Case Report

Recurrent bilateral gestational gigantomastia after reduction mammoplasty: A case report and review of the literature

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ABSTRACT

Gestational gigantomastia (GG) is a benign condition characterized by excessive breast enlargement during pregnancy, which can lead to significant physical and psychosocial distress. We present a 22-year-old woman who experienced progressive bilateral breast enlargement for 20 weeks. She was G3P1+1 A1, with her last menstrual period being 28 weeks prior. The patient had experienced similar breast enlargement during her last pregnancy, which was treated with bilateral reduction mammoplasty. Upon examination, both breasts were disproportionately enlarged with no nipple-areolar complex. Initially, she was treated with oral bromocriptine 5 mg daily, which reduced the enlargement. However, her condition worsened in the immediate post-partum period, resulting in significant breast enlargement, deformity, pain, spontaneous ulceration, and tissue necrosis. Ultimately, she underwent a bilateral simple mastectomy. GG is a rare condition that can severely impact a woman's quality of life. The previous occurrence of GG increases the likelihood of recurrence in subsequent pregnancies.

Key words: Bromocriptine, Mastectomy, Recurrent gestational gigantomasti, Reduction mammoplasty

Gestational gigantomastia (GG) is a rare condition marked by swift and excessive breast enlargement during pregnancy, typically occurring in the first or early second trimester [1]. It is a benign breast disease with unclear cause; however, it is believed to stem from hormonal imbalances, hormone overproduction, or heightened sensitivity of breast tissue. This hormonal theory is supported by the fact that most gigantomastia occurs during puberty or pregnancy. GG can lead to significant complications, including ulceration, bleeding, sepsis, and adverse maternal and fetal outcomes. In addition, the psychological impact of having excessively large and disfiguring breasts can be profound. Therefore, early diagnosis and prompt, effective treatment are essential to minimize these complications. The recurrence of GG is particularly concerning, as it may result in severe physical and psychosocial distress for the affected woman and pose a therapeutic challenge for the attending surgeon.

This case report highlights the importance of proper counselling and follow-up for women with GG, given the likelihood of recurrence in subsequent pregnancies. It also adds to the limited literature on the recurrence of GG after reduction mastectomy for previous cases of this condition.

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CASE REPORT

A 22-year-old Hausa lady presented to the plastic surgery clinic with a complaint of progressive bilateral breast enlargement that had been occurring for 20 weeks (Fig. 1). She reported experiencing discomfort and heaviness in her breasts. She was G3P1+1 A1, and her last normal menstrual period was 28 weeks before her presentation. She had experienced similar breast enlargement during her last pregnancy 2 years ago, which led to her undergoing bilateral reduction mammoplasty [2]. Unfortunately, she lost both nipples during that procedure and experienced an intra-uterine fetal death. She had no comorbidity and was not on any routine medications.

On examination, she was conscious, with a temperature of 36.4°C, blood pressure of 100/70 mmHg, a pulse rate of 96 beats/min, and oxygen saturation of 98%. Respiratory and cardiovascular examinations were unremarkable. Both breasts were disproportionately enlarged, with no nipple-areola complex bilaterally. No distinct masses were palpable, and there were no significantly enlarged axillary lymph nodes. Her abdomen was consistent with approximately 28 weeks of gestation.

An obstetric ultrasound scan revealed a live singleton fetus at 29 weeks and 2 days. Her serum prolactin levels were normal.

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She was prescribed oral bromocriptine at a dosage of 5 mg daily, which helped reduce breast enlargement while she was monitored at the outpatient clinic every 2 weeks. She gave birth to a healthy female neonate at 38 weeks and 4 days through spontaneous vaginal delivery.

However, in the early post-partum days, the breast enlargement worsened. The breasts became severely deformed and were associated with pain, spontaneous ulceration, and breast tissue necrosis (Fig. 2), accompanied by a foul-smelling discharge. These complications necessitated her hospitalization despite her ongoing bromocriptine treatment. After adequate resuscitation and counseling, she consented to undergo a bilateral simple mastectomy (Fig. 3) 3 weeks post-partum, fully understanding the possibility of recurrence in future pregnancies. Post-operatively, she developed a surgical site infection, which was managed with dressings and secondary direct wound closure.

Her wounds have healed completely, and she was last seen for a follow-up visit 8 months after her discharge, using improvised breast prostheses due to her inability to afford further breast reconstruction.

Figure 1: (a) Frontal and (b) right lateral views of the disproportionately enlarged breasts with previous surgical scars and absent nipples

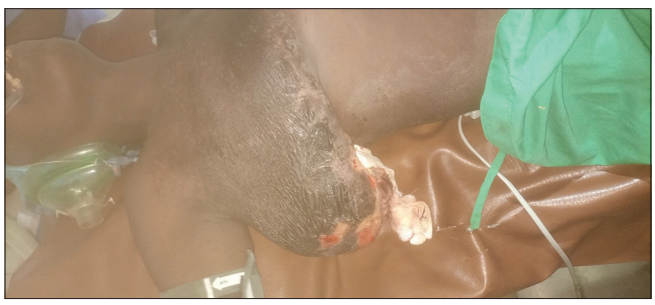

Figure 2: Right breast ulcer and breast tissue herniation

DISCUSSION

The incidence of GG varies between 1/28,000 and 1/100,000 pregnancies worldwide [3]. Reports of GG seem to be increasing, as noted in a 2017 review by Mangla and Singla [1], possibly due to a better understanding of the pathology and increased awareness of the disease. Most cases occur in women aged 20–30 years [1,3-6], similar to the case presented here. While GG may resolve after pregnancy [4], there is a possibility of recurrence in subsequent pregnancies [7]. The condition is characterized by diffuse and relentless breast enlargement, which often leads to breast pain, back pain, and difficulty with ambulation [1,2,4,7,8]. Complications reported in the literature include ulceration, herniation of breast tissue, bleeding, sepsis, and potentially serious outcomes for both the mother and the fetus [2,6,7,9,10]. The psychosocial impact, including challenges with clothing and overall quality of life, can be significant.

