CASE REPORT

Idiopathic Granulomatous Mastitis: Surgery, Treatment, and Reconstruction

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Abstract: Idiopathic granulomatous mastitis is a rare inflammatory breast disease that mimics breast diseases such as inflammatory breast carcinoma, infective mastitis, and inflammatory breast disease of known cause. It is a diagnosis made only after excluding other causes, and although the disease is nonmalignant it may be both locally aggressive and recurrent. Definitive treatment may require radical excision and adjunctive treatment with immunosuppressants. Reconstruction following excision of disease has not been previously described. In those patients who undergo reconstruction, both the surgeon and the patient must be aware of complications associated with residual disease, and the potential involvement of donor sites. Treatment should be undertaken as part of a multidisciplinary team including surgeons and physicians with an interest in inflammatory breast disease. We present two patients diagnosed with idiopathic granulomatous mastitis who were referred to our unit for consideration of reconstruction. One patient underwent autologous breast reconstruction and the other contra lateral surgery to achieve symmetry.

Key Words: granulomatous mastitis, idiopathic, inflammatory breast disease, reconstruction

Idiopathic granulomatous mastitis (IGM) is a rare breast disease of unknown etiology, which can mimic breast cancer, both clinically and radiologically. It may also be difficult to distinguish from infective mastitis and inflammatory breast disease of known etiology. It is a diagnosis of exclusion and histology is the mainstay of diagnosis with characteristic features including granulomatous inflammation of the breast lobules and microabscess formation (1). Although treatment protocols are well established for both breast cancer and infective mastitis, there are no such pathways established for treatment of this condition. Nonetheless, wide local excision with or without steroid therapy is most commonly advocated (2). The disease has a high rate of recurrence and persistence even after surgery (2). In addition, there is little evidence regarding the role and the timing of breast reconstruction in these patients. We present two patients with the above condition who were referred to our plastic surgery department. We describe the presenting features and investigations to establish diagnosis of this condition, and propose options for surgical reconstruction.

PATIENTS AND MATERIALS

Two patients were referred to our multidisciplinary breast team with inflammatory breast disease; one for definitive treatment of disease and the other for reconstruction following previous bilateral mastectomy.

CASE REPORTS

Case 1

A 46-year-old nonsmoker patient presented to the Accident and Emergency department with a history of malaise and left breast pain was treated with oral antibiotics having undergone an unsuccessful aspiration for presumed infective mastitis.

She was referred to the local surgical team with a left breast abscess and superficial skin necrosis. She had no history of previous breast disease or other inflammatory conditions and was on no medication at the time of presentation. She had undergone bilateral silicone breast implantation 18 years before presentation, and had undergone nine closed capsulotomies to treat a left capsular contracture.
She was admitted with left breast cellulitis and received a course of intravenous antibiotics for presumed infection. The skin over the lower pole of the breast deteriorated with leakage of pus and blood stained fluid through the previous aspiration site. No organism was isolated. The Initial clinical impression was that of an atypical infection, with suspicion of underlying breast carcinoma. Ultrasound showed a left breast abscess with bilateral capsular contractures but no evidence of implant leakage. The patient was subsequently referred to our care. At the time of review the left breast was indurated and red with ulceration and pus leakage from the lower pole (Fig. 1). A planned surgical exploration was undertaken and revealed an intact implant and grossly indurated breast tissue. The implant was removed and wide excision of indurated breast tissue was performed. Histology showed idiopathic granulomatous mastitis with no evidence of silicone leakage, carcinoma, fat necrosis, or infective organisms. The patient made a good recovery and wounds healed satisfactory in the postoperative period (Fig. 2). No adjunctive treatment was given. Following discussion with the patient, the right breast implant was also removed 6 months later and bilateral Benelli mastopexy was performed to achieve symmetry (Fig. 3). No recurrence of the disease was noted on outpatient review eighteen months following the wide local excision.

Case 2

A 42-year-old Para 3 + 0 woman gave a history of a tick bite to the right breast 3 years previously which had initially healed with topical dressings. She subsequently developed an enlarged right axillary lymph node associated with pain and inflammation around the right nipple (Fig. 4). The lymph node was biopsied and showed only reactive changes. Core biopsy from the nipple was reported as benign. However, she
developed an abscess at the site of the biopsy which required incision and drainage. This area was further complicated by subsequent multiple abscesses. Ultrasound demonstrated a chronic abscess cavity and the patient had a persistent nonhealing wound in the upper pole of the right breast with generalized induration and peau d’orange appearance. All microbiology samples were found to be unremarkable and long-term treatment with multiple antibiotic regimes brought no improvement in her symptoms. The patient finally underwent a right mastectomy 3 years following initial presentation as no definitive diagnosis had been reached. Histology at this point demonstrated idiopathic granulomatous mastitis. Contralateral recurrence of the disease developed a year later, and a left mastectomy was performed again confirming the diagnosis of IGM. No adjunctive treatments had been given.

