A Rare Case of Gestational Gigantomastia with Hypercalcemia: The Challenges of Management and Follow up

Bahram Moazzami 1, Shahla Chaichian 2*, Mohammad Reza Farahvash 3, Saeedeh Taheri 1, Seyed Ali Ahmadi 4, Majid Mokhtari 5, Kourosh Sheibani 6

1- Pars Advanced and Minimally Invasive Manners Research Center, Pars Hospital, Iran University of Medical Sciences, Tehran, Iran
2- Minimally Invasive Techniques Research Center in Women, Tehran Medical Sciences Branch, Islamic Azad University, Tehran, Iran
3- Department of Plastic Surgery, Faculty of Medicine, Tehran University of Medical Sciences, Tehran, Iran
4- Department of Pathology, Faculty of Medicine, Tehran University of Medical Sciences, Tehran, Iran
5- Department of Medicine, Pulmonary and Critical Care Medicine, Shahid Beheshti University of Medical Sciences, Tehran, Iran
6- Clinical Research and Development Center, Imam Hossein Medical Center, Shahid Beheshti University of Medical Sciences, Tehran, Iran

Abstract
Background: Gigantomastia is a breast disorder marked by exaggerated rapid growth of the breasts, generally bilaterally. Since this disorder is very rare and has been reported only in sparse case reports its etiology has yet to be fully established. Treatment is aimed at improving the clinical and psychological symptoms and reducing the treatment side effects; however, the best therapeutic option varies from case to case.

Case Presentation: The present report described a case of gestational gigantomastia in a 30-year-old woman, gravida 2, parity 1, 17 week pregnant admitted to Pars Hospital, Tehran, Iran on May 2014. The patient was admitted to hospital at week 17 of pregnancy, although her breasts initially had begun to enlarge from the first trimester. The patient developed hypercalcemia in her 32nd week of pregnancy. The present report followed this patient from diagnosis until the completion of treatment.

Conclusion: Although gestational gigantomastia is a rare condition, its timely prognosis and careful examination of some conditions like hyperprolactinemia and hypercalcemia is essential in successful management of this condition.

Keywords: Breast, Diagnosis, Gestational age, Gigantomastia, Hypercalcemia, Iran, Therapeutics.


Introduction

Gestational hypertrophy of the breast or gestational gigantomastia is a very rare clinical condition characterized by fast, unusual and painful breast growth during pregnancy first reported by Palmuth in 1648 (1, 2). Since then, only about one hundred cases have been reported in the literature (1, 2).

This condition can be accompanied by ulcerations, infections, and areas of local necrosis causing severe maternal morbidity and even mortality (1). The etiology of this condition is unknown but various theories including endocrine imbalance, hyperprolactinemia and target organ hypersensitivity have been suggested by different researchers (3, 4).

The choice of treatment for these patients is different based on the severity of each case and the time of pregnancy (5). Only few cases of complete spontaneous postpartum resolution among these patients have been reported and surgical management may be performed by breast reduction or by mastectomy and delayed reconstruction (1).

The objective of this paper was to describe a
case of a gravid 2 patient diagnosed with the dis-
order in the second trimester of pregnancy. All
procedures, included in the present report, were
carried out in compliance with the appropriate eth-
cal principles, with approval by the institutional
review board of the Pars Hospital, Tehran, Iran,
and after obtaining informed consent from the
patient.

**Case Presentation**

A 30-year-old woman, gravida 2, para 1, 17 week
pregnant, was hospitalized in gynecology and ob-
stetrics department of Pars Hospital on May 2014
with chief complain of bilateral breast enlarge-
ment, as well as breast skin edema and inflamma-
tory changes. Patient had a series of previous
breast ultrasounds, indicating fibrocystic changes
with a progressive enlargement from the 9th week
of pregnancy. The chest circumference was 109 cm
and sonography examination was normal except
for fibrocystic findings. Two days after admis-
sion, the patient developed breast skin ulceration
which was bandaged and the sample was sent for
culture examination. The results of wound culture
were negative for microorganism growth. The
patient was released 10 days after admission with
stable condition.

She was again admitted to emergency depart-
ment at 25th week of pregnancy with coughs and
dyspnea increasing gradually since 15 days before
admission. Ultrasonography exam again revealed
fibrocystic changes and the breast size had in-
creased to 122 cm in circumference. Heart exami-
nation showed tachycardia and ejection fraction of
60% in echocardiography. Patient also had anemia
and her prolactin level was 49.7 ng/ml at first ad-
mission and rose to 67 ng/ml before delivery
which were both in normal range for pregnant
women. It was reduced to 27 ng/ml one week after
breast surgery. With thorough evaluation of the
patient, the diagnosis of asthma was established
and she received Clexane, Betamethasone, Veno-
fer, and Azithromycin and was discharged after 7
days with stable condition.

She was admitted again to the hospital in 31th
week of pregnancy with dry coughs, dyspnea, and
tachycardia. No peripheral edema was
observed. The breast circumference had increased
to 134 cm and patient was treated by Clexane,
methylprednisolone, and Azithromycin. She was
released after 3 days with stable condition.

She was admitted at week 32 for the fourth time
with dry coughs, dyspnea, vomiting, reduced urine
volume and depression. Her breast circumference
at this time had reached 155 cm (Figure 1). She
also showed hypercalcemia (Ca=13.5) in two oc-
tasions. Considering the patient’s condition, she
was a candidate for cesarean section after normal-
izing the Ca level and gave birth to a male new-
born with Apgar score of 7 and 8 in the first and
fifth minute after delivery, 1.38 kg of weight, and
good respiratory conditions. Although it was sup-
posed that the breast enlargement might be due to
pregnancy hormonal changes, the breast size did
not change after delivery and considering the size
of breasts as well as respiratory and cardiac com-
lications, patient underwent total mastectomy in
two surgeries with nipple and a flap of areola transplanted back 10 days after delivery. Each
breast weighted 16.5 kg before the start of the
surgery. Pathology report indicated fibrocystic
changes (Figure 2), increased stroma and CD34
positivity. The calcium levels returned to normal
levels one week after the total mastectomy and
patient left the hospital 12 days after the surgery
with good general condition.

