CASE REPORT

Gigantomastia Complicating Pregnancy: A Case Report

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ABSTRACT

***Background**: Mammary gigantism is a rare, cosmetically embarrassing complication of pregnancy that may ulcerate and have potentially fatal bleeding.*

***Methods**: A case report of a 20-year old primigravida with bilateral massive breast enlargement is presented to highlight the clinical presentation and management challenges of the condition.*

***Results**: She was treated with local debridement, bromocriptine, antibiotics and blood transfusion with good results. The pregnancy however terminated at 27 weeks of gestation.*

***Conclusion:** Gigantomastia is a rare complication of pregnancy, which may pose a major management challenge. A favorable outcome may be achieved with prompt recognition of the condition and conservative management in selected cases.*

***KEYWORDS:** Gigantomastia; Pregnancy; Nigeria.*

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INTRODUCTION

Physiological enlargement of the breast occurs during puberty and in pregnancy. Rarely massive diffuse enlargement of the mammary gland may complicate pregnancy in a condition known as gigantomastia[1]. This condition, first described in the German literature in 1648 by Palmuth[2] and in the English literature by Simpson[3] in 1920, was extensively reviewed in 1989 by Beischer *et al*[4]. With an incidence of less than 1 in 100,000 pregnancies, mammary gigantism is a rare yet potentially fatal and cosmetically embarrassing complication of pregnancy. This paper is the report of one case managed in the Obstetrics unit of Aminu Kano Teaching hospital in Northern Nigeria.

CASE REPORT

The patient Mrs M.J. a 20-year old primigravida, presented at 15 weeks of gestation with a three-week history of rapid enlargement of both breasts. She was well and the pregnancy largely uneventful, prior to the onset of this symptom. Initially she thought the enlargement to be the normal pregnancy related breast changes. However, when she began to have pains she consulted a private practitioner who treated her with ciprofloxacin and diclofenac potassium with little effect. She subsequently felt some lumps in both breasts. The breast enlargement continued and the left breast developed a blister, which later ulcerated with severe haemorrhage. She was then referred to Aminu Kano Teaching Hospital (*figure 1* shows the breasts at presentation). She had associated difficulty lying on her side or turning over in bed. There was no history of use of hormonal contraception, or any breast disorder in her or her family members.

On examination the breasts were massively enlarged and oedematous. There was an ulcer measuring 4cm in diameter on the left breast.

Her haematocrit was 22%. Serum electrolytes and liver enzymes were normal. A swab from the ulcer grew *Staphylococcus aureus*. Fine needle aspiration biopsy from both breasts revealed stromal hypertrophy with ductal proliferation but no evidence of malignant change. Hormonal assay was not done for lack of funds.

She was admitted and the breasts were rested on pillows. They were supported with a corset when she walked. The ulcer was dressed twice a day and she was treated with antibiotics and analgesics. Bromocriptine 2.5mg twice daily was prescribed. It was later increased to four times a day and was well tolerated. She was transfused with two pints of blood. After two weeks, the pressure symptoms had improved, the ulcer was clean and patient requested for discharge to be coming for wound dressing from her nearby home. When she didn't have money to buy bromocriptine she defaulted follow up and had to be traced by the social welfare services of the hospital.

By this time the breasts were much more enlarged and oedematous. The right breast had also ulcerated, and both ulcers were infected by *Staphylococcus aureus* (*figure 2*). Both nipples were however preserved. She had another episode of heavy bleeding from the breast ulcers and her haematocrit dropped to 18%. There was associated deterioration in her general condition. She was resuscitated with antibiotics, intravenous fluid and three pints of blood. Analgesics, antibiotics, bromocriptine and tamoxifen were given with the aid of social welfare services of the hospital. She was counseled for mastectomy or reduction

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mammoplasty to both of which she declined consent.

