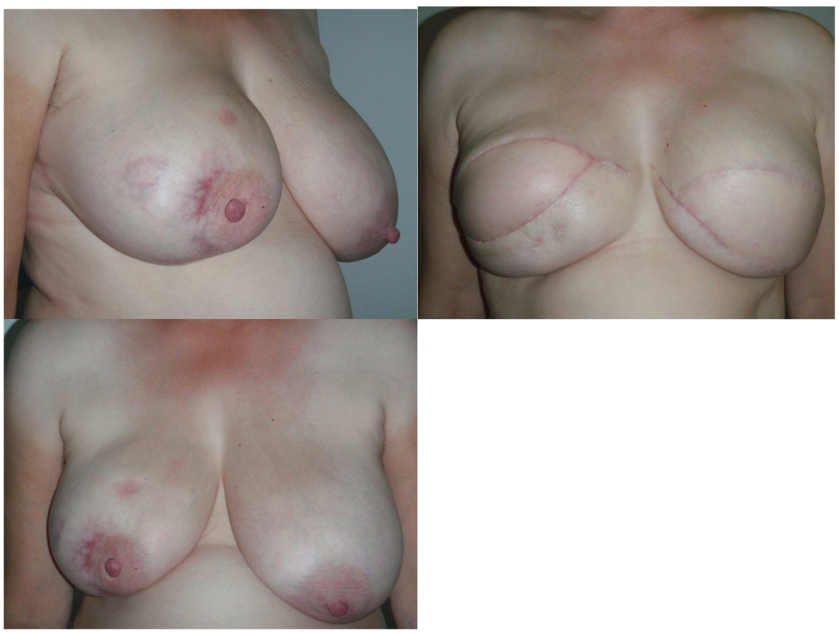


**Fig. 5** Patient with bilateral disease requiring bilateral mastectomies and subsequent reconstruction with bilateral latissimus dorsi flaps and implants

The sequence of events that leads to the development of IGM has been postulated as such: injury to the ductal epithelium, spilling of luminal secretions into the lobular connective tissue, local inflammation to the connective tissues, followed by macrophage and lymphocyte infiltration and a subsequent local granulomatosis inflammatory response [16, 17]. However, the triggering factor causing the ductal epithelial damage is yet to be elucidated [18]. Pregnancy, oral contraceptive use, local trauma to the breast, alpha-1 antitrypsin, smoking and diabetes mellitus are thought to be potential triggering factors [18], although the evidence for smoking and the contraceptive pill as trigger factors has not been conclusive [19, 20]. Nonetheless, smoking, pregnancy and breast infection have been reported to be associated with disease recurrence [21].

Three main hypotheses have been put forward to describe the development of IGM; these include an autoimmune response, infectious disease and hormonal disruption [13]. The autoimmune hypothesis is currently viewed as the basis of IGM. The favourable response of IGM to steroids and immunosuppressants; the presence of extra-mammary disease, such as erythema nodosum and arthritis; and the presence of T lymphocytes have supported the autoimmune basis for the disease [3, 18]. However, the findings of classical serological markers, such as antinuclear antibody (ANA) and rheumatoid factor (RF), seen in autoimmune disorders, such as antinuclear antibody and rheumatoid factor, have not been consistent in various studies [18, 22, 23]. In our series, all four patients were tested for ANA, RF and angiotensin-converting enzyme (ACE), and were noted be negative. In our unit, we have not found the use of serological markers particularly helpful in making the diagnosis of IGM.

Hormonal disturbance in the form of raised levels of prolactin has also been suggested as a potential cause of IGM. It is thought that the hyperprolactinaemia leads to overstimulation of the breast parenchyma, as well as changes similar to the lactation period [24]. It may also affect the severity of the disease and its recurrence rate [18]. However, findings of raised prolactin levels associated with IGM have been inconsistent [3, 7]. Reports of IGM associated with hyperprolactinaemia were either cases of antipsychotic drug-induced or associated with recurrent disease [19, 25]. Moreover, no association between smoking and the use of the contraceptive pill and hyperprolactinaemia has been demonstrated [18]. Given the inconsistent association of hyperprolactinaemia with IGM, its relevance is still debatable.

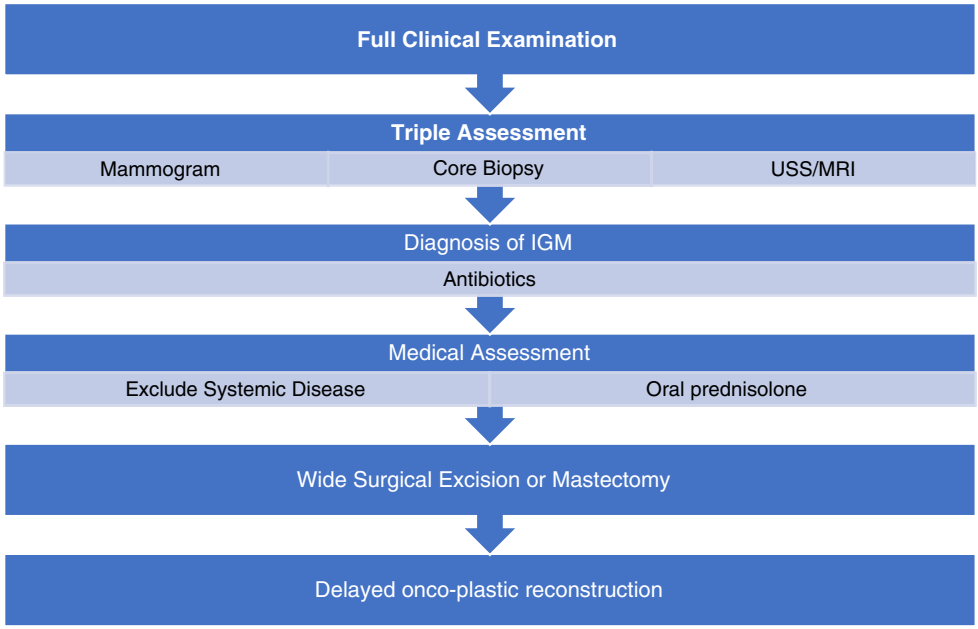
Infections have also been suggested to be a cause of IGM, especially since a granulomatous reaction to specific agents such as fungi, parasites and bacteria can happen [26]. Various studies have reported on the presence of *Corynebacterium* in patients with IGM [17, 27]. However, the bacterium was not isolated in all cases, bringing into question its true role in the aetiology of the disease. Uysal et al. noted a significant relationship between disease recurrence and systemic bacterial infection [21]. Of note however, the bacterial infection was not the same as when the initial diagnosis of IGM was made. More recently, a study carried out by Yu et al. profiled the microbiota of patients with IGM and noted the presence of Corynebacterium in all 19 patients, and in particular the presence of *C. kroppenstedtii* in 11 of the 19 patients [28].