The patient was referred to our care for consideration of reconstruction 2 years following the second mastectomy. At this point there was no evidence of active disease and a bilateral autologous latissimus dorsi breast reconstruction was planned (Fig. 5). Multiple granulomatous deposits were noted within the chest wall intraoperatively and excised, followed by completion of the reconstruction. The initial recovery was uneventful, however the patient continued to experience chronic seroma formation in both her latissimus dorsi donor sites 2 years from the time of reconstruction. These were treated with local seroma cavity steroid injection but the seroma continued to persist. As a final measure the patient underwent excision of a chronic seroma capsule with quilting of the donor site, which eventually resolved her donor symptoms. Histology of the seroma capsule demonstrated chronic inflammatory changes without any evidence of granulomatous deposits. The reconstruction was completed with bilateral nipple reconstruction and nipple areolar complex tattooing, resulting in a satisfactory outcome (Fig. 6).

**DISCUSSION**

Idiopathic granulomatous mastitis is a rare inflammatory breast disease first described by Kessler and Wolloch in 1972 (3). Histology shows chronic granulomatous lobulitis in the absence of an obvious etiology although some postulated causes include an autoimmune response, undetected organisms, and oral contraceptives (4–6). It is usually a unilateral condition that affects women of childbearing age and is frequently associated with pregnancy and lactation (4,7). IGM can present with clinical features including a breast mass, inflammation, galactorrhea, and ulceration of the overlying skin, hence raising the suspicion of breast carcinoma. Radiologic findings may often be misleading with characteristics mimicking cancer. Mammography may demonstrate microcalcifications and spiculate calcifications, while ultrasound may show areas of hypoechoegenecity and nodularity (8). Due to a high incidence of false-positive and false-negative findings when used to diagnose granulomatous mastitis, mammography, and ultrasonography, should be used to exclude malignancy rather than to confirm the diagnosis of IGM.

The diagnosis of IGM is one of exclusion where atypical infection and breast carcinoma are among the possible differentials. Histology in the form of core and excision biopsy is the mainstay of diagnosis. Findings include noncaseating granulomas, necrosis, giant

Figure 5. Preoperative planning for a bilateral latissimus dorsi reconstruction of mastectomy sites.

Figure 6. Completed bilateral breast reconstructions with the autologous latissimus dorsi flaps and nipple areolar complex tattooing.
cell formation, and background neutrophils (9). Other systemic granulomatous conditions such as sarcoidosis, Wegener’s granulomatosis or tuberculosis must be excluded, and a negative microbiology with standard and special stains has to be demonstrated (1). Neither patient in our report had any evidence of systemic disease following extensive investigations. Given the history in both patients of local trauma, in one as a consequence of multiple closed capsulotomies and in the other a given history of an insect bite, the possibility of fat necrosis or foreign antigen as a trigger for the disease must be considered, although we do not believe this has been described in previous literature. Alternatively, the history of local trauma in both cases may be coincidental.

There is currently no standard treatment for idiopathic granulomatous mastitis. The recommended management in some of the recent literature is complete surgical excision of the affected area with possibility of adjuvant therapy in cases of recurrence or delayed wound healing (2,4). Steroids have been found in some recurrent cases to reduce the size of the lesion as well as enhancing the postoperative healing process (1). The protracted disease process in the second case might well have benefited from earlier steroid treatment following the histologic confirmation of the diagnosis.

Exclusion of an infective etiology is essential before commencing steroid therapy given its potential for precipitating a flare-up of infective diseases. In addition, we feel that in cases where bacterial superinfection is a possibility, empirical antibiotic therapy should be initiated and sustained until a definitive diagnosis is reached.

