Case Report

Massive Bilateral Gestational Gigantomastia Mimicking Malignancy: A Case Report of a Rare Breast Disorder

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Abstract

Gestational gigantomastia (GG) is a rare benign disorder of the breast usually occurring in the 1st or 2nd trimester of pregnancy of unknown etiology. It causes significant physical and psychological symptoms with occasional complications such as sepsis and haemorrhage. We present a 20-year-old G2P1 + 0 A1, whose LMP was 18 weeks with complaints of progressive bilateral breast enlargement associated with breast, neck, and back pain. She developed ulcers on both breasts spontaneously and subsequently had an episode of massive bleeding from the left breast ulcer necessitating emergency admission and multiple blood transfusions. She had a reduction mammoplasty of both breasts due to her desire to preserve her breast and lack of funds for postmastectomy reconstruction. However, she had intrauterine fetal death at 26 weeks due to severe oligohydramnios. GG could mimic malignancy and become complicated by severe bleeding with possible maternal or fetal mortality. A multidisciplinary approach in the management of the patient is required to achieve the desired treatment outcome.

Keywords: Bleeding, breast, case report, gestational, gigantomastia, mammoplasty

Introduction

Gestational gigantomastia (GG) or macromastia is a rare disorder of the breast during pregnancy whose etiopathogenesis remains unknown, evident by the numerous theories that exist, of which none is universally applicable to all cases.[1] It is a benign disease characterised by diffuse and disproportionate enlargement of the breast often occurring in the 1st or 2nd trimester and may resolve after pregnancy,[2] but has been reported to persist postpartum.[3-5] It is associated with significant physical and psychological symptoms. Some cases of gestational macromastia are complicated with ulcers, sepsis, or severe bleeding, sometimes causing fetal loss and, rarely, maternal death.[1,6-8] The best outcome to this condition has been reported to be surgical in the form of reduction mammoplasty or mastectomy.[5]

We report a case of bilateral GG complicated by massive haemorrhage and fetal demise. This represents the first in our collective experience.

Case Report

A 20-year-old Hausa lady presented to the outpatient clinic with a 16-week history of progressive bilateral breast enlargement with associated breast, neck, and back pain. She also complained of difficulty coping with routine activities and mobility. She was G2P1 + 0 A1 with her last normal menstrual period (LMP) 18 weeks at presentation, and her last confinement was three years before presentation. No history of similar breast enlargement in her previous pregnancy and no other morbidity. Both breasts were markedly enlarged with Peau d'orange. The nipple–areola of both breasts was stretched out. On examination, there was no discrete mass palpable and negative palpable axillary lymph node.

Report of the core needle biopsy of both breasts, done by the general surgery unit before referral, suggested gestational macromastia. Her pregnancy test was positive, and obstetric ultrasound showed a live singleton fetus at 19 weeks + five days.

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However, she developed ulcers on both breasts spontaneously around the nipple–areola region three weeks after presentation, with rapid herniation of breast tissue into the ulcers presenting as a fungating mass [Figure 1], and subsequently had an episode of massive bleeding from the left breast ulcer necessitating emergency admission. Both breasts were grossly enlarged with slight asymmetry, the left larger than the right. The left breast ulcer was about 30 cm × 15 cm containing necrotic tissues, clotted blood, no visible nipple, and an extensive area of skin hyperpigmentation and distended veins. The right breast ulcer was about 15 cm × 10 cm containing necrotic tissues, a flattened nipple–areola complex, and an extensive area of skin hyperpigmentation and distended veins. She had an application of pressure dressings and multiple blood transfusions on account of a packed cell volume of 17%.

Obstetric ultrasound scan showed a single viable fetus at 26 weeks and four days with severe oligohydramnios and no fetal anomaly.

She was counseled for surgery and opted for a reduction mammoplasty due to her desire to preserve her breast, and lack of funds for further reconstruction if a mastectomy was done. She had bilateral reduction mammoplasty with free grafting of the right nipple–areola. The left nipple–areola was necrotic at the time of surgery.

Histological report of the excised breast tissue showed that the right and left breasts measured 40 cm × 30 cm × 15 cm and 36 cm × 25 cm × 10 cm and weighed 6.0 kg and 6.8 kg, respectively. Transection showed gray-white to yellowish tissue surfaces. Microscopically, both breasts were similar, showing lobular hypertrophy, proliferation of the ducts, and periductal fibrosis [Figures 2-4]. These were disposed within a fibrocollagenous stroma.

