Case Report

Gestational Gigantomastia: A Patient Case Review and Literature Review

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ABSTRACT

Gestational gigantomastia (GG) or gravidic macromastia is defined as a disorder characterized by a diffuse, extreme and incapacitating enlargement of one or both breast during pregnancy. Gestational Gigantomastia is a very rare condition with a little over a 100cases reported in history. Gigantomastia has been seen to occur in the face of normal hormonal profile. Due to increase in breast mass, the overlying skin is stretched and may undergo necrosis which in the face of pregnancy can cause severe mastitis and haemorrhage.

We present a rare case of GG in a 36-year-old para 7 gravida 8, Zambian woman at 28weeks gestation with classical features of typical GG with normal hormonal profile with respect with pregnancy. She was managed conservatively on Bromocriptine 5mg once a day during pregnancy and postpartum until breast size had reduced to near normal pre-pregnancy size.

We report a rare case of "True gigantomastia which develops rapidly during pregnancy, undergoes regression after delivery, and recurs with subsequent pregnancies" in Zambia.

INTRODUCTION

Gestational gigantomastia (GG) or gravidic macromastia is defined as a disorder characterized by a diffuse, extreme and incapacitating enlargement of one or both breast during pregnancy. Although its true definition is still elusive due to rarity of condition, it is defined as rapid and disproportionate growth of breast during pregnancy, where more than 1500g of breast tissue needs to be removed from the breast.

Breasts in response to hormones will enlarge physiologically at puberty and pregnancy. However, exaggerated hypertrophy with massive enlargement of the breast causing discomfort, pain and embarrassment may occur. This is termed Gigantomastia. Gestational Gigantomastia is a very rare condition with a little over a 100cases reported in history. Its pathophysiology is not well understood but theories look at hormonal changes to be the main cause. However, Gigantomastia has been seen to occur in the face of normal hormonal profile. Due to increase in breast mass, the overlying skin is stretched and may undergo necrosis which in the face of pregnancy can cause severe mastitis and haemorrhage. With breast milk being invaluable in the survival of the new born, Gigantomastia should be managed promptly as it negatively affects infant feeding.

LITERATURE REVIEW

Very little literature is available on this subject due to the rarity of the condition. It was first reported in 1648 by Palmuth.[5] Since his initial description,

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Key words: Gestational Gigantomastia (GG), gravidic macromastia, mastitis, bromocriptine

approximately 100 cases have been reported in the literature [6]. The incidence of GG has been reported to be between 1 in 28,000 and 1 in 100,000 pregnancies between the years of 1935–1960 [7] and 1989–2009, respectively [8]. Till date there is no clear definition, however the commonly used definition is retrospective as it depends on the weight of breast tissue removed during mastectomy. However, there is no clear accepted weight with ranges from 0.8 to 2kg [9]. This condition may be unilateral or bilateral. The enlargement can cause muscular discomfort and over-stretching of the skin envelope which can lead in some cases to ulceration [10]. Gigantomastia is characterized by massive enlargement of the breasts resulting in tissue necrosis, ulceration, infection and haemorrhage, complications that can be life threatening in certain cases [11].

The aetiology is not well understood. Many theories have been brought up, these include over sensitivity to or over-production of hormones such as estrogen, human chorionic gonadotrophin, human placental lactogen and prolactin [12]. However, many cases of Gigantomastia have occurred in a setting of normal hormone levels or even after medical suppression with bromocriptine [13,14]. And in some cases, with normal hormone profile, receptor activity has been found to be normal [15]. In addition to aberrant hormone levels or possible hormone receptor sensitivity, several authors have suggested a possible autoimmune aetiology to Gigantomastia [16]. Several cases of Gigantomastia have been described in association with autoimmune disorders, such as systemic lupus erythematosus (SLE), myasthenia gravis, Graves' disease, and rheumatoid arthritis [8]. Vinicki et al. proposed that pregnancy could be a trigger for SLE emergence [17], suggesting that Gigantomastia in pregnancy might be the result of an underlying autoimmune disorder.

Most cases, patients present with complaints of excessive load including backache, dyspnoea, joint pains and difficulties with movement [6,18]. It usually presents with milk discharge from the breasts [18].

To exclude bilateral breast malignancies associated with pregnancy, some authors have performed biopsy with histopathology. Histologic features of tissues in Gigantomastia commonly include glandular hyperplasia, abundant stromal tissue, acinar/peri-acinar stromal fibrosis, interstitial oedema, and, in some cases, lymphocytic infiltration [19].

The management of gestational Gigantomastia is variable as different case studies have recommended different approaches. In some cases, conservative management has been successful [18], these measures include proper brassiere support, good skin hygiene, analgesia, and adequate nutrition [19]. Bromocriptine is the mainstay conservative management. It has been seen to reduce breast size, promote regression and prevent ulceration, promote healing and decreasing the need for surgery [20]. However, it has been associated with intrauterine growth restriction hence making frequent foetal monitoring invaluable. Conservative management with bromocriptine has been unsuccessful in some case studies [10] and this is an absolute indication for surgical management. Other indications for surgery include severe infection, ulceration and haemorrhage. Surgery depends on the size of pregnancy and risk of miscarriage during surgery [21,22]. Since there is a possibility of recurrence with simple mastectomy or reduction mammoplasty, bilateral mastectomy with reconstruction is the treatment of choice in women who desire future pregnancies [14]. In the literature review of data from 1968 to 2006 by Swelstad MR et al, medical management successfully avoided surgery in 39% of cases. However, 35% eventually underwent breast reduction (12%) or mastectomy (88%) during pregnancy. 30% had spontaneous or elective termination of pregnancy. Patients who underwent breast reduction and then became pregnant had a 100% chance of recurrence [23].