The exact cause of GG remains unclear. Common theories related to hormone imbalance or overproduction does not explain instances of unilateral breast involvement reported in the literature. Previous GG is a risk factor for recurrence [8], as observed in this case. Although multi-parity has been suggested as a risk factor for GG, there is considerable variation in the parity of women reported to have GG [4,6-8,10,11]. The condition has also been noted in some primiparous women [7]. The index patient had an uneventful first pregnancy but developed GG during her second pregnancy, with recurrence in her third.

It is commonly mentioned that the remaining breast tissue following reduction mammoplasty can undergo significant hyperplasia [1,10,11], as evidenced in our patient. However, there are limited reports of GG recurrence after reduction mammoplasty in the literature. It is possible that cases managed with reduction mammoplasty did not involve subsequent pregnancies or were not adequately followed up. The reports of recurrent gigantomastia for which the full texts were available are summarized in Table 1,

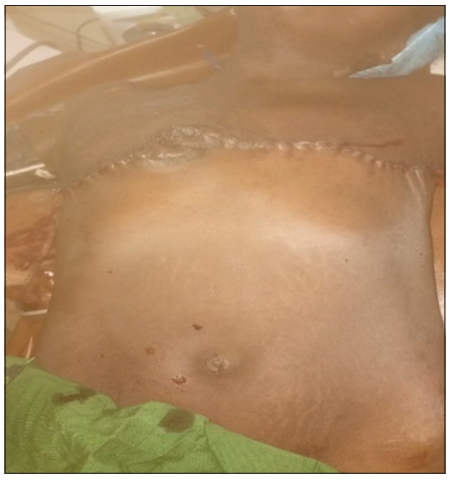

Figure 3: Immediate post-mastectomy

Table 1: Summary of reviewed literature on recurrent gestation gigantomastia

Authors (year)	Age	Reproductive history	Onset of symptoms	Comorbidity	Previous diagnosis	Previous treatment	Interval between recurrences	Treatment for recurrence
Zhou *et al*. (2017) [12]	26	G1P0	12 weeks	Hyperthyroidism	Juvenile macromastia	Reduction mammoplasty	3 years	Bilateral mastectomy
Begum *et al*. (2015) [13]	32	G3P4	3weeks	None	Gestational gigantomastia	Conservative	NA	Conservative
Isik *et al*. 2011) [14]	31	G6P0+5	NA	Prolactinoma	Gestational gigantomastia	Reduction mammoplasty	NA	Bilateral mastectomy
Vidaeff *et al*. (2003) [15]	NA	G1P0	NA	Mirror syndrome	NA	Reduction mammoplasty	2 years	Bilateral mastectomy
Ahcan *et al*. (2003) [16]	27	NA	10 weeks	None	Adolescent macromastia	Reduction mammoplasty	4 years	Bilateral mastectomy
Ship and Shulman (1971) [5]	16	NA	NA	None	Adolescent macromastia	Reduction mammoplasty	6 months	Bilateral mastectomy
Ship and Shulman (1971) [5]	22	G2P1	NA	Myasthenia gravis Ovarian cyst	Bilateral fibrocystic disease of the breast	Reduction mammoplasty	8 months	Bilateral mastectomy

NA: Not available

detailing the age, reproductive history, comorbidity, previous diagnoses and treatments, intervals between recurrences, and the treatments for recurrence [5,12-16].

Table 1 shows that the earliest report of GG occurring in patients who had undergone reduction mammoplasty was by Ship and Shulman in 1971 [5]. When all seven patients with recurrent gigantomastia were evaluated, only two had a prior diagnosis of GG, while three were previously managed for juvenile/adolescent macromastia. One patient had a reduction mammoplasty for bilateral fibrocystic disease of the breast, and the diagnosis of the seventh patient was not clearly stated. The onset of symptoms is often in the first trimester. Recurrence occurred as early as 6 months after the reduction surgery. One patient experienced four recurrences, all managed conservatively, and saw complete resolution of the breast enlargement after childbirth. Notably, patients reported with recurrence had bilateral breast involvement.

The recurrence of GG poses significant challenges for both the affected women and their healthcare providers. Our patient declined a mastectomy during the previous episode of GG about 2 years ago due to her desire to preserve her breasts, which are culturally associated with womanhood in her society. Her initial encouraging response to Bromocriptine suggests it may be an effective treatment option for GG. Bromocriptine is the most commonly reported medical treatment for this condition, with some successful outcomes noted [7,13]. The reasons for the rapid enlargement, ulceration, and necrosis that occurred in the immediate post-partum remain unclear. This may be due to hormonal factors coupled with the increased breast engorgement following the loss of her nipples during the previous episode of GG [2]. However, this rapid post-partum deterioration of the breast enlargement with ulceration and breast tissue necrosis necessitating surgical intervention has been previously observed [15].

CONCLUSION

GG is an uncommon benign breast disease that can cause significant physical and psychosocial disturbances, negatively affecting a woman's quality of life. A history of GG increases the likelihood of recurrence in subsequent pregnancies, underscoring the importance of thorough counseling for women with GG who are contemplating reduction mammoplasty. Mastectomy should be considered early, once fetal safety is confirmed, to prevent serious and potentially life-threatening complications.

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