There is still no agreed protocol for the management of patients with idiopathic granulomatous mastitis. Treatment regimens that have been suggested include conservative treatment, drug therapies, surgery or combination of all the treatments available [12]. Some authors have managed patients with mild features of IGM with either observation, or with antibiotics exclusively, and have reported no recurrences after 15 months of treatment [29, 30]. However, this modality of treatment may not be appropriate for all patients, as although mild, IGM can be painful and can be emotionally distressing for patients.

More recently, the use of steroids, on its own or in conjunction with surgery, have become more prominent. The use of steroids reduces the dimensions of the disease and can help to promote healing after excision of the disease [22, 31]. However, there is still debate regarding the timing on its use and the concentrations to be used. The administration of steroids pre-surgery does reduce the size of the lesion and therefore allows for a more conservative resection [11, 31]. Also, stopping the steroids too early can lead to an increased rate of recurrence of the disease [11, 32]. Nonetheless, the use of steroids is associated with complications such as glucose intolerance, Cushing syndrome, avascular necrosis and potential exacerbation of any infections [33]. In resistant cases or where the use of steroids is contraindicated or can potentially lead to the complications mentioned, the use of immunosuppressants, such as methotrexate, azathioprine, bromocriptine and colchicine, has been shown to be effective [34].

Surgical excision of the affected areas, following incision and drainage of any abscess, is another option for the management of patients with IGM. It allows for definitive diagnosis of the disease and faster recovery [33]. Wider surgical excision has gained further acceptance in recent years, especially since limited excision was typically associated with increased rates of recurrence [3, 14]. Wider excision and, in some advanced cases, mastectomy should be surgical options that are discussed with patients. These options would reduce the time required for healing and also reduce the number of operations required for complete resolution of the disease. Whilst more extensive surgery affects the shape of the breast, the availability of more advanced reconstructive techniques, ranging from implant-based techniques to the use of local or free tissue

**Table 4** Management protocol used at the local breast unit




transfer, has made the restoration of the breast aesthetics a more achievable result [4–6, 35]. It further emphasises the necessity of managing these patients within a multidisciplinary team, where both breast and plastic surgeons are able to plan the excision and reconstruction of the breast. It is also crucial to involve all the members of a breast unit to be involved so that patients are given the appropriate psychological support as required. In our series, all four patients were managed in our local breast unit with input from both a plastic and a breast surgeon. All patients underwent wide local excision or mastectomies following the initial incision and drainage of abscesses. A delayed oncoplastic reconstructive approach was adopted, and reconstructions were either implant based or flap and implant based. No recurrence of the disease was noted at a mean of 2.5 years following reconstruction.

Our breast unit's experience in managing these four patients have allowed for the development of a simple treatment protocol that has been successful so far (Table 4). Patients undergo a complete medical assessment and are triple assessed to exclude to possibility of carcinoma. Once a diagnosis of IGM is made, the patients are started on the appropriate antibiotics. They are also assessed by a medical specialist to exclude the possibility of any systemic disease and are started on oral prednisolone. Any breast abscess noted is drained. Resection and surgical reconstruction are discussed at the multidisciplinary meeting. The patients would then either undergo wider local excision or mastectomy, depending on the severity of the disease. Delayed oncoplastic reconstruction is only carried when there is no evidence of recurrence of the disease.

## Conclusion

Idiopathic granulomatous mastitis is an extremely challenging problem to manage. Confirmation of the diagnosis can be difficult. A lack of consensus on the aetiology of the pathology and subsequent development of any treatment pathway has not been forthcoming. However, current evidence points towards a combined use of steroids and wide surgical excision of the diseased area, followed by reconstruction as appropriate. In our series, all the patients were of Caucasian background, and three of the patients had bilateral disease, which is unusual. Also, three patients were reconstructed using oncoplastic techniques, whilst the fourth patient awaits reconstruction. There is paucity in the literature describing the use of such an approach to the management of patients with IGM, and our series addresses this issue. We feel that the use of oncoplastic breast reconstructive techniques plays a fundamental role in the management of patients afflicted with IGM, as it gives the surgeon the freedom to perform a wider resection, thus reducing the rate of recurrence, whilst at the same time, knowing that there are reconstructive options to restore any breast aesthetics. As such, the value of a multidisciplinary team cannot be underestimated.

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## Compliance with ethical standards

**Funding** No funding was available for this review.

**Conflict of interest** Author A and Author B declare that they have no conflict of interest with any person and/or organisations that can bias the outcome of the work submitted.

**Ethical approval** The study was registered with the research department.



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