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Gestational gigantomastia on a Saudi woman: A case report on surgical removal and reconstruction and management of complications, KFSH&RC

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ABSTRACT

INTRODUCTION: Gestational gigantomastia (GG) is a rare condition manifesting as a fast and excessive growth of the breasts in pregnant women. Its etiology is still unclear, with theories ranging from hormonal imbalances, unregulated immune response, to hypersensitivity. Medical interventions are mainly surgical in nature, though some pharmacological medications are of debatable efficacy.

CASE PRESENTATION: A 33-year old Saudi gravida 3 para 2 presents continuous breast enlargement since the start of her pregnancy. She complains of skin ulcerations and discharge which was initially treated conservatively with topical antibiotics. Days after she came back with worsening GG symptoms, and was admitted for bilateral skin sparing mastectomy and reconstruction, and successfully recovered. The patient came back with problems concerning the surgical implant and wound infection. Emergency operation was performed for implant removal and wound treatment. Labor induction was performed by the OB-GYN on her 39 week. The patient opted for autogenous reconstruction by bilateral latissimus dorsi flap months after delivery. After treatment of minor surgical complications, the patient successfully recovered.

DISCUSSION: Surgery is one of the most effective interventions for GG. Total mastectomy is preferred due to lesser risk of recurrence in subsequent pregnancies. Reduction mammoplasty offers the breastfeeding option if conducted before the delivery, but poses higher risk of recurrence in future pregnancies.

CONCLUSION: The patient's gestational gigantomastia condition was complicated by several ulcerations and infections. Surgery was conducted alongside antibiotic treatment. This report also highlights the importance of follow ups in managing complications.

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1. Introduction

Gestational gigantomastia (GG) is a rare condition occurring in women during pregnancy. In around 1 in 100 000 pregnancies, GG is manifested through exceptionally fast and uncontrolled growth

of breasts in terms of size [1]. The etiology for this condition has not yet been established, although there are speculations about changes in hormone levels being one of the important factors. The criteria for diagnosis include excessive breast size and weight, possibly in one (unilateral) or both (bilateral) breasts [2]. It is important to note that aside from the physical burden of this condition, pregnant women also experience the concomitant psychological strain and reduced quality of life. This report was written in accordance to the SCARE guidelines [3].

2. Case presentation

A 33-year old pregnant woman from Saudi Arabia was admitted at her 23rd week of gestation, presenting symptoms of gestational

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Fig. 1. Patient manifestation of GG before bilateral