

Gestational Gigantomastia (Spontaneous Resolution after Delivery) a Rare Case Report

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Abstract: Gestational gigantomastia (GG) is a very rare type of gigantomastia, occurring during pregnancy. Less than 100 cases have been reported in literature till date. Here we are reporting a 25-years old female secondgravid, Presenting as gigantomastia in the 2nd trimester. She noticed sudden enlargement of bilateral breast in her 26th week of gestation. (LMP: 6 Apr 2018) G2P1+0+0+1. This enlargement started at 12 weeks of gestation and continued to increase rapidly as the pregnancy progressed and complicated by infection, skin atrophy, marked venous engorgement, ulcerations, necrosis and subsequent hemorrhage. On examination, both breasts were extremely enlarged & she was not able to walk or even sit up properly. Hormonal studies were found to be within the normal range. Emergency LSCS was done by gynae team at 36th week POG & a single live female baby of 2kg was delivered. The gigantomastia subsided post- delivery spontaneously.

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I. Introduction

GG is a rare breast condition characterized by rapid, diffuse, and excessive enlargement of one or both breasts during pregnancy (> 1.5 kg/ breast).¹ the definition of this rare disease is although not clear, Lewison et al used beautiful words to describe this typical cases, "True gigantomastia develop rapidly during pregnancy, undergoes regression after delivery, and recurs with subsequent pregnancies."² Less than 100 cases of gestational gigantomastia have been described in literature till the date.³ The term gigantomastia was first introduced by Palmuth in German literature in 1648.⁴ It was not introduced into English literature until centuries later, when Simpson used it in 1920.⁵ According to etiology gigantomastia, can be subclassified into four types (a) Puberty induced or juvenile gigantomastia (b) Gravid (gestational) gigantomastia (c) Drug-induced gigantomastia (d) Idiopathic gigantomastia.⁶ Gestational gigantomastia is listed as a "rare disease" by the Office of Rare Diseases (ORD) of the National Institutes of Health (NIH).⁷ Caucasian women are more likely to be affected than black women, with a ratio of 9:4.⁸⁻⁹ Although only rare cases have been reported in literature that underwent complete spontaneous resolution after pregnancy, majority of cases need either medical or surgical treatment. It is a much bigger problem in developing countries where the importance of breast feeding for the newborn child cannot be underrated.

II. Case Report

A 25-years-old woman, second gravida presented at 26 weeks of gestation, (LMP: 6 Apr 2018) G2P1+0+0+1 with marked bilateral breast enlargement. This enlargement started at 12 weeks of gestation and continued to increase rapidly as the pregnancy progressed and was complicated by infection, skin atrophy, marked venous engorgement, ulcerations, necrosis and subsequent hemorrhage. She reported that her previous pregnancy (4 years back) was successful and she did not have similar problem. She was initially seen by a local doctor who referred her to RIMS surgery department. Her past medical history and family history were insignificant. During her hospitalization breasts continued to increase in size (bra size passed from fifth to tenth measure), with skin atrophy, necrosis and multiple ulcers measuring 2-5 cm in diameter (Figure 1-2). The patient was complaining of mastalgia, severe back pain, breathing difficulties, inability to do routine work and pain near ulcerations. She was also complaining of sudden increase in her weight during this pregnancy from 54 kg to 70 kg within 4 months. Patient was admitted in our general surgery ward and was managed conservatively with daily dressing for ulcer under full sterile precautions, oral antibiotic coverage, iron and calcium tablet. Regular fetal wellbeing was monitoring by USG Doppler and regular Obs & Gynae consultations were done during the admission.

On examination, both breasts were extremely enlarged, she was not able to sit or walk due to heavy weight of breasts. The right breast was slightly larger than the left breast and both nipples exuded serous fluid. The skin covering the breasts showed marked pigmentation, dilated veins, and multiple ulcers. Breast ultrasonography was normal other than enlarged size. The patient underwent full laboratory investigations,

including complete blood count, liver function tests, renal function tests, hormonal assay (estrogen, progesterone, prolactin, testosterone, follicle stimulating hormone, luteinizing hormone, thyroid stimulating hormone, T3, T4, growth hormone, and insulin), lipid profile, immunological assay (ANA, anti-dsDNA, ASMA, AMA, antiphospholipid IgM, C3), serum calcium, tumor markers (CEA, CA15.3, AFP, CA19-9) and virology markers (HCV, HBV, HIV). All the results for these investigations were within normal limit.

After these preliminary investigations, an excision biopsy was performed, which included part of the ulcerated skin. Results of pathological examination revealed adenosis, moderated epitheliosis, fibrosis, and stromal lymphocytic infiltration.



Fig 1- breast size at the time of presentation



Fig 2- multiple ulceration over breast



Fig 3- size of breast 1 week after delivery



Fig 4- size of breast after 3 months of delivery

III. Discussion

GG is a rare condition in which the breasts attain huge size during pregnancy. In some instances, such benign condition can turn out to be a life-threatening condition. GG was first introduced by Palmuth in a German literature in 1648. To date only around 125 cases have been reported in literature. The incidence of gigantomastia occurring in pregnancy is 1:28,000–1:100,000.

The exact pathogenesis of this condition is still not understood. Many hormones are assumed to play a role in this process, for example, estrogen, progesterone, prolactin, testosterone, and cortisone.⁸ Among these hormones, the dramatic increase of the estrogen plasma level during pregnancy appears to be the main contributor in triggering gestational gigantomastia.¹⁰ Another proposed cause of gestational gigantomastia is the associated rise in the serum prolactin level during pregnancy. Lafreniere et al¹¹ showed high serum prolactin levels in GG. Bromocriptine which is a dopamine agonist that inhibits the release of prolactin from the pituitary, has been used on a wide scale to control cases of gestational gigantomastia. But in our case serum prolactin level was within normal limit (16.15 ng/ml).

