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Case Report

# Gestational Gigantomastia: A Rare Case

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**Abstract**

Gestational gigantomastia is a rare condition of unknown etiology with psychological and physical adverse effects on the mother. Our case presented as an unbooked case at 37 weeks of gestation and was managed conservatively till delivery followed by spontaneous resolution post delivery.

## ABBREVIATIONS

OPD: Outpatient Department; CBC: Complete Blood Count; AEC: Absolute Eosinophil Count; ESR: Erythrocyte Sedimentation Rate; Anti-DSDNA: Anti-Double Stranded De-Oxy Ribonucleic Acid; ANA: Anti-Nuclear Antibody; RA Factor: Rheumatoid Factor; Anti-TPO: Anti-Thyroid Peroxidase Antibodies; FNAC: Needle Aspiration Cytology; SLE: Systemic Lupus Erythema

## INTRODUCTION

Physiological enlargement of the breasts normally occurs during puberty and pregnancy but when the breast weight exceeds 3% of the total body weight, it is known as macromastia [1]. If the condition occurs during puberty, it is known as juvenile macromastia or virginal hypertrophy and when it occurs in pregnancy, it is known as gestational macro-mastia or gigantomastia. Breast hypertrophy can occur in both the breasts or it can solely affect one breast causing breast asymmetry. We describe here a case of gestational gigantomastia with a favorable outcome along with the current understanding of pathogenesis, diagnosis, management and the review of literature.

## CASE PRESENTATION

Mrs. X, 28 year old lady, G2P1L1 with previous full term normal vaginal delivery, presented to the Antenatal Outpatient department (OPD) for the first time at 37 weeks of gestation for her massively enlarged breasts. She had discomfort while walking and standing and had severe back pain because of them. She complained of breathlessness off and on and had to support her breasts with a sheet as no brassiere would fit her. She gave history of rapid growth of breasts since third month of pregnancy which continued up till sixth month after which it stabilized. She had no history of breast enlargement in previous pregnancy. There was no history of drug intake. On examination, her breasts were massively enlarged, erythematous with peude orange appearance. On palpation, they were non tender with pitting edema. Bilateral nipple areola complex was normal. There was no galactorrhoea, no palpable lump or lymphadenopathy. There were en-gorged veins over the breast but there were no areas of ulceration. On examination of the abdomen, the uterus was term size with a single live intrauterine fetus in cephalic presentation. She was admitted with a differential diagnosis of microfiliriasis or phylloides tumor of the breast. Her breast measurements were taken. The circumference was 72cm and 69cm of right and left breast respectively. The total chest circumference was 152cm. Laboratory work up included complete blood count (CBC) with peripheral smear to rule out microfilariasis, absolute eosinophil count(AEC), erythrocyte sedimentation rate(ESR), serum calcium, serum estrogen, progesterone, prolactin, thyroid profile, anti-double stranded de-oxy ribonucleic acid (anti-dsDNA), anti-nuclear antibody(ANA), rheumatoid factor( RA factor) and anti-thyroid peroxidase (anti-TPO) antibodies. All of these were within normal range. On ultrasound, there was hypertrophied breast parenchyma with diffuse skin thickening and subcutaneous tissue edema. There was no evidence of any abscess or any pus collection. Ducts were dilated at some places, bilateral axilla were normal. Fine needle aspiration cytology (FNAC) revealed monolayer sheets and highly cohesive clusters of benign ductal epithelial cells with interspersed myoepithelial cells and occasional stromal cells, no microfilaria was seen. Literature was reviewed and diagnosis of gestational gigantomastia was made. Patient was kept on conservative management with adequate breast support, hydration, analgesics and rest. She went into spontaneous labor at 40 weeks but had a caesarean section with bilateral tubal ligation in view of fetal distress and delivered a 3.4 kg healthy baby girl. Post partum patient was uncomfortable because of the enlarged and engorged breasts and chose not to breast feed the baby. We gave her tablet cabergolin 0.25miligram twice a day for 3 days as breast milk secretion had already been established. Breast secretion stopped and breast became soft but were still of the same size as in antenatal period. Patient was discharged on post op day 7 after the stitch removal in a stable condition. Her breasts continued to be of the same size but were soft. Patient was followed up after two weeks and six weeks post partum. The breasts had started to decrease in size from six weeks post partum and had a complete resolution to normal size as before within 10 months of delivery.

