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Syed Sarmad Bukhari

Aga Khan University, sarmad.bukhari@aku.edu

Haris Manan

Teaching Hospital, Peshawar, Pakistan.

Mah Muneer Khan

Teaching Hospital, Peshawar, Pakistan

Syed Shahmeer Raza

Hayatabad Medical Complex, Hayatabad-Peshawar, Pakistan.

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CASE REPORT

RESOLUTION OF GESTATIONAL GIGANTOMASTIA WITH
TERMINATION OF PREGNANCY

Syed Sarmad Bukhari, Haris Manan*, Mah Muneer Khan*, Syed Shahmeer Raza**

Department of Neurosurgery, Aga Khan University Hospital, Karachi, *Department of Surgery, Khyber Teaching Hospital, Peshawar,

**Department of Surgery, Hayatabad Medical Complex, Hayatabad-Peshawar-Pakistan

Less than 100 cases of gestational gigantomastia have been described in literature. The aetiology and risk factors are not well-established. Various treatments have been used with some consensus. We present the case of a 47-year-old female who presented to us with bilateral gigantomastia in her 16th week of gestation. She had massively enlarged breasts which were very painful. Relevant laboratory investigations were normal. An incisional biopsy done prior to, and two trucut samples at presentation to us, showed normal breast tissue proliferation. In the absence of adequate pain control, it was decided to electively terminate the pregnancy and give a trial of tamoxifen. She made a rapid recovery following termination without requiring the use of tamoxifen

Keywords: Gigantomastia; Pregnancy; Gestational gigantomastia

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INTRODUCTION

Breast enlargement is a natural phenomenon that occurs during puberty and pregnancy due to interplay of hormones like oestrogen, progesterone, growth hormone, prolactin and adrenal steroids. Any disturbance in this balance can potentially lead to Gestational Gigantomastia (gravidic gigantomastia, mammary hyperplasia of pregnancy) or virginal gigantomastia (if it occurs at puberty).¹ According to one criterion, gigantomastia can be defined as excess in Breast weight that exceeds 3 % of body weight.² Excess weight of Breast can lead to muscular discomfort and ulceration due to stretching of skin.³ Breasts are of vital importance to the health of baby especially in a developing country. There are only 100 cases so far reported in literature. A case study in 2011 led the doctors to believe Gigantomastia as one of the possible causes of Mondor's syndrome and by treating Gigantomastia at an early stage could potentially prevent development of Mondor's syndrome.⁴

CASE PRESENTATION

A 47-year-old female patient presented to the surgical clinic with massively enlarged breasts for the past 2 months. She was 4 months pregnant at the time of presentation. Both breasts had started enlarging insidiously without any precipitating cause and were exquisitely painful. Dimensions are given in table-1.

On examination, the skin overlying the breasts was erythematous and painful to touch. The nipple was normal in appearance and there was no discharge. Skin was intact without any ulceration, except for a scar on the right indicating the previous biopsy site (Figure-1). Palpation of the breasts did not reveal any distinct masses but a diffuse enlargement

was appreciated. There was no axillary lymphadenopathy. She denied any weight loss. Physical examination was otherwise unremarkable. Her medical and surgical history was unremarkable. She had 8 children and never had any complications in any of those pregnancies.

Standard prenatal labs; CBC with differential; serum chemistry panel; liver function tests; serum calcium and albumin levels; hormone profile: oestrogen, progesterone, and prolactin were all within normal range.

Prior to presentation she had an incisional biopsy done at a different centre. There was a strong suspicion of inflammatory breast cancer hence several Trucut biopsies were taken and sent for histopathology. The other differential considered was lymphoma and tuberculosis of the breast. Results from the earlier biopsy and trucut specimen showed the tissue contained chronic inflammatory cells with no evidence of breast neoplasm. Considering the presentation of the patient it was considered prudent to have the Trucut repeated from different sites. This biopsy confirmed the histopathology to be the same as the previous sample.

The diagnosis of gestational gigantomastia was then considered. The patient was counselled regarding her disease and she decided to terminate the pregnancy before commencing tamoxifen. A week following elective therapeutic abortion, her condition rapidly improved. The breasts returned to the pre-gestation size and her pain resolved. No further intervention was done and the patient was not asked to come for follow up since she lived in a remote area of the country. However, inquiry about her status was made over the telephone.