She had postoperative wound infection, which was managed conservatively with dressings, and loss of the left breast nipple–areola graft. She was planned for amnioinfusion, but had intrauterine fetal death before it could be done. She was discharged 10 days after surgery [Figure 5].

Figure 1: A 20-year-old G2P1 + 0 A1 with bilateral breast enlargement and septic ulcer

Discussion

GG is a benign disease of the breast occurring during pregnancy, characterised by diffuse and disproportionate enlargement of the breast. The incidence of GG varies from 1/28,000 to 1/100,000 pregnancies.[5] Most reports are from North America and Europe.[9] Less than 10 cases have been reported from Africa according to a recent report by Abdullahi.[6] However, it is possible that most African patients do not seek medical care due to poverty as well as low health coverage in the region. Like the index case, most cases occur in women in their 2nd or 3rd decade.[2,5-7,9] Majority of cases of GG start in the first or second trimester and are often bilateral.[6,10] Rutherford *et al*.[3] reported a rare case of unilateral GG in a 34-year-old woman that persisted 24 months postpartum necessitating reduction mammoplasty.

Breast enlargement is a physiological change that occurs in pregnancy. However, unrelenting and disproportionate enlargement as seen in GG, with associated symptoms, causes a reduction in the quality of life of the pregnant woman. Many authors define gigantomastia as breast enlargement that requires a reduction of over 1.5 kg of tissue per breast. Dafydd *et al*., however, defined gigantomastia as excess breast tissue that contributes more than 3% of the patient's total body weight.[11]

The etiology of GG remains unclear, and no known predisposing factor has been associated with it.[9] The most commonly reported risk factor is the occurrence of GG in the previous pregnancy.[9] It is considered that GG results from varying disease processes, including increased tissue receptor sensitivity, hormonal abnormalities, and/or autoimmune mechanisms.[1,12] However, none of these theories have been widely accepted due to varied findings in reported cases.

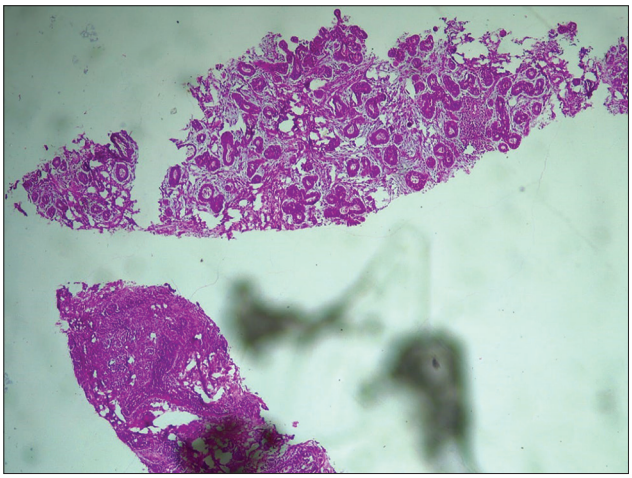

Figure 2: Photomicrograph of GG showing lobular hypertrophy, proliferation of the ducts, and periductal fibrosis (H and E, ×40). GG: Gestational gigantomastia

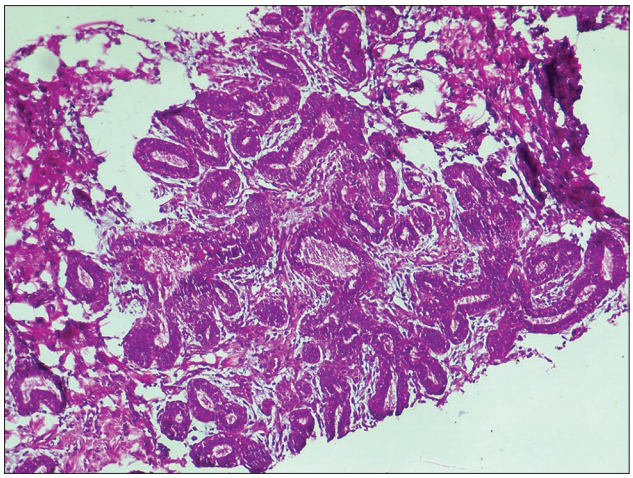

Figure 3: Photomicrograph of GG showing lobular hypertrophy, proliferation of the ducts, and periductal fibrosis (H and E, ×100). GG: Gestational gigantomastia

