

Rare disease

Gestational gigantomastia with complete resolution in a Nigerian woman

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Summary

Physiological enlargement of the breasts occurs at puberty and during pregnancy. It is known as gestational gigantomastia when enlargement in pregnancy becomes excessive, uncomfortable and embarrassing. Gestational gigantomastia may have far reaching effects for the mother and fetus. This rare condition is associated with considerable morbidity but may be associated with good fetal outcome. Our case was very interesting because it was managed conservatively and postpartum there was complete spontaneous resolution.

BACKGROUND

Gestational gigantomastia is a very rare condition and only about 100 cases have been reported in the literature. This is the first case of gigantomastia in pregnancy in our collective experience and is significant because to the best of our knowledge, few cases of postpartum complete spontaneous resolution have been reported. The breasts are of vital importance to the newborn child, particularly in developing countries where breast feeding is common. Breast feeding confers numerous advantages on the infant including reduced mortality rate and improved neurological development. Physiological enlargement of the breasts

occurs at puberty and during pregnancy, when it starts very early and is sustained until delivery. The factors controlling breast growth are complex and not completely understood, although oestrogens, progesterones, prolactin, growth hormone and adrenal steroids are all known to play a role. Sometimes this process goes wrong resulting in an excessively large and painful breast called gestational gigantomastia (gravidic gigantomastia, mammary hyperplasia of pregnancy) or virginal hyperplasia when it occurs at puberty. This rare but important condition of the breast not only interferes with breast feeding but may cause severe maternal morbidity and even mortality.



Figure 1 Erect patient with gigantomastia.



Figure 2 Patient with gigantomastia in supine a position.

CASE PRESENTATION

A 24-year-old teacher presented at 26 weeks gestation with marked bilateral breast swelling which started when she was 10 weeks pregnant. She was para 2+0 with one child alive. The swelling started insidiously in the left breast and 2 weeks later extended to the right breast. There was no itching or pain initially, but there was a history of milky discharge from both breasts. She subsequently developed severe pain and discomfort in her breasts, neck and back with marked limitation of movement as the breasts enlarged.

The patient was pale, afebrile and had bilateral axillary lymphadenopathy. Her pulse rate was 86 bpm, blood pressure was 80/60 mm Hg and the neck veins were engorged. Heart sounds were normal and the chest was clinically clear.

Both breasts were grossly enlarged and extended to the level of the tubercle of the iliac crest (figures 1, 2).

The right breast was slightly bigger than the left. Anteriorly, the left breast had two infected ulcerated areas, the result of herbal preparations applied by a native doctor. Both breasts had dilated veins and exhibited peau d'orange, and a milky discharge could be expressed from each nipple. There was no organomegaly and the uterus was enlarged to a size consistent with a 26-week pregnancy. A diagnosis of gestational gigantomastia complicated by infection, ulceration and anaemia was made. The patient was admitted and started on antibiotics, daily dressing, haematinics and analgesics. Investigations showed a packed cell volume (PCV) of 21% and a white blood cell (WBC) of $18.7 \times 10^9/\text{dl}$. Urinalysis, urea, liver function tests and hormone studies were normal. The patient often bled severely, spontaneously and during dressing, and this led to severe anaemia. Her PCV subsequently fell to 18% at the onset of labour.

Wound swabs were done regularly and antibiotics were changed accordingly. The pregnancy was monitored by serial ultrasound and proceeded satisfactorily. The patient went into labour at 38 weeks gestation and had a caesarean section for fetal distress. A live female infant weighing 2.4 kg was delivered. Breast feeding is desirable particularly in developing countries, but in this case, the massive size of the breasts made it difficult for the patient to safely breast feed her baby. The postoperative period was complicated by severe anaemia which was treated with blood transfusion and haematinics. The patient was discharged on the 8th postoperative day and given an appointment for reduction mammoplasty. However, the gigantomastia underwent complete spontaneous resolution within the puerperium.

INVESTIGATIONS

- ▶ PCV of 21%
- ▶ WBC of $18.7 \times 10^9/\text{dl}$
- ▶ Urinalysis, blood urea and electrolytes, liver function tests and hormone studies were normal.

DIFFERENTIAL DIAGNOSIS

- ▶ Phylloides tumour
- ▶ Lymphoma of the breast.

TREATMENT

Conservative management was employed in this case of gigantomastia in pregnancy.

OUTCOME AND FOLLOW-UP

Outcome and follow-up were good for both mother and baby.



Figure 3 Patient with resolved gigantomastia.

The gigantomastia had resolved completely by 8 weeks after delivery when the patient returned for a follow-up visit.

