

A Rare “Big” Problem: A Case Study of Gestational Gigantomastia

ABSTRACT

Aim: Gestational gigantomastia (GG) is an extremely rare condition, with an estimated global incidence rate of approximately 1 in 100,000. This case report aims to examine possible aetiologies of GG and discuss management strategies, with a focus on symptom relief and achieving a safe pregnancy to full term.

Presentation of case: We report a case of a 32-year old female at 37-weeks of gestation who presented with rapidly increasing bilateral breast enlargement accompanied with severe back pain, resulting in significant functional impairment. Breast ultrasonography showed heterogenous breast parenchyma with a well-defined heterogenous breast lesion in the right breast. On examination, both breasts were grossly enlarged, tense, tender, with excoriation of the infra-mammary folds. Foetal assessment showed intrauterine growth retardation (IUGR), however amniotic fluid index (AFI) was within normal range. The initial management was for early elective delivery. However, the patient entered spontaneous labour and delivered a healthy baby boy with normal birth weight. During the immediate post-partum period, there was marked reduction in size of both breasts with improved symptoms.

Discussion: The aetio-pathology of GG is not yet fully understood and is hypothesized to be linked to hormonal imbalance, auto-immune diseases or malignancy. Management should be focused on symptomatic relief, prevention of complications in order to carry the pregnancy to term safely and to prevent recurrence in future pregnancies.

Conclusion: Although GG poses minimal risk to the unborn foetus, it can significantly impact the mother’s physical and psycho-social well-being. Early referral to a specialised centre is crucial to effectively manage this rare condition, and optimise maternal and foetal outcomes.

Keywords: Gestational gigantomastia, gestational breast hypertrophy, enlarged breast in pregnancy, benign breast disorder, pregnancy

1. INTRODUCTION

Gigantomastia is a rare condition characterized by diffuse and excessive breast growth⁶, while gestational gigantomastia (GG) or gravidic macromastia is a subtype of gigantomastia that arises in pregnancy, typically during the first or second trimester¹. It is a disorder that is characterized by diffused, extreme and incapacitating enlargement of one or both breasts during pregnancy¹; and is often accompanied with enlarged nipple and areolar size, and prominent superficial veins over the anterior chest⁶. It is not known to harm the developing foetus but can have a significant impact on the mother’s physical and psycho-social well-being³⁻⁵. We report a case of GG who presented to us at 37 weeks of gestation with bilateral breast enlargement in pregnancy.

2. CASE PRESENTATION

A 32-year old Indonesian female gravida 3 para 2 at 37 weeks of gestation presented with rapidly increasing bilateral breast enlargement for the past 8 months. She also complained of associated bilateral breast discomfort and severe back pain which had significantly limited her mobility and activities of daily living (ADL). She had initially presented to a primary healthcare clinic at 19 weeks of gestation with bilateral breast enlargement since the third week of gestation. Her pre-morbid bra size was a cup C, but as both her breasts rapidly increased in size, she was unable to wear any supportive

32 undergarments. The increase in weight of her excessively large breasts had caused further strain to her back and
33 rendering her mostly bed-bound. She also complained of pain over the infra-mammary folds due to the increase in skin
34 friction and felt social discomfort and self-consciousness to go out in public due to her grossly enlarged breast size.
35 Besides that, she did not have fever or nipple discharge, and there was no loss of weight or loss of appetite. Otherwise,
36 there was no family history of malignancy. She is an unemployed foreigner from Indonesia, married to her husband who is
37 of Indian nationality and works earning a minimum-wage. Thus, she is subjected to foreigner hospital fees which is much
38 more costly. This is her first pregnancy of her second union, with no prior history of abnormal breast growth or other
39 complications in her previous two pregnancies. Her last childbirth was more than 8 years ago. She achieved menarche at
40 14 years old and has regular menstrual cycle. She had previously used hormonal contraception for four years and
41 practiced turmeric liquid consumption as part of traditional Indonesian practice, known as "jamu". Turmeric-based jamu is
42 deeply rooted in Indonesian culture and is believed to promote overall health. They are sold commercially in health stores
43 and supermarkets, and are commonly taken by Indonesian women as part of their daily routine¹¹. A provisional diagnosis
44 of Phyllodes breast tumour was made and the patient was given an outpatient appointment to the Breast & Endocrine
45 Clinic. However, she did not attend her appointment due to financial constraints as she could not afford the clinic
46 consultation fee. She finally presented again due to the worsening symptoms and her concern to her pregnancy.

