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CASE REPORT

Idiopathic Gestational Gigantomastia: A case report

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ABSTRACT

Background: Gestational gigantomastia (GGM) is described as a diffuse and rapid enlargement of the breasts during pregnancy. It is a rare condition with an incidence of 1 in 28,000 to 1 in 100,000 pregnancies worldwide.

Case presentation: We present a case of a 29-year-old lady, G_2P_1 at 15 weeks gestation who presented with 2-months history of painful bilateral breast swelling. Clinical examination revealed erythematous bilateral breast enlargement. Results of multiple breast biopsies range from lactational adenoma and acute on chronic mastitis changes. She was treated with multiple courses of antibiotics for bilateral cellulitis with mastitis, however both breasts continued to enlarge. The revised diagnosis of gestational gigantomastia was made and she was started on steroid. Nevertheless, she failed to respond. Oral Bromocriptine commenced at 29 weeks showing some reduction in her breasts size. The fetus was however found to have a growth restriction requiring delivery at 37 weeks. Her breast has reduced by half of the initial volume during postnatal review. She was offered bilateral reductive mammoplasty and mastopexy, but she was not keen and opted for conservative management.

Conclusion: A thorough investigation is necessary to rule out other causes in women presenting with gigantomastia in pregnancy. GGM treatment ranges from conservative, hormonal therapy, reduction mammoplasty, mastectomy with or without reconstruction. This depends largely on the severity of disease and the patient's wish.

Keywords: gestational gigantomastia, mastitis, breast reduction, mastopexy.

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Introduction

Gestational gigantomastia (GGM) is characterized by a rapid excessive and disproportionate enlargement of the breasts during pregnancy. There is no objective definition of GGM worldwide from literature search, some suggest the quantification of GGM is based on the weight of removed breast tissue when surgery is performed, however this was not widely accepted[1,2]. It is occasionally associated with ulceration, infection, and necrosis of the overlying skin. It is a rare condition with an incidence of 1 in 28,000 to 1 in 100,000 pregnancies worldwide[3]. The etiology of GGM is still uncertain with many proposed theories. GGM has been reported to be associated with a response of breast receptors to gestational hormones and with hyperprolactinemia. It causes physical and psychological problems that severely affect the patient's quality of life. A thorough workup including serum markers for infection, electrolytes, hormonal profile and tissue biopsy should be done in order to rule out other causes in women presenting with gigantomastia in pregnancy. Treatment ranges from conservative hormonal therapy, reduction mammoplasty, mastectomy with or without reconstruction.

Case report

We present a case of a 29-year-old lady, G_2P_1 at 15 weeks gestation presented with 2-months history of painful bilateral breast swelling for the past 5 weeks. She was breastfeeding her first child when she embarked into this pregnancy and has stopped breastfeeding once this pregnancy was confirmed at 10 weeks gestation. She reported her original breast size was a B cup and denied similar problems during her first pregnancy. Her obstetric history includes previous caesarean section for dysfunctional labor at term.

Her height was 155 cm with a weight of 47 kg, making her BMI 19.56 kg/m^2 during her first presentation. Clinical examination revealed non tender erythematous bilateral breast enlargement (Fig. 1), in which she required a D cup size. She was initially treated with multiple courses of antibiotics for bilateral cellulitis with mastitis. Ultrasonography of the breasts did not reveal any underlying suspicious lesions. During her second visit at 18 weeks, her breasts continued to enlarge (Fig. 2) and became more inflamed despite multiple courses of antibiotics. Hence, the breast surgeon decided to perform biopsy to exclude inflammatory breast carcinoma. As there was no specific target or focus lesion identified on ultrasound, several punch biopsies were taken randomly from both inflamed looking breasts ensuring the whole thickness of dermis and subcutaneous layer were captured. Fortunately, the biopsies results range from lactational adenoma and acute on chronic mastitis changes. Blood parameters and hormonal profile, including estrogen, progesterone and prolactin levels were within normal limit of pregnancy values. The pain and erythema improved with the antibiotics, however both breasts continued to enlarge excessively throughout the pregnancy, in which she could not even wear a bra of her suitable size from 22 weeks gestation onwards (Fig. 3). This rapid growth resulted in back pain and difficulty in movement. The revised diagnosis of gestational gigantomastia was made and she was started on steroid. Nevertheless, she failed to respond. Oral bromocriptine 2.5 mg once daily was commenced at 29 weeks and there was some reduction in her breast size, hence it was continued.