Idiopathic granulomatous mastitis is also a disease of secondary complications such as infection, sinus formation, and delayed wound healing as experienced in the cases illustrated above. Case 2 had the challenging problem of chronic seroma formation which may have been related to her underlying diagnosis, and which appeared to be unresponsive to local steroid therapy. Recurrence rates following surgery as high as 50% have been reported (2). It has been suggested that for reasons unknown, this condition can remain dormant for a long period before recurrence at a much later date from initial presentation (2). Thus, it is perhaps important to maintain long-term follow-up for patients with chronic granulomatous mastitis. However, no measure for prevention of the disease has been devised.

Due to the possibility of high recurrence of IGM, surgical management poses a dilemma both to the surgeon and patient in that extensive resection of breast tissue may be required to ensure a disease-free zone. Wide local excision or mastectomy in the absence of malignant disease is a challenging decision. However, involvement of oncoplastic procedures can provide options of reconstruction in conjunction with the initial surgical management and help to reduce the otherwise potentially poor cosmetic outcome, and address the psychological impact on patients. Avoidance of implant-based methods of reconstruction following resection, and focus on autologous techniques is advisable. Despite lack of evidence to relate silicone to aggravation of IGM, it would seem sensible to avoid foreign material in the presence of granulomatous disease, as foreign-body reactions and granulomatous disease have biologic similarities.

Well-established techniques of autologous breast reconstruction such as the autologous latissimus dorsi flap can provide a safe and effective option with high patient satisfaction (10). In addition, techniques for wide local excision and breast reshaping are being increasingly utilized with safe and satisfactory results (10,11). Given the high rate of associated complication and recurrence, reconstruction should be undertaken as a delayed procedure to avoid potentially extensive surgery in the presence of florid inflammation, and to allow completion of adjunctive treatment with steroids or other immunosuppressants, if needed.

**CONCLUSIONS**

Idiopathic granulomatous mastitis is a rare and challenging condition. It is a diagnosis of exclusion with histology as its main diagnostic tool. It should be considered as a possible differential diagnosis for inflammatory breast disease when carcinoma, infection, and systemic disease have been excluded. Surgical intervention in the form of wide excision is necessary and there is place for adjuvant treatment in selected and recurrent cases. Reconstruction following initial surgery should be performed as a delayed procedure. Adjunctive treatment with steroids or immunosuppressants should be considered in cases of extensive disease. Although this may minimize residual disease, recurrence is possible, and may involve adjacent tissues, and regional donor sites. The diagnosis and treatment remain challenging and are best treated within a multidisciplinary setting.
REFERENCES

1. Erhan Y, Veral A, Kara E, et al. A clinicopathologic study of a rare clinical entity mimicking breast carcinoma: idiopathic granulomatous mastitis. *Breast* 2000;9:52–56.

2. Azlina AF, Ariza Z, Arni T, Hisham AN. Chronic granulomatous mastitis: diagnostic and therapeutic considerations. *World J Surg* 2003;27:515–18.

3. Kessler E, Wolloch Y. Granulomatous mastitis: a lesion clinically simulating carcinoma. *Am J Clin Pathol* 1972;58:642–46.

4. Bani-Hani KE, Yaghan RJ, Matalka II, Shatnawi NJ. Idiopathic granulomatous mastitis: time to avoid unnecessary mastectomies. *Breast J* 2004;10:318–22.

5. Cserni G, Szajki K. Granulomatous lobular mastitis following drug-induced galactorrhea and blunt trauma. *Breast J* 1999;5:398–403.

6. Brown KL, Tang PH. Postlactational tumoral granulomatous mastitis: a localized immune phenomenon. *Am J Surg* 1979;138:326–29.

7. Going JJ, Anderson TJ, Wilkinson S, Chetty U. Granulomatous lobular mastitis. *J Clin Pathol* 1987;40:535–40.

8. Memis A, Bilgen I, Ustun EE, et al. Granulomatous mastitis: imaging findings with histopathologic correlation. *Clin Radiol* 2002;57:1001–06.

9. Tse GM, Poon CS, Law BK, et al. Fine needle aspiration cytology of granulomatous mastitis. *J Clin Pathol* 2003;56:519–21.

10. Delay E, Gounot N, Bouillot A, Zlatoff P, Rivoire M. Autologous latissimus breast reconstruction: a 3-year clinical experience with 100 patients. *Plast Reconstr Surg* 1998;102:1461–78.

11. Asgeirsson KS, Rasheed T, McCulley SJ, Macmillan RD. Oncological and cosmetic outcomes of oncoplastic breast conserving surgery. *Eur J Surg Oncol* 2005;31:817–23.