**Figure 1.** Patients breasts before the surgery

**Figure 2.** The small slit-like spaces in the stroma (pseudo-
angiomatous spaces) are lined with cells that are stained with
smooth muscle actin (SMA) marker in immunohistochemis-
try (IHC) study. Note that the basal cells around the mamma-
ry ducts are also stained with this marker, which is presumed
as a normal histologic finding. Also, note that the benign
proliferation and branching of breast ducts is closely similar
to the benign fibroadenoma of the breast (X100; IHC staining
for SMA marker)
**Discussion**

Gestational gigantomastia is a complication whose etiology and pathogenesis have yet to be fully clarified; however, various theories including endocrine imbalance, hyperprolactinemia and end organ hypersensitivity have been suggested by different researchers (3, 4). Gestational macromastia can manifest either in first pregnancy or after previous normal pregnancies (6). Manifestations usually begin in the first trimester (7) but in the current presentation, the patient was admitted to hospital at week 17 of pregnancy, although her breasts initially had begun to grow in size from the first trimester. Similar cases of admission at second trimester of pregnancy have previously been reported. In a case reported by Eler Dos Reis et al. (8), their 24 year old pregnant patient was referred to hospital at 22nd week of pregnancy. Agarwal et al. reported that their 22 year old pregnant patient came to their hospital at week 24th of her pregnancy. Following the increase in the size of the breasts, ulcerations, infections, and areas of local necrosis, causing severe maternal morbidity and even mortality might be seen (1). In our patient, the ulcerations appeared at her first visit. Similar ulceration in cases of gestational gigantomastia has been reported by Ezem et al. (1), Eler dos Reis et al. (8) and Agarwal et al. (9).

In the case reported by Eler dos Reis et al. (8), because of the progressive increase in the size of their patient’s breasts as well as necrosis and recurrent infections, her pregnancy had to be interrupted at 28 weeks of gestational age but the delivery was postponed by cesarean section in our patient until week 32 of pregnancy.

Depression due to combination of pregnancy and unbearable size of the breasts limiting the patients mobility and interaction with others has been previously reported among patients with gigantomastia (1, 8). Our patient developed depression at the 32nd gestational week.

The exact role of hyperprolactinemia is not clear. Most patients with hyperprolactinemia do not present with breast hypertrophy, yet those with pregnancy induced gigantomastia usually respond well to high doses of anti-prolactin agents such as the dopamine agonist bromocriptine (10). The normal range of prolactin for non pregnant women is 4.8-23.3 ng/ml, which might increase to 34-386 ng/ml in pregnant women. In our patient, the prolactin reading during pregnancy was in normal range for pregnant women. Bloom et al. (11) have reported increased levels of prolactin in their 32 year old patient with gestational gigantomastia.

Our patient also developed hypercalcemia in her 32nd week of pregnancy. Hypercalcemia in patients with pregnancy-induced gigantomastia has been attributed to the excessive production of parathyroid hormone-related protein (PTHrP) by the (hypertrophied) breasts (12). The exact reason for the abnormally high level of PTHrP production in pregnancy-induced gigantomastia is unknown (10). It seems that in patients with the combination of gigantomastia and excessive PTHrP of mammary origin causing hypercalcemia, radical breast reduction and in some cases bilateral mastectomy may be the only option available (10). The majority of pregnant patients with hypercalcemia remain asymptomatic during gestation so the careful examination of calcium levels in patients with gigantomastia has been suggested (13).

The main objective of the palliative treatment among patients with gestational gigantomastia is improving the adverse conditions affecting pregnancy, minimizing the pain and providing psychological support for patients suffering from depression (8). The effect of drug therapy on gigantomastia is limited, so the surgical treatment remains the mainstay treatment method (14-16). Options for surgical management have included reduction mammoplasty and mastectomy (15). Although spontaneous involution might happen in rare cases it is sufficient to return the breast to its premorbid condition (11, 14). Considering the high chance of recurrence during the next pregnancies, it has been recommended that patients undergo mastectomy rather than reduction mammoplasty when future pregnancy is desired (11, 14).

The decision was made to perform a total mastectomy due to unbearable size of breasts not being reduced after delivery, as well as respiratory and cardiac complications and also hypercalcemia. In a similar case, Eler dos Reis et al. (8) decided to perform simple bilateral mastectomy 52 days after their patient gave birth due to continuation of complications and the size of their patient’s breasts not reducing after delivery. In another case reported by Karagulle Et al. (17), patient underwent surgery about two months post delivery due to initial refusal of patient for resection of breast mass. Alternatively, successful management of gestational gigantomastia using reduction mammoplasty has been previously reported (11).
Usually, the pathology of the surgical specimens shows glandular hyperplasia with an increase in the connective tissue and no involvement of the adipose tissue (8). In the present case, pathology revealed breast tissue with fibrocystic changes without atypia.

**Conclusion**

Although gestational gigantomastia is a rare condition, its timely prognosis and careful examination of some conditions like hyperprolactinemia and hypercalcemia is essential in successful management of this condition.

**Conflict of Interest**

None of the authors has a conflict of interest with the subject matter of the present study.

**References**


