Gestational Gigantomastia

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ABSTRACT
Gestational gigantomastia is a rare condition characterized by fast, disproportionate and excessive breast growth, decreased quality of life in pregnancy, and presence of psychologic as well as physical complications. The etiology is not fully understood, although hormonal changes in pregnancy are considered responsible. Prolactin is the most important hormone. To date, 125 cases of gigantomastia have been reported in the literature. In this case presentation, we report a pregnant woman aged 26 years with a 22-week gestational age with gestational gigantomastia and review the diagnosis and treatment of this rare disease in relation with the literature.

Keywords: Breast, gigantomastia, hypertrophy, mastectomy

Introduction
Gigantomastia can be defined as excessive breast growth where 1500 gr or more tissue has to be removed from the breast (1). Gestational gigantomastia is exceptionally rare and occurs in 1 out of every 100 000 pregnancies (2).

Case Presentation
A pregnant woman aged 26 years with no apparent systemic disease and medication history who was 22 weeks pregnant was admitted to our breast surgery outpatient clinic because of rapid growth in both breasts, which caused back pain, and difficulty in movement. The patient was in her third pregnancy and had experienced breast growth within physiologic limits in her previous pregnancies. She noticed excessive and rapid breast growth after the 14th week of pregnancy; there was no family history of a similar condition. Physical examination findings were extreme growth in both breasts, distinct subcutaneous venous structures, and some necrotic areas on the skin (Figure 1). Additionally, the patient had back pain, difficulty in movement, and difficulty in meeting daily needs. The patient weighed 75 kg and was 165 cm in height, with a body mass index (BMI) of 28 kg/m². Breast ultrasonographic examination revealed diffuse hypoechoic areas with increased vascularity; there were no subcutaneous fat planes or solid/cystic masses in either breast. The findings from the preoperative laboratory investigation were as follows: Hemoglobin: 11.2 g/dL (normal: 11.5-15.02 g/dL), sedimentation rate (ESR): 68 mm/hr (normal 2-20 mm/hr), urea: 9 mg/dL (normal: 70-1009 mg/dL), Cre: 0.45 mg/dL (normal: 0.56-0.85 mg/dL), AST: 12 IU/L (normal: 11-25 IU/L), ALT: 6 IU/L (range: 7-28 IU/L), TSH: 2.73 mIU/Ml (normal: 0.35-4.94 mIU/Ml), and prolactin: 110 ng/Ml (normal: 1.2-29.9 ng/Ml). At the 24th gestational week, the patient was scheduled bilateral subcutaneous mastectomy and implant placement. However, the operation was finalized after completion of bilateral subcutaneous mastectomy due to acute hemorrhage causing hemodynamic instability and severe anemia (intraoperative hemoglobin; 5.7 11.2 g/dL). Therefore, the reconstruction was postponed to another session. The measurements of the excised tissue from the right and left breasts were 3750 gr and 3700 gr, respectively. On 6th postoperative day, surgical debridement was performed for necrosis that had developed on the left areola and parts of skin. The histopathologic examination of the specimen revealed marked lactation changes of the epithelial component and increased vascularization in the stroma. The patient’s follow-up went smoothly and she was discharged after post-natal reconstructive surgery was scheduled. The decision to presenting this case report was made after receiving written and oral consent from our patient.

Discussion and Conclusion
Gestational gigantomastia was first described in 1648 by Palmuth. Its etiology and pathogenesis are not well established; however, it is believed to be triggered by placental hormones. This hypothesis is supported by the fact that excessive increase in breast size is seen
most frequently during the first trimester when the highest amount of gonadotropin is produced (3). Prolactin hormone is the first of the hormones shown as a target in etiology. Additionally, other hormones such as progesterone, estrogen, thyroxine, growth hormone, cortisol, insulin and human placental lactogen are also considered to have an effect (4).

Lafreniere et al. (5) demonstrated that prolactin levels were high in this type of patient in their study. In our study, the prolactin level was 110 ng/mL (normal: 1.2-29.9 ng/mL). Furthermore, a patient with rheumatoid arthritis was reported to have gigantomastia due to D-penicillamine use in the etiology (6). Drugs such as cyclosporine and bucillamine are also blamed in the etiology. Moreover, Touraine et al. (7) stated that immunologic and hormonal reasons were effective in their study. It was proven that breast tissue was a potential target tissue in autoimmune diseases such as myasthenia gravis, chronic arthritis, and Hashimoto’s thyroiditis, and that an autoimmune mechanism was effective in the etiology of the disease and immunohistochemical analysis of breast tissues (7). In the differential diagnosis, a phyllodes tumor, fibroadenom, Non-Hodgkin’s lymphoma and lymphoblastic lymphoma can be excluded through biopsy. Having analyzed mastectomy samples of patients with gestational gigantomastia histologically, Swelstad et al. discovered significant lobular hypertrophy, ductal proliferation and periductal fibrosis (8). Furthermore, gestational gigantomastia can be accompanied by histologic alterations such as extensive lobular hyperplasia, dilated tracts, and pseudoangiomatous hyperplasia. Although the effects of the disease can be seen more frequently in multiparous women, there is no relationship between the disease and the number of pregnancies (2, 9, 10).

Patients with this disease might experience social and psychologic problems, as well as difficulty with movement and breathing.

Conservative treatment with bromocriptine, a dopaminergic receptor agonist, is the preferred option for the treatment. Even though it halts breast growth, it has no apparent effect on reducing breast size (11). Furthermore, tamoxifen, hydrocortisone, diuretics, and medroxyprogesterone are included in the conservative treatment. Breast-conserving surgery could cause relapse; therefore, mastectomy is recommended for patients with this disease (8). In a study by Swelstad (8), 100% of the patients (4 patients) who underwent breast reduction surgery for gestational gigantomastia relapsed when they were pregnant again after the operation. We also preferred mastectomy in consideration of possible relapses after breast reduction surgery.

Consequently, gestational gigantomastia may begin in any pregnancy and recur during following pregnancies. Hyperprolactinemia is a common condition in patients with gestational gigantomastia; however, it does not require the termination of the pregnancy (3). The best possible treatment option is total mastectomy. We believe potential problems that may arise should be considered and measures should be taken in order to cope with a possible venous lake and severe anemia due to hemorrhage during surgery.

Informed Consent: Written informed consent was obtained from patient.

Peer-review: Externally peer-reviewed.


Conflict of Interest: No conflict of interest was declared by the authors.

Financial Disclosure: The authors declared that this study has received no financial support.

References