CASE REPORT

Idiopathic granulomatous mastitis associated with hyperprolactinemia: A nonoperative approach

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Abstract
There is increasing evidence associating idiopathic granulomatous mastitis (IGM) with hyperprolactinemia. All documented cases have involved the patient having at least one operative procedure before the association has been made. We present a 55 year old female with IGM associated with risperidone induced hyperprolactinemia. She was successfully treated with a dopamine agonist, bromocriptine. We demonstrated that complete resolution can be achieved without surgical intervention, by targeting serum prolactin levels. We hope this will increase awareness of this rare clinically entity and avoid potentially unnecessary surgery.

KEYWORDS
bromocriptine, hyperprolactinemia, idiopathic granulomatous mastitis, non-surgical

1 INTRODUCTION

Idiopathic granulomatous mastitis (IGM) is a rare clinical entity involving chronic inflammation of the breast without any associated infection, trauma, or exogenous material. Most cases of IGM are associated with a recent history of pregnancy and breast feeding, coinciding with when hyperprolactinemia is most commonly seen. The prevalence of hyperprolactinemia in the general population is less than one percent, however, it has been reported as high as 88% with those on long term risperidone. It usually causes menstrual cycle dysfunction, decreased bone density and galactorrhea, however, its role in inflammatory breast conditions are increasingly recognized. Due to the rarity and lack of awareness, prolactin levels are often not tested, and if so only on recurrence or lack of clinical progress. As such, to our knowledge, all documented cases of IGM associated with hyperprolactinemia have involved the patient having at least one operation before the diagnosis and association was establish. This is largely due to the uncertainty and similarities with malignancy in a nonresolving lesion. There have been few cases of recurrences that have been successfully managed with therapies aimed at normalizing prolactin levels. To our knowledge, we present the first successful case of nonsurgically treated IGM associated with hyperprolactinemia with a dopamine agonist, bromocriptine.

2 PRESENTING CONCERNS

A 55 year old Caucasian female presented to our surgical outpatient clinic with a 1 month history of swelling and pain in her left breast. Mammogram and Ultrasound of her breast were suspicious for an inflammatory carcinoma (Figure 1). Subsequent core biopsies of the lesions revealed focal inflammation with no evidence of malignancy. Her medical history included depression and schizoaffective disorder which had been successfully treated with risperidone for a number of years. Her medical and surgical history was otherwise unremarkable. She went through menopause at age 53, was nulliparous, with no history of oral contraceptive use or hormone replacement therapy and no personal or family history of breast cancer.

On examination she had a large mass with erythema and suppurating over the lateral aspect of her left breast. She had bilateral nipple inversion which was long-standing. There was no lymphadenopathy or other clinical signs on physical examination. The presumptive diagnosis based on the physical exam and biopsy was infective duct ectasia and she was trialled on oral antibiotics, Amoxycillin and Clavulanic Acid.

After 2 weeks of antibiotic therapy, there was no improvement. The overlying skin had broken down and was suppurating (Figure 2A). A further biopsy was conducted and confirmed suspected IGM (Figure 3). A literature review on IGM revealed...
increasing evidence that hyperprolactinaemia may play a pathophysiological role. Given the patient’s history of risperidone use and its association with hyperprolactinemia, a serum prolactin level was ordered, which was 935 (58-416). She was then trialled on a dopamine agonist, bromocriptine 2.5 mg twice daily. The patient showed interval improvements to full resolution within a month of therapy (Figure 2B). After a year of remission, she was trialled off bromocriptine with no signs of recurrence a year and a half of follow-up postcessation.

Sections of the breast biopsy showed replacement of breast tissue by a diffuse inflammatory infiltrate of histiocytes and lymphocytes with some epithelioid multinucleated giant histiocytes. Lipid vacuoles characterized by clear spaces were present. Features are consistent with IGM.

**FIGURE 1** Initial mammogram of left breast: left Cranio-caudal view

Idiopathic granulomatous mastitis clinically and radiologically simulates breast malignancy. Biopsies are often reported as chronic inflammation without evidence of malignancy, however due to a lack of awareness, a specific diagnosis of IGM may not be made. Patients are then often trialled on antibiotics or corticosteroids. With a lack of response, many clinicians would proceed to an excisional biopsy or mastectomy, in fear of missing an occult malignancy.3,6,7 With a long history of risperidone use we suspected that our patient may have IGM, which was confirmed on repeat core biopsy. Lin recently published the first case of risperidone induced hyperprolactinemia related IGM which shared a similar clinical course as our patient. However, their diagnosis was made on formal histology after their patient was scheduled for a simple mastectomy. Attention was then brought to her long term use of risperidone. Risperidone has been known to cause hyperprolactinemia.3 However, until recently the role of hyperprolactinemia in IGM has not been well described.

**FIGURE 2** (A) Before treatment. (B) After treatment [Color figure can be viewed at wileyonlinelibrary.com]

**FIGURE 3** Histology slide from repeat core biopsy [Color figure can be viewed at wileyonlinelibrary.com]
Rowe was the first to describe a patient with IGM associated with hyperprolactinemia. Their patient had recurrent breast abscesses despite having multiple incision and drainages. The diagnosis of IGM was only made after she had surgical excision of 75% of her right breast. Her hormonal profile was suggestive of a pituitary prolactinoma and she was put on bromocriptine and did not have any further breast abscesses. Cserni also described a patient with IGM that was likely associated with hyperprolactinemia. In addition to this, they also suggested that trauma may have a role in IGM. More importantly, they suggested the multiple operations that patients whom have not been correctly diagnosed with IGM often have, could correspond to trauma and hence advised against recurrent surgical intervention. The case we present adds to the limited cases reports associated hyperprolactinemia with IGM. More importantly, we demonstrated that complete resolution can be achieved without surgical intervention, by targeting serum prolactin levels. We hope this will increase awareness of this rare clinically entity and avoid potentially unnecessary surgery.

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