47 Upon examination, she was alert, clinically pink with good pulse volume and hydration. Vital signs were normotensive, not
48 tachycardic and afebrile. Her bilateral breasts were grossly enlarged, extending beyond the level of her umbilicus. There
49 was peau d'orange skin appearance and dilated superficial veins over the anterior chest (Figures 1-3). Both breasts were
50 tense and tender on palpation. There were no breast or axillary lumps palpable. Examination of the inferior aspect of
51 bilateral breast revealed superficial excoriation of the infra-mammary folds, but there were no skin ulceration or wounds.
52 No other lymph nodes were palpable.

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57 *Figures 1-3: Bilateral breast swelling with dilated superficial veins at 37 of weeks gestation. (1) Bilateral breasts, anterior*
58 *view. (2) Right breast, medial view. (3) Left breast, medial view*

59 Full blood count, renal profile and electrolytes were all within normal range. Breast ultrasonography demonstrated diffused
60 heterogenous hyperechogenicity of bilateral breast parenchyma, with a well-circumscribed hypoechoic lesion with edge
61 shadowing at the upper inner quadrant of right breast, measuring 3.3 x 4.4 x 3.4cm (AP x W x CC). Rest of breasts
62 showed evidence of heterogenous breast parenchyma echotexture bilaterally. There were no enlarged axillary lymph
63 nodes bilaterally. Foetal assessment showed evidence of intrauterine growth retardation (IUGR), but the amniotic fluid
64 index (AFI) and abdominal circumference were within normal range. She was advised for further hormonal testing and fine
65 needle aspiration cytology of the right breast lump, however refused due to the financial concerns on the cost of the
66 laboratory investigation.

67 She was admitted into the Obstetrics ward for pain management and was planned for early delivery via induction of
68 labour. However, she entered spontaneous labour spontaneous at 38 weeks of gestation and delivered a healthy baby
69 boy vaginally, with a birth weight of 2.3kg. At two days post-partum, there was a significant change in the appearance of
70 the bilateral breasts (Figures 4-6). Both breasts were less tense and had reduced in size. The previously-seen dilated
71 superficial veins over her anterior chest had resolved. She had reduced breast pain, significantly less back pain marked
72 and was able to ambulate with ease and carry her newborn baby. She and her baby were both healthy and was able to be
73 discharged home three days post-partum.

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77 *Figures 4-6: At post-partum Day 2, bilateral breasts appeared less tense with significant reduction in size and resolved*
78 *dilated superficial veins over overlying skin. (4) Bilateral breasts, anterior view. (5) Right breast, lateral view. (6) Left*
79 *breast, lateral view*

80 Medical therapy was not offered as she had presented late in pregnancy. A follow-up review was planned for her in the
81 post-partum period to assess her symptoms and the need for surgery. However, she did not attend her appointment due
82 to financial constraints. Attempts to contact her were futile as her contact number and address given were incorrect.
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84 3. DISCUSSION

85
86 Gigantomastia can be classified into 4 subtypes based on aetiology: juvenile, idiopathic, gestational and drug-induced². It
87 is a rare condition with an incidence rate of 1 in 100,000 globally⁶. It was first reported by Palmuth in 1684¹² with only 50
88 published case reports from 1976-2016. Demographic distribution shows that gestational breast hypertrophy is more
89 prevalent in North America, followed by South East and Central Asia¹. Cases have been reported in patients at the ages
90 of 16-35 years, with the majority presenting at 26-30 years old^{1,6-8}, which correlates to our patient's age.

91 The aetio-pathology of this condition is not yet fully understood. However, it has been hypothesized to be associated with
92 systemic disorders such as hormonal imbalance, auto-immune diseases or malignancy⁹. Published case reports have
93 linked GG to hyperprolactinaemia, hypercalcaemia, deranged liver function, myasthenia gravis, anti-phospholipid
94 syndrome and lymphoma (Hodgkin's and non-Hodgkin's lymphoma and T-cell lymphoblastic lymphoma)^{1,10}. Another
95 theory that has been proposed is related to hormone receptor hypersensitivity or excess in circulating hormones,
96 associated to hormonal changes during pregnancy, such as oestrogen, progesterone, prolactin, testosterone and
97 cortisol^{1,2,7}. When unrelated to underlying malignancy, GG is a benign condition with no known threat to the foetus in-
98 utero. However, it carries a significant social, emotional and physical disability to the expectant mother. The excess weight
99 and size of breast can remarkably limit their movement, causing significant pain and ulcerations. If poorly treated, infected
100 wounds may lead to sepsis and death^{1,8-10}. Our patient was not known to have autoimmune diseases or malignancy,
101 therefore hormonal abnormalities could have been a possible cause. Unfortunately, this could not be proven as she
102 refused not further laboratory testing due to her financial constraints.