During the treatment course, the fetus was found to have asymmetrical fetal growth restriction (FGR) at 34 weeks with normal amniotic fluid volume and Doppler study. She was otherwise normotensive, with no other risk factor for FGR. She subsequently delivered a healthy female baby weighing 2,100 grams at 37 weeks gestation via caesarean section. She declined the continuation of bromocriptine as she was keen to breastfeed her baby. Follow-up at 6 weeks postpartum showed her breast volume has decreased to about half of the enlarged volume and has become severely ptotic (Fig. 4). Unfortunately, due to the still excessively enlarged breast, she was unable to breastfeed her baby. Review at 3 months postpartum showed her breast size has reduced to D cup size but

remained ptotic. She was counselled for bilateral reductive mammoplasty and mastopexy by the breast surgeon but she declined as she was still able to endure her symptoms.

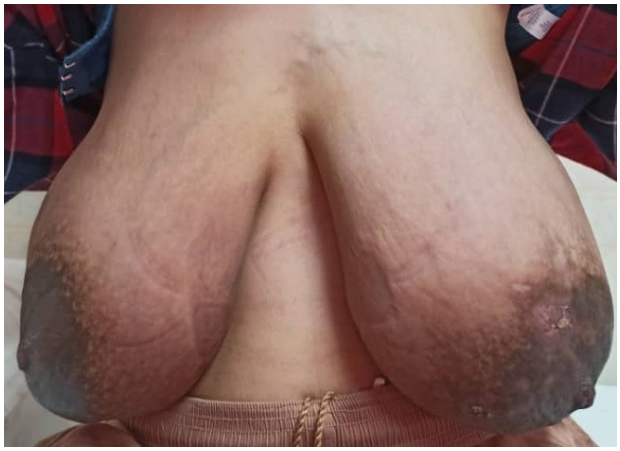

Fig. 1. Photograph showing bilateral gigantomastia at 15 weeks (initial presentation).

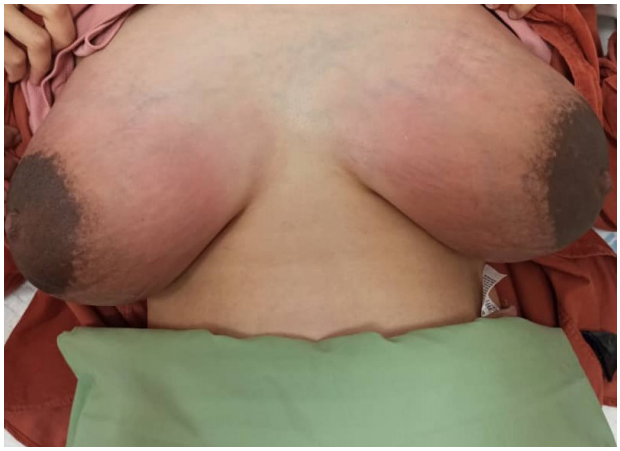

Fig. 2. Photograph showing gestational gigantomastia complicated with cellulitis at 18 weeks.