Table-1: Breast dimensions of patient with gestational gigantomastia at 16 weeks of pregnancy

Breast dimensions	Right	Left
Breast circumference	46 cm	55 cm
Breast length from base to nipple	30 cm	33 cm
Breast length from underarm to the tip of nipple	47 cm	50 cm



Figure-1: Photograph showing the patient's breasts at presentation. Peau d'orange appearance of the skin. Incision site of the incisional biopsy can be seen on the right breast.

DISCUSSION

The occurrence of gigantomastia during pregnancy and puberty has given support to the theory that hormones such as oestrogen and prolactin can be implicated in the pathogenesis. This also explains the response to bromocriptine. Increased risk in multiparous women is poorly understood.⁵ Some authors have suggested that increased receptor sensitivity in the presence of normal hormone levels may be one explanation. Several cases have been described in association with autoimmune diseases.

The major challenge in treating this case was pain control in the presence of a live foetus. Paracetamol was tried in the maximum possible dose without much benefit while NSAIDs and tramadol were withheld to prevent possible foetal complications. It was difficult to ambulate the patient and it was uncomfortable for her to lie down in any position. The early gestational period coupled with the severe pain demanded a more rapid solution. A discussion with the patient and her husband ensued and they decided that they would terminate the pregnancy and the patient would be started on tamoxifen.

It is recommended to have an elective delivery (by induction of labour if baby is at the age of viability) if the condition is complicated by haemorrhage and necrosis of the breast, but in our case the severe pain was the deciding factor. Following elective termination of the pregnancy and bilateral tubal ligation, the patient's condition rapidly improved on the second day. It was decided to

withhold tamoxifen. The patient's breasts eventually reduced in size considerably and she was not followed up because of financial constraints.

It is interesting to note that removal of the foetus and the placenta led to rapid resolution. We believe that besides other hormones, progesterone and human placental lactogen may have a yet unidentified trophic role in these cases. On the other hand, since some cases of gigantomastia have been reported in the presence of autoimmune disease, we do not know whether autoimmune disease could have triggered the gigantomastia in this case. However, since the patient's condition resolved on termination of pregnancy, and she did not have any clinical features suggesting autoimmune disease, this is less likely.

It is recommended to follow the preliminary protocol described by Rezaei S *et al* in diagnosing and treating these patients.⁶ This protocol includes the following steps:

- 1 - Immediate workup for a patient presenting with unilateral or bilateral gigantomastia includes
 - a- standard prenatal labs;
 - b- CBC with differential;
 - c- serum chemistry panel;
 - d- liver function tests;
 - e- serum calcium and albumin levels;
 - f- hormone profile: oestrogen, progesterone, and prolactin.
- 2 - Send the following tests for anti-dsDNA, ANA, RF, anti-Smith, CCP, antithyroglobulin, and anti-TPO, in addition to ESR and CRP, to investigate possible concomitant autoimmune disorders.
- 3 - To evaluate malignancy, perform a breast biopsy. Any finding suspicious of malignancy should be followed up with an MRI of the head and CT scan of the thorax, abdomen, and pelvis to detect areas of metastasis.
- 4 - Surgical intervention is not recommended in uncomplicated cases of gestational gigantomastia due to potential foetal compromise.
- 5 - Delivery via caesarean section or induction of labour is recommended for pregnancies complicated by breast necrosis, maternal sepsis, or haemorrhage.
- 6 - Trial of postpartum bromocriptine (2.5 mg twice daily) with cessation of breastfeeding is recommended to possibly reduce breast size before surgical intervention. If bromocriptine is used during pregnancy, serial foetal growth monitoring is recommended.
- 7 - Final surgical intervention with bilateral total mastectomy instead of reduction mammoplasty or simple mastectomy is recommended, especially in women desiring future pregnancy. There is an increased rate of recurrence in patients who underwent simple mastectomy or reduction

mammoplasty. Total bilateral mastectomy is the preferred surgical option.

CONCLUSION

Very few cases of gestational gigantomastia have been described in literature. It is important to understand the presentation and complete the full work up to avoid mistaking it for malignant conditions. Most cases of gigantomastia resolve spontaneously in the postpartum period, as was the case with our patient.

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Address for Correspondence:

Dr. Syed Sarmad Bukhari MBBS, Department of Neurosurgery, Aga Khan University Hospital, Karachi-Pakistan

Cell: +92 331 883 2628

Email: sarmad.bukhari@aku.edu