103 During the intra-partum period, her foetus showed evidence of IUGR and was born with a birth weight below the 10th
104 centile¹³. However, he was healthy without any congenital anomalies, and did not require any special neonatal care. In
105 contrast, the patient had suffered great physical and social disability due to the weight of her rapidly-growing breasts that
106 had made her a social recluse and severely limiting her daily activities. Fortunately, she did not have any ulcerations or
107 wounds that could have led to infections, though there were excoriation of the inferior mammary folds as a result of
108 continuous friction.

109 Management of this condition is focused on symptomatic relief, prevention of complications in order to carry the
110 pregnancy to term safely and to prevent recurrence in future pregnancies. The first line of care is medical therapy with the
111 use of bromocriptine; a D2-agonist that arrests further hypertrophy of breast tissue¹. It is safe to use in the gestational
112 period and has not been linked to abortion or congenital anomalies¹⁴. However, there have been isolated cases that report
113 IUGR, such as in our case, therefore foetal growth should be serially monitored on a regular basis¹⁰. Other options include
114 pain relief with analgesics and the management of any wounds, if present. Elective termination of a viable foetus is not
115 ethical nor indicated as this condition rarely affects the foetus and this does not guarantee a cure.

116 Most cases do not spontaneously resolve after delivery thus, surgical treatment is offered in the post-partum period as
117 definitive therapy to prevent recurrence in future pregnancies¹. A multi-disciplinary approach is required to postpone
118 surgery as late as possible until the foetus is viable with matured lungs to avoid complications. An earlier elective delivery
119 may be planned with the administration of corticosteroid for foetal lung maturity if urgently indicated. Options include
120 reduction mammoplasty and mastectomy with or without breast reconstruction^{1,15}. Mammoplasty gives the patient the
121 choice for breastfeeding in the post-partum, which is essential in developing countries. However, it does not eliminate the
122 risk of recurrence as the remaining breast tissue may undergo hyperplasia in subsequent pregnancies. For patients who
123 have a desire for future pregnancies, bilateral mastectomy and breast reconstruction can be considered¹⁵.

124 Fortunately, her symptoms showed spontaneous improvement almost immediately after delivery. She did not require
125 medical therapy and was able to deliver safely a healthy baby without major complications throughout pregnancy.
126 However, the challenge we encountered with our patient is compliancy to seeking medical attention due to her financial
127 difficulties. Her symptoms had appeared at 2 weeks of gestation, but she presented at 19-weeks of gestation to a primary
128 healthcare clinic, and then defaulted until 37 weeks of gestation to a tertiary hospital. If she had presented earlier, medical
129 therapy could have been initiated to help reduce her symptoms. Proper work-up could also have been done to determine
130 the pathological cause and prevent her condition from worsening, or recurring in the future. We are also unable to

131 determine the long-term outcome for her due to lost in follow-up; whether her condition had completely resolve and, or if, it
132 recurs in future pregnancies.

133 4. CONCLUSION

134 More awareness must be made of this rare condition amongst primary healthcare doctors and obstetricians for early
135 referral to a specialised centre, as it requires a multi-disciplinary management. Further research is needed to better
136 understand and treat this condition to attain optimal foeto-maternal outcome.

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144 COMPETING INTERESTS

145 All authors declare that there are no competing interests.

148 AUTHORS' CONTRIBUTIONS

149 Author A performed history-taking and examination on the subject matter, managed literature review and wrote the
150 manuscript. Author B proof-read, edited and approved the manuscript.

153 CONSENT (WHERE EVER APPLICABLE)

154 All authors declare that informed consent was obtained from the patient for publication of this case report and
155 accompanying images.

158 ETHICAL APPROVAL (WHERE EVER APPLICABLE)

159 All authors hereby declare that all measures taken for the completion of this case report are in accordance to the 1964
160 Declaration of Helsinki.

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