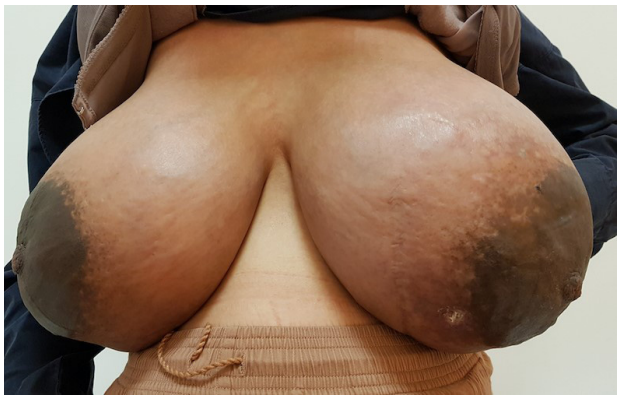

Fig. 3. Photograph showing bilateral breasts continue to enlarge with left breast ulceration at 22 weeks.

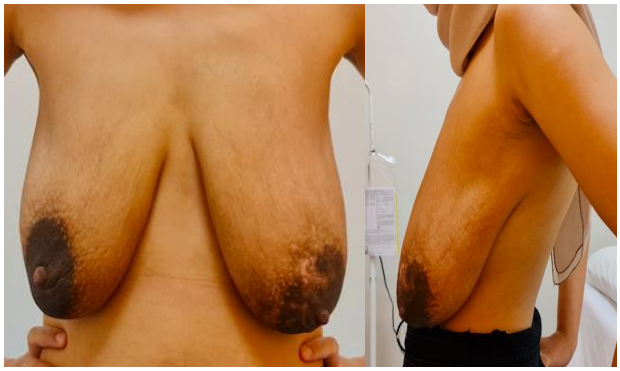

Fig. 4. Photograph showing reduction in size of bilateral breasts at 6 weeks postpartum.

Discussion

GGM has been documented to have associations with several conditions including hormonal imbalances such as hyperprolactinemia, hypercalcemia, deranged liver function tests, autoimmune diseases like systemic lupus erythematosus, and underlying malignancy[5,6,7]. Certain medications have also been reported to have associations with GGM such as prednisolone, D-penicillamine and cortisone[8,9,10]. However, the mechanism of action on how these drugs causes GGM remains unclear. Nevertheless, there have been limited cases which reported such associations; hence it may not be a direct causal relationship. Therefore,

the workup for GGM should focus on identifying these potential associations. Basic laboratory investigations including full blood count to look for white cell count and systemic inflammatory markers (erythrocyte sedimentation rate and C-reactive protein), estrogen, progesterone, prolactin and testosterone level should ideally be evaluated[6]. In addition to the laboratory tests, breast ultrasound scan or magnetic resonance imaging (MRI), and breast biopsies may be obtained to exclude underlying malignancy as performed in this case[6]. Other disease processes must be considered before making a diagnosis of benign GGM. This comprises of both benign conditions; infectious mastitis, juvenile breast hypertrophy, fibrocystic change or fibroadenoma and/or normal pregnancy-related breast enlargement and malignancy. Rapid breast enlargement with axillary swelling may mimic malignancy. Other signs such as oedema and peau d'orange skin changes consistent with inflammatory carcinoma need to be excluded via cytomorphological evaluation[3,4].

GGM, although benign, can be a significantly debilitating disease. It does not only result in physical complications if left untreated but also affects emotional and social incapacity. It is important to recognize it early to ensure treatment can be started as soon as possible to prevent future complications. Conservative measures include suitable brassiere support, proper skin hygiene, and analgesia[4,5]. During pregnancy, medical management is often preferred over surgical management due to the risk of fetal harm, although the latter may be pursued in the case of massive hemorrhage, ulceration or sepsis[4,5,11]. Women diagnosed with GGM should ideally be seen more frequently by both the obstetricians and breast surgeons to identify for any ulceration, necrosis or hemorrhage. Fatal cases of hemorrhage and sepsis from GGM due to partially treated ulcers have been reported[7,8,12].