DISCUSSION

Gestational gigantomastia was first described in 1684 by Palmuth¹ and is very rare. An incidence of 1 in 28 000 to 1 in 100 000 pregnancies has been quoted.² It is a severely debilitating condition in which massive enlargement of the breasts may be accompanied by thinning of the skin, tissue necrosis, infection and haemorrhage. Movement and respiratory difficulty and emotional, social and psychological problems may also occur.³

To date there is no universally accepted definition.⁴ Some authors have suggested the amount of breast tissue removed at surgery should determine the definition. However, this has the disadvantage of being a retrospective diagnosis. Moreover, there is disagreement among authors on the amount of tissue removed to substantiate the diagnosis, with figures ranging from 0.8 to 2 kg.

The aetiology is unknown but various factors have been proposed. These include over-sensitivity to or over-production of hormones such as oestrogen, human chorionic gonadotrophin, human placental lactogen and prolactin.⁵ In this patient hormone studies were normal. Abnormal excessive breast enlargement at puberty, which may have an endocrine aetiology, is known as juvenile gigantomastia. Gigantomastia has also been reported in cases of mirror syndrome in pregnancy.⁶ The description of a few cases outside puberty and pregnancy⁷⁻⁸ makes the hormonal aetiology even more uncertain. It has also been suggested that the condition could arise as a complication of D-penicillamine therapy.⁹

In this case the poultice applied to the breasts by a native doctor precipitated ulceration, which led to recurrent

bleeding and severe anaemia. Such a scenario is not unusual in developing countries where native doctors still play a key role in healthcare delivery. Ulcers have also been known to arise spontaneously secondary to thinning of the skin and subsequent tissue necrosis.

This condition can occur in any pregnancy but once it has occurred recurrence is likely in subsequent pregnancies.

Apart from the physical problems of pain and ulceration, this condition may be associated with severe social problems, as demonstrated by the abandonment of this patient by her husband. Other authors have recorded depression and low self-esteem.

Phylloides tumour, fibroadenoma, non-Hodgkin's lymphoma¹⁰ and lymphoblastic lymphoma¹¹ are possible differential diagnoses, which can be excluded by biopsy.

Breast biopsy was avoided in our patient because she had anaemia in pregnancy and there was significant risk of haemorrhage. Swelstad *et al* examined mastectomy specimens from gestational gigantomastia histologically and found significant lobular hypertrophy, ductal proliferation and periductal fibrosis,¹² consistent with the changes found in the pregnant breast.¹³ Other histological features of gestational gigantomastia include extensive lobular hyperplasia, dilated ducts and pseudoangiomatous hyperplasia. Some areas also revealed interstitial oedema, lymphoplasmocytes in the stroma and foci of increased fat and connective tissue.¹⁴ Lymphoma usually progresses postpartum.¹¹

Treatment may be medical or surgical. The plethora of medical remedies, including tamoxifen, bromocryptine, testosterone, dydrogesterone, hydrocortisone, diuretics and medroxy progesterone acetate,¹⁴ which have been used in its treatment belies its unknown aetiology. Medical treatment is indicated if there is no morbidity. Bromocryptine is the drug of choice as it has been reported

to stop progression and cause regression.^{2 15 16} Although the results of bromocryptine treatment are inconsistent,^{17 18} it is worth trying since, if successful, the need for surgery during pregnancy will be avoided in 39% of patients.^{12 18} However, care should be taken as fetal growth restriction has been recorded.¹²

When complications such as cardiac failure,¹² massive haemorrhage,¹⁹ ulceration or breast necrosis occur, surgical treatment is advised.¹⁴

Surgery was not carried out in this patient as she declined this option after delivery. Mastectomy with implant reconstruction is the treatment of choice as reduction mammoplasty results in more bleeding and a higher chance of recurrence.¹² However, breast prostheses are not yet widely used in developing countries, and implants have their own complications.²⁰ Also, since mastectomy without implant may have adverse psychological effects, reduction mammoplasty may be better in developing countries despite the risk of recurrence and the possibility of multiple operations. This patient had complete spontaneous resolution of gigantomastia by 8 weeks after delivery (figure 3), as also reported by others.²¹

CONCLUSION

This fortunately rare condition is particularly important in developing countries as it prevents breast feeding, which is crucial for the development of the infant, and prevents effective contact between mother and baby, thus making bonding difficult. Gestational gigantomastia does not preclude a normal delivery,¹⁹ although in this case caesarean section was carried out for obstetric reasons. However, the

severe anaemia secondary to haemorrhage from the lesion in this case could have caused maternal death. Since complete spontaneous resolution of gigantomastia can occur postpartum, there is a need for caution before resorting to surgical treatment.

Competing interests None.

Patient consent Obtained.

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Learning points

- ▶ Gigantomastia in pregnancy progresses fairly rapidly.
- ▶ Spontaneous haemorrhage from breast ulcers is worrisome.
- ▶ Conservative management of gigantomastia can lead to good fetal and maternal outcome.
- ▶ Gigantomastia makes breast feeding impossible, a serious problem in developing countries where poverty limits the provision of alternatives to breast milk.
- ▶ In this case, spontaneous complete resolution of gigantomastia was observed postpartum and breast surgery was unnecessary.

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