CASE PRESENTATION

A 36-year-old para 7 gravida 8, Zambian woman at 28weeks gestation, was referred to our hospital from her rural clinic with a history of excessive bilateral

breast enlargement. She had been treated as mastitis on antibiotics at the local clinic with no improvement. She recounted that the problem started about 2months into her pregnancy. The referral was prompted by worsening symptoms including ulceration of the left breast, postural breathing difficulties with heaviness on the chest, back/neck pain and some breast pain. She had seven previous pregnancies with spontaneous vaginal deliveries (SVD), and no associated history of excessive breast enlargement with any of the past pregnancies. Her past medical history was unremarkable, and was not any medications.

Examination

On examination she was diaphoretic, not in obvious respiratory distress, had markedly enlarged breasts and a right axillary accessory breast. The left breast was larger than the right, indurated and hyper-pigmented with a clean ulcer (figure 1 and 2). Obstetric examination revealed a height of fundus of 28/40 with a viable singleton pregnancy in cephalic presentation.

Figure 1: showing enlarged breasts, with right axillary tailand ulcer on right breast. Picture courtesy of Dr Musa Gerald, Livingstone Central Hospital.

Figure 2: Patient in right lateral position. Picture courtesy of Dr Musa Gerald, Livingstone Central Hospital.

Investigations

Full blood count- Hemoglobin - 7.8g/dl, hematocrit-23.6%, MCV- 90.1fl and rest of cell lines normal.

Hormone profile - Essentially normal: prolactin, progesterone, estradiol, FSH and LH (table 2).

Table 1. Hormonal third trimester profile

Test	Result
Prolactin	157ng/ml
Progesterone	190.8nmol/L
Estradiol	7890pmol/L
FSH	0.1U/L
LH	< 0.1U/L

Obstetric ultrasound- Obstetric ultrasonography confirmed a singleton viable pregnancy with fetal weight of 1600 grams.

Breast ultrasound- Suggested breast hypertrophy with no solid nor cystic lesions

Treatment

Conservative management in a Multidisplinary approach as an outpatient with 2weekly reviews on Bromocriptine 5mg per day with adequate analgesia as required.

Wound care with povidone iodine cream/solution. Was advised to be nursing, resting in bed in propped up position.

She delivered a 2.6kg female baby with an APGAR score of 9/10 at 33weeks 4days by dates via spontaneous vaginal delivery. Post-delivery, the patient was reviewed every fortnight

Bromocriptine was continued after delivery at 5mg per day until breast size had reduced to near normal pre-pregnancy size.

Patient counselled on high recurrence on next pregnancies and opted for permanent contraception.

DISCUSSION

Gestational Gigantomastia is a very rare complication in pregnancy with very few reported in literature. The management of this condition is not fully agreed upon as some authors have tried conservative management with good outcome and others have had to employ surgical management. Like most cases reviewed in literature, our patient presented with painful and abnormally enlarging breasts around the first trimester at about 8-12 weeks. She had already started developing ulcers on the breasts at presentation. The ulcers are due to pressure necrosis of the skin as a result of stretch and increase in breast volume. The hormonal profile was done and quantitatively the hormones were within normal range for the pregnancy. This is similar to the findings in literature. However, no receptor studies were done to test the receptor activity in response to normal hormonal stimulation. She had no history of similar condition in the family or previous pregnancies and no symptoms of autoimmune disease.

The patient was managed conservatively after discussion on the condition and management options. She was started on bromocriptine 5mg once daily. The medication was continued up to 6 weeks postpartum. One month into the medical therapy she reported slight reduction of the pain and the size of the breast. Objectively the breasts had reduced size by 5cm and 3cm, right and left respectively. This is in keeping with literature on the symptomatic benefits of bromocriptine. She delivered at 33weeks a 2.6kg female baby. This was normal weight based on an average of her other children. Hence in this case there was no fetal growth restriction as postulated with bromocriptine use. She was lactating and was encouraged to express breast milk to feed the neonate. She reported symptomatic relief and reduction in breast size postpartum. The ulcers progressively reduced in size as the breast size reduced and had healed by 6weeks post-partum.

CONCLUSION

Gestational Gigantomastia is a rare complication of pregnancy and may have debilitating and fatal complications to the patient. With massive breast sizes it makes neonatal breast feeding very difficult and may cause malnutrition in the neonate. The management of the condition should be individualized taking into consideration presence of potentially life-threatening complications like hemorrhage and severe infection. However, in the absence of said complications, conservative medical management maybe employed with close monitoring. This is an example of a good outcome with conservative management.

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