The pregnancy progressed normally, until 27 weeks of gestation when she had a stillbirth of a female fetus (weight 900g). The breasts became even further enlarged despite addition of frusemide to the above regimen. A lot of necrotic tissue was found extruding from the ulcer. She had local debridement, and some bulk reduction in both breasts. There was massive haemorrhage introoperatively necessitating the transfusion of four pints of blood. She did well postoperatively and was discharged three weeks post-operatively. She requested for contraception and had CUT 285 intrauterine device inserted postpartum after adequate counseling. *Figure 3* shows the breasts after treatment.

Histology revealed extensive ductal proliferation with stromal oedema but no evidence of malignant change. The IUCD was removed after one year when she expressed the desire to get pregnant. She subsequently conceived and delivered a live male fetus at term. The antenatal period was uneventful and there was no recurrence of the breast enlargement.

Fig 1. The patient at presentation.

Fig 2. The patient immediately pre-operative.

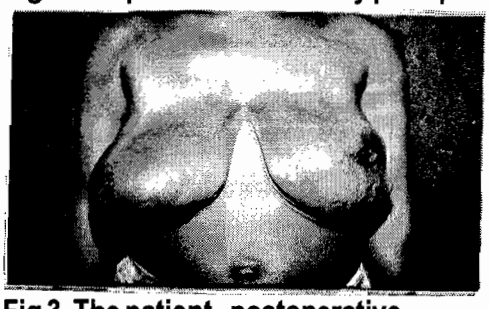

Fig 3. The patient - postoperative.

DISCUSSION

The profile of patients affected by this disorder is not well established, but majority are young women in their first pregnancy (9 of 17 cases)[4]. However, the condition was first recorded amongst grandmultiparae[1]. Similarly the time of onset of the breast enlargement has been variable; usually commencing in early pregnancy[5,6] as in this case, but the 4 cases reported by Geschicktar all started in the puerperium[7]. In most cases, breast enlargement was noticeable in early pregnancy and the breasts were already large by the end of the first trimester as in this case. Other clinical features include ulceration, oedema, progressive anaemia, infection, necrosis, skin fissures and intrauterine growth retardation[4,8]. Bakeoff has reported its association with habitual abortion, which resolved following reduction mammoplasty[9].

Pregnancy outcome is similarly variable but majority of patients have term delivery. In this case the pregnancy terminated at 27 weeks of gestation with a female fetus weighing 900g. Pre-term delivery, was similarly reported by Leis *et al*[10] and generally female babies predominate[4]. In this case however, infection, primiparity and low socio-economic status may be important confounding factors.

Why this enlargement occurs remains uncertain. Several theories are advanced. Endocrine imbalance appears most likely[8], but hormone assay in many cases in the literature fail to reveal a pattern[8,11]. Report of hyperprolactinaemia by Wolner-Haussen *et al* was isolated. Efforts to establish increase receptors for oestrogen, progesterone, steroids and androgen in affected mammary specimens has been inconclusive[11]. Hypersensitivity to these hormones unrelated to receptor density is plausible but has not been investigated.

Since the aetiology of this disorder is yet to be defined conclusively, its treatment has also remained speculative[12]. Oestrogens, androgens and vitamin E are ineffective[1]. Norethindrone advocated by Lewison *et al*[13] was not found useful by others[4]. Similarly the initial response we observed in this case with bromocriptine was not sustained in other reports[4]. We used tamoxifen here in the hope that the effect of androgens on mammary gland may result in regression of the growth, but this was ineffective, again in pregnancy its effect on the commonly associated female fetus must be considered. A tempting consideration of therapeutic abortion has been associated with poor results[11]. Furthermore, there are ethical concerns on sacrificing a life for an organ.

Surgical treatment of this disorder is also controversial, the options being subcutaneous mastectomy, total mastectomy and reduction mamoplasty[8,14]. Our patient declined all three options, leaving only the opportunity for local treatment of the ulcers. Contraception in this patients is apprehensive, as it is not known what role exogenous sex hormones play in this disorder. This patient tolerated Cu T285 IUCD well for one year. Several authors have reported the recurrence of massive hyperplasia of the breast in subsequent pregnancy[4,6] . This patient had an uneventful pregnancy and delivered a live fetus following this treatment.

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