Gestational gigantomastia: A case report and brief review of the literature



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INTRODUCTION

Gigantomastia is a rare condition characterized by diffuse and excessive breast growth.¹ Gestational gigantomastia (GG) is a subtype of gigantomastia in which the condition presents during pregnancy, typically in the first or early second trimester.^{2,3} The earliest reported case of GG was described by Palmuth in 1648, and it is reported to affect roughly 1 in 100,000 pregnancies.^{4,5}

GG typically presents as markedly disproportionate growth of either one or both breasts accompanied by grossly dilated nipples and areolae, prominent superficial veins, and occasionally ulceration, infection, and necrosis of the overlying skin. The affected breasts are firm and tender to palpation.¹

Various associations with GG have been documented, including hormonal imbalances such as hyperprolactinemia, hypercalcemia, deranged liver function tests (LFTs), autoimmune conditions, and underlying malignancy; therefore the workup is focused on identifying these potential associations.²⁻⁶

Treatment depends mainly on the severity of disease and can range from medical management with bromocriptine to surgical management with reduction mammoplasty or bilateral mastectomy.^{1,4,7}

CASE REPORT

A 31-year-old woman, gravida 5, para 4 at 18 weeks' gestation presented with a 4-month history of bilateral breast swelling, erythema, and pain (Fig 1). She reported her original breast size was a B cup, and she denied similar complications with prior pregnancies. Her breast swelling had previously been managed with dicloxacillin for suspected mastitis without improvement. Bilateral breast ultrasound scans were negative for fluid collections or Abbreviations used:

GG: gestational gigantomastia

LFT: liver function test

masses, and bilateral breast magnetic resonance imaging studies exhibited bilateral symmetric breast skin thickening suggestive of mastitis. The surgical oncology department performed bilateral breast incisional biopsies to rule out inflammatory breast carcinoma, and pathology findings showed chronic, dermal inflammation without evidence of malignancy. C-reactive protein, erythrocyte sedimentation rate, calcium, LFTs, and prolactin levels were normal. In the dermatology clinic, repeat biopsy findings of the right breast were consistent with lymphedema. Tissue cultures were negative.

The patient experienced worsening breast pain requiring inpatient admission. Prolactin, estradiol, progesterone, and thyroid-stimulating hormone levels were within normal limits. An initial autoimmune workup found a low positive antinuclear antibody titer of 1:40, making it unlikely to be relevant. Gestational gigantomastia was diagnosed, and the patient was started on bromocriptine, which was effective in decreasing galactorrhea, erythema, and tension of her breasts, but they remained significantly enlarged. Her course was complicated by spontaneous local hemorrhage in her left breast. To avoid further complications and because of severe pain associated with gestational gigantomastia, the patient was induced at term. Following a cesarean section, she gave birth to a healthy child who received routine postpartum care. The patient continued bromocriptine postpartum. Because she continued to have significant breast discomfort, she

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Fig 1. Gestational gigantomastia: Bilateral breast swelling with dilated superficial veins present. Healing incisional biopsy sites to bilateral, inferior breasts and punch biopsy site to right, superior breast visible.

discontinued the bromocriptine and underwent a breast reduction/mammoplasty. She reported that she went from a size M cup down to a D cup with significant relief secondary to the reduction in breast size.

DISCUSSION

Gigantomastia, a rare condition characterized by diffuse and excessive breast growth, can be subdivided into 4 major types based on etiology. These include idiopathic, juvenile (puberty associated), gestational, and drug induced (most associated with bucillamine and penicillamine administration).^{1,7,8} The pathogenesis of juvenile gigantomastia and GG is still not entirely elucidated. The postulated cause is related to hormonal changes surrounding puberty and pregnancy, which can come in the form of either hormonal excess or hypersensitivity of the breast tissue to normal levels of circulating hormones.^{8,9} The hormones likely implicated are estrogen, progesterone, prolactin, testosterone, and cortisol, and consideration may be given to thyroxine, growth hormone, insulin, and human placental lactogen.^{7,9} Specifically, with regard to GG, prolactin is the primary hormone to show potential as a target for therapy.^{2,7}

In addition to hormonal abnormalities, like hyperprolactinemia, other laboratory abnormalities can be associated with GG, and, as such, an appropriate workup should be considered. Hypercalcemia has been attributed to excessive production of parathyroid hormone—related protein of mammary origin, although the reason for this abnormality is unknown.³ Deranged LFTs have also been reported in conjunction with GG although the origin is not fully understood.^{2,4} Autoimmune serologies may show abnormalities, as certain autoimmune conditions, including myasthenia gravis, systemic lupus erythematosus, rheumatoid arthritis, and autoimmune thyroiditis, have been reported in patients presenting with GG.² Furthermore, workup should include a complete blood count with differential, serum chemistry panel, and systemic inflammatory markers (erythrocyte sedimentation rate and C-reactive protein). In addition to the aforementioned laboratory tests, breast ultrasound scan/magnetic resonance imaging, and breast biopsies may be obtained to investigate for underlying malignancy.⁶ Barring any malignancy, breast histology in patients with GG often shows lobular hypertrophy, ductal proliferation, abundant stromal tissue, acinar and periacinar stromal fibrosis, and occasionally lymphocytic infiltration.4,5,7

GG can be a significantly debilitating disease. Skin ulceration, necrosis, infection, and hemorrhage can occur and, more rarely, sepsis, multiorgan dysfunction, and even death.^{1,2} In light of this finding, it is important to recognize GG early on in the disease process to begin treatment if warranted. Conservative measures include proper brassiere support, appropriate skin hygiene, and analgesia.⁴ During pregnancy, medical management is preferred over surgical management because of the risk of fetal harm, although the latter may be pursued in the case of massive hemorrhage, ulceration, sepsis, or necrosis.⁴ If medical management is pursued, bromocriptine, a dopaminergic agonist, is the first-line medication choice.^{2, $\bar{4},7$} Although bromocriptine is safe to use during pregnancy, fetal growth should be monitored because of anecdotal evidence of fetal intrauterine growth retardation secondary to bromocriptine therapy.⁴ Ideally, bromocriptine is continued throughout pregnancy and in the post-partum period to suppress lactation and reduce breast size, to allow for surgical intervention if desired.⁴ There are 2 main surgical approaches, including reduction mammoplasty and mastectomy.^{2,3,9} Because of the high chance of recurrence of GG in subsequent pregnancies after treatment with reduction mammoplasty, it is recommended for patients planning on future pregnancies to undergo mastectomy.³ Every patient's treatment course should be managed with a multidisciplinary approach in conjunction with an obstetrician or maternal fetal medicine specialist.

GG is a rare disorder characterized by excessive breast growth with many possible complications, all of which may lead to a significant decline in quality of life. Underlying disease processes that may trigger or cause gigantomastia should be ruled out via assistance from oncology, endocrinology, and histopathologic evaluation. Treatment is often pharmacologic throughout pregnancy and in the early post-partum period, although the definitive treat-

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ment is surgical.

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