Bromocriptine, a dopaminergic agonist, has been the medication of choice for GGM[3,5,13]. It has been shown to reduce breast size and suppress lactation while allowing surgical intervention if desired[3,5,10]. Bromocriptine is found to be safe in pregnancy with no teratogenic risk to the fetus. It has been safely used in pregnant women with pituitary prolactinoma[9]. Nevertheless, maternal blood pressure and fetal growth require close monitoring due to hypothetical risk of hypertension and fetal growth restriction as described in this case. Ideally, bromocriptine is continued throughout pregnancy and in the postpartum period to reduce breast size, to allow for surgical intervention when preferred[3,10]. Surgical approaches, including reduction mammoplasty and mastectomy are warranted when medical treatment fails, or patient develops debilitating symptoms[4,6,13]. Since there is a possibility of recurrence with simple mastectomy or reduction mammoplasty, bilateral mastectomy with reconstruction may be the treatment of choice in women who desire future pregnancies[4,6].

Conclusion

GGM is a rare disorder with many potential complications, all of which may lead to a significant reduction in the quality of life. Other various benign and malignant diseases should be ruled out through endocrinology and histopathologic evaluation. Treatment is often pharmacological throughout pregnancy and in the early postpartum period, although the definitive treatment is surgical. Fetal growth should be monitored with bromocriptine therapy due to case reports describing fetal growth restriction. Knowledge on this rare condition is necessary especially for obstetricians, whom will be the first line seeing these women.

Potential conflicts of interest

The authors declare no conflicts of interest.

References

1. Mangla M, Singla D. Gestational gigantomastia: A systematic review of case reports. J Midlife Health 2017;8:40-4.
2. Okere UC, Margenthaler JA, Vanko S, Kennard K.

A multi-disciplinary approach to gestational gigantomastia management: a case report. AME Surg J 2023;3:47.
3. Pleasant V, Chapel E, Momoh A, Manorot A, Carver A. Gestational gigantomastia: Case report and review of treatment options. Ann Case Rep 2024;9:1700.
4. Jeong HY, Kang T, Park H, Kim KE, Nam SB, Go JY, Bae SH. Management of gestational gigantomastia with goldilocks procedure after mastectomy: A case report and review of literature. Arch Plast Surg 2024;7;51:62-6.
5. Mohan V, Singla A, Agarwal R. Gestational gigantomastia: The sentinel event to systemic lupus erythematosus. Obstetric Medicine 2023;1:1-3.
6. Rezai S, Nakagawa JT, Tedesco J, Chadee A, Gottimukkala S, Mercado R, Henderson CE. Gestational gigantomastia complicating pregnancy: A case report and review of the literature. Case Rep Obstet Gynecol 2015;10:892369.
7. Biancha-Vasco JM, Muñoz P AL, Gómez J, Rios-Serna LJ, Ruiz-Ordoñez I, Tobón GJ, et al. Idiopathic gigantomastia exacerbated during pregnancy. Its relationship with autoimmunity: A case report. Int J Rheum Dis 2023;26:2567-71.
8. Mori T, Yokogawa N, Higuchi R, Tsujino M, Shimada K. Bucillamine-induced gigantomastia with galactorrhea and hyperprolactinaemia. Modern Rheumatol Case Rep 2020; 4:122–5.
9. Jabaiti S, Fayyad L, Isleem U. Prednisolone-induced virginal mammary hypertrophy: Case report. Int J Surg Case Rep 2019;59:140–3.
10. Fletcher MB, Corsini LM, Meyer MD, Osswald SS. Gestational gigantomastia: A case report and brief review of the literature. JAAD Case Rep 2020;21;6:1159-61.
11. Neblett C, Venugopal R, Johnson M, Mitchell D, Kunc V. Gestational gigantomastia- a rare entity complicated by life-threatening haemorrhage. J Surg Case Rep 2021; 4: rjab050.
12. Rakislova N, Lovane L, Fernandes F. Gestational gigantomastia with fatal outcome. Autops Case Rep 2020;10:e2020213.
13. Alhindi N, Mortada H, Alzaid W, Al Qurashi AA, Awan B. A systematic literature review of the clinical presentation, management, and outcome of gestational gigantomastia in the 21st Century. Aesthetic Plast Surg 2023;47:10-29.