Touraine et al¹² reported 8 cases of gigantomastia in a context of autoimmune diseases, for example, myasthenia gravis, systemic lupus erythematosus, Grave's disease, and rheumatoid arthritis. Two of these cases occurred during pregnancy. These authors claimed that GG could be an inflammatory breast hypertrophy condition aggravated by hormonal changes associated with pregnancy. The authors, however, were not able to detect organ-specific autoantibodies in breast tissue sections and thus failed to prove on a scientific basis that breast tissue is targeted by these autoimmune diseases. In our case immunological assay (ANA, anti-dsDNA,

ASMA, AMA, antiphospholipid IgM, C3) were found to be normal. In the differential diagnosis a phyllodes tumor, fibroadenoma, Non-Hodgkin's lymphoma and lymphoblastic lymphoma were excluded through biopsy.

Reduction mammoplasty and simple mastectomy with delayed reconstruction are the available current surgical options. Wolf et al¹³ stood against reduction mammoplasty and mastectomy as methods of treatment in gravid gigantomastia cases and proposed elective termination of the pregnancy as an alternative treatment. Here in our case patient's condition improved spontaneously after delivery deviating the need for surgery.

In a study by Swelstad et al¹⁴ reported that 100% of the patients (4 patients) condition relapsed when they were pregnant again after the breast reduction surgery for GG. Conservative treatment with a dopaminergic receptor agonist (bromocriptine) is the preferred option for the treatment. Even though it stops the further breast growth, but it has no effect on reducing the breast size.¹⁵

In our patient Breast size remained hugely enlarged during admission & complete healing of superficial ulceration did not occur despite regular dressing and treatment with medication. She remained bedridden during admission period. She also complained of difficulty in respiration with increasing breast size, though she maintained normal SpO₂ during this period. At 36 weeks of pregnancy she could no longer tolerate her condition in consultation with Gynaecologist, LSCS was done & she delivered a healthy 2kg female baby. The baby was managed initially in paediatric ICU unit and subsequently discharged in good health.

She came for regular follow up in surgery OPD and the post-delivery breast size gradually started decreasing in size and she was able to sit and walk around normally by 2nd week of postpartum. Skin engorgement also subsided gradually & ulceration healed completely by 3rd week of postpartum.

IV. Conclusion

GG is a rare disease occurring during pregnancy, which can result in great social, emotional & physical disability. It requires multidisciplinary team effort in the form of general surgeon, obstetrician, anaesthetist, & paediatrician for a successful foeto-maternal outcome.

References

- [1]. Zingaretti N, De Biasio F, Riccio M, Nardini N, Mariuzzi L, Parodi PC. A case of gestational gigantomastia in a 37-years-old woman associated with elevated ANA: a casual linkage?. *The Pan African medical journal*. 2017;27.
- [2]. Lewison EF, Jones GS, Trimble FH, da Lima LC. Gigantomastia complicating pregnancy. *Obstetrical & Gynecological Survey*. 1960 Jun 1;15(3):338-40.
- [3]. Bukhari SS, Manan H, Khan MM, Raza SS. Resolution of gestational gigantomastia with termination of pregnancy. *Journal of Ayub Medical College Abbottabad*. 2018 May 27;30(2):298-300.
- [4]. Palmuth T. Observations Medicuarum Centinae Tres Posthumae. Braunschweig; Cent II, Obs 1684:89.
- [5]. Simpson BS. Notes on a case of diffuse hypertrophy of the breasts. *Edinburgh Med*. 1920;24:176-179.
- [6]. Ship AG. Virginal and gravid mammary gigantism: recurrence after reduction mammoplasty. *Br J Plast Surg*. 1971;24: 396-401.
- [7]. Office of Rare Diseases. National Institute of Health. <http://rarediseases.info.nih.gov/GARD/Disease.aspx?PageID=4&diseaseID=9450>. Accessed July 16, 2008.
- [8]. Beischer NA, Hueston JH, Pepperell RJ. Massive hypertrophy of the breasts in pregnancy: report of 3 cases and review of the literature, "never think you have seen everything." *Obstet Gynecol Surv*. 1989;44:234-243.
- [9]. Swelstad M, Swelstad B, Rao V, Gutowski K. Management of gestational gigantomastia. *Plast Reconstr Surg*. 2006; 118:840-848.
- [10]. Krupp P, Monka C. Bromocriptine in pregnancy: safety aspects. *Klin Wochenschr*. 1987;65:823-827.
- [11]. Lafreniere R, Temple W, Ketcham A. Gestational macromastia. *Am J Surg*. 1984;148:413-418.
- [12]. Touraine N, Youssef M, Alyanakian X, et al. Breast inflammatory gigantomastia in a context of immune-mediated diseases. *J Clin Endocrinol Metab*. 2005;90:5287-5294.
- [13]. Wolf Y, Puzner D, Groutz A, Walman I, David MP. Gigantomastia complicating pregnancy: case report and review of the literature. *Acta Obstet Gynecol Scand*. 1995; 74:159-163.
- [14]. Swelstad MR, Swelstad BB, Rao VK, Gutowski KA. Management of gestational gigantomastia. *Plast Reconstr Surg*. 2006;118:840-848.
- [15]. Hedberg K, Karlson K, Lindstedt G. Gigantomastia during pregnancy: effect of a dopamine agonist. *Am J Obstet Gynecol*. 1979;133:928-931.

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