## DISCUSSION

Gestational gigantomastia is a rare condition with an incidence of 1 in 28,000 to 1 in 1,00,000 [2,3]. The first case was reported in 1648 by Palmuth [4] and since then about 100 [1] cases have been reported. Surprisingly, more than 50 cases have been reported in the last four decades [5]. This is the only case we have encountered so far. As there is no fixed definition of gigantomastia [5], we used the breast circumference at the level of the nipple for breast size measurement, as we were not aware of the jugular nipple distance, which is one of the standard breast measurement used in the plastic surgical clinical practice [6]. However, retrospectively, by measuring the length and the width of the bed from the Figure 1, an approximate jugular nipple distance has been extrapolated to be 145 cm and 139 cm for right and left breast respectively.

Risk factors for breast hypertrophy have not been well understood but it is seen more commonly in Caucasians, obese and multiparous women [2]. Gestational gigantomastia can occur in any pregnancy with a tendency to recur in subsequent pregnancy. Swelstad et al., reported a recurrence rate of 100 percent in the women who underwent breast reduction mammoplasty [7].

The etiopathogenesis of breast hypertrophy has not been well elucidated with a number of theories being proposed. These include either over sensitivity of the breast tissue to the hormones or over production of hormones such as estrogen, progesterone and prolactin [2,8]. It can be a response to an underlying autoimmune disorder [9,10]. A number of drugs have also been indicated in breast hypertrophy like penicillamine, bucillamine, indinavir, cy-closporin and neothetazone [8].

Over production of hormones can occur in conditions such as aromatase excess syndrome resulting in hyperestrogenism or with pituitary adenomas resulting in hyper prolactine-mia Figure 2. In our case, hormonal profile was normal. Hypersensitivity of the receptors to the hormones has been proposed as a cause but would result in hypertrophy across all the pregnancies. A number of authors have suggested an underlying autoimmune disorder such as SLE, rheumatoid arthritis and graves' disease [9,10] giving rise to gigantomastia. In our patient, anti ds DNA and RA factor both were negative and serum TSH was normal. Microfilariasis, phylloides tumor, non-hodgkin's lymphoma [11] and lymphoblastic lym-phoma [12] are other differential diagnoses which were ruled out through FNAC.

**Figure 1** Supine pregnant patient.

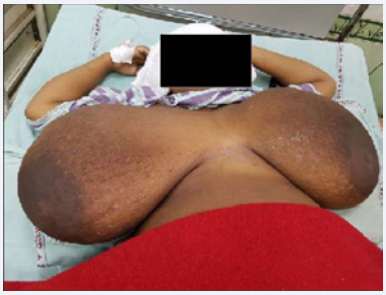

**Figure 2** supine post partum patient.

**Figure 3** Resolved gigantomastia 10 months after delivery.

Apart from causing physical and psychological stigma for the woman, gigantomastia is associated with a number of complications; pain, ulceration, breast necrosis, massive haemorrhage, sepsis, cardiac failure and even cases of death have been reported. Khosla et al., reported hypercalcemia to be associated with a case of macromastia, possibly due to an increased production of parathyroid hormone related protein by the hypertrophied breast tissue Figure 3 [13].

Treatment may be medical or surgical and varies from case to case. Conservative man-agement includes optimal support for the breast, good skin hygiene, adequate nutrition and topical ice application to cool the breast. A plethora of medical treatment includes tamoxifen, danazol, progesterone, testosterone and bromocriptine [2,14]. Till date bromocriptine is the drug of choice in antenatal period, but the results are inconsistent [5,15,16]. How-ever, prolonged usage of bromocriptine results in fetal growth restriction, hence caution has to be exercised. In our case, since the patient came to us at term, she was managed conservatively and post delivery she was given cabergolin to suppress milk production.

Surgical management is needed in the cases where complications or failure of medical therapy arise. Both reduction mammoplasty and bilateral mastectomy are recommended. Bilateral mastectomy has no chances of recurrence and is the treatment of choice if fur-their pregnancies are desired [17]. Post partum spontaneous resolution, although limited in number, have been reported [2,5], so caution is advised before going ahead with the surgery. Mangla et al., in 2017 carried out a systemic review of all the cases of gestational gigantomastia from 1976 to 2016

and analyzed 50 case reports of gestational gigan-tomastia [5]. Out of 50 cases, only two underwent postpartum spontaneous resolution. Three cases were managed conservatively and two cases managed medically with bromocriptine. Two most successful treatments were reduction mammoplasty, done in 22 patients and simple mastectomy done in 15 patients respectively.

Gestational gigantomastia is a rare disease of unknown etiology. It is of major concern in developing countries as it prevents the mother from breast feeding the infant. A complete knowledge about the possible factors leading to the breast enlargement and its management helps the obstetricians to manage this distressing situation.

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