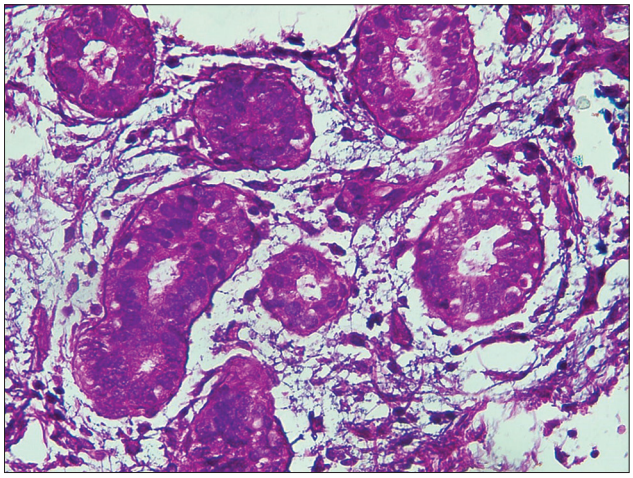

Figure 4: Photomicrograph of GG showing lobular hypertrophy and cystic dilatation of breast ducts (H and E, ×400). GG: Gestational gigantomastia

Figure 5: Postreduction mammoplasty of bilateral gigantomastia

GG causes various physical and psychological symptoms. Patient usually complains of breast heaviness,[7] breast pain,[2,6-8,12] neck pain,[2,5] back pain,[2,5] difficulties in breathing,[6,7] and movement.[2,5-8,12] Some present with complications such as skin atrophy and ulceration,[2,6-8,12,13] bleeding,[7] and sepsis.[2,7,8] Many of these features were observed in our patient.

Histology of breast tissue biopsies of patients with GG most often shows glandular hyperplasia, overgrowth of connective tissue, and tissue fibrosis as seen in this case report. These findings are consistent with normal breast tissue changes during pregnancy.[1]

The management of GG depends on individual presentation and severity of symptoms.[5,14] Bromocriptine is the most commonly used drug with variable response.[9,10] It is an ergot-derived dopamine agonist, resulting in a significant decrease in the release of prolactin from the anterior pituitary gland. Bromocriptine can slow breast hyperplasia, but may not restore the breast size as desired.[1,5,10] Surgical treatment during pregnancy can, thus, be avoided until postpartum. Although safe in pregnancy, bromocriptine has been associated with intrauterine growth retardation.[9] Improvement in symptoms as well as reduction in breast size was reported with the use of cabergoline in the postpartum period.[14] Other hormonal manipulations have not yielded considerable responses.[1,9]

Surgery is the mainstay of treatment for macromastia[3,9] as it helps restore breast size and relieve symptoms. It is indicated when medical treatment fails or complications occur.[5] Surgical options are reduction mammoplasty or mastectomy with delayed reconstruction. Reduction mammoplasty has the potential benefit of allowing for postoperative breastfeeding, but with an attendant risk of recurrence of breast hypertrophy in the subsequent pregnancy.[1] Mastectomy, with delayed reconstruction, is usually reserved for those who have recurrent gigantomastia following reduction or in the presence of life-threatening complications such as severe sepsis and haemorrhage[3] because it is faster with less intraoperative blood loss.[14] Our patient strongly desired to preserve her breast despite understanding the risk of recurrence. She also lacked funds for further reconstruction postmastectomy. Thus, she had a reduction mammoplasty. Ibrahim *et al*. also reported a case of GG in the same region, in which their patient and her caregivers refused mastectomy.[8] These refusals of mastectomy for a benign breast disease may be influenced by cultural beliefs and the notion of "whole body" by females in Africa. Furthermore, breastfeeding is an integral part of motherhood in Sub-Saharan Africa.

Conclusion

GG is a rare benign disorder of the breast in pregnancy that could mimic malignancy and is associated with significant morbidity from the markedly enlarged breast. A multidisciplinary approach in the evaluation and management of the patient is required to improve the outcome of treatment, which is mainly surgical excision of the excess breast tissue in the form of reduction mammoplasty or mastectomy. Patient's wish to preserve "self" will affect decision-making, limiting options

of treatment to use of medications or reduction mammoplasty, especially in the face of a complication.

Declaration of patient consent

The authors certify that they have obtained all appropriate patient consent forms. In the form, the patient has given her consent for her images and other clinical information to be reported in the journal. The patient understands that her name and initials will not be published, and due efforts will be made to conceal her identity, but anonymity cannot be guaranteed.

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Conflicts of interest

There are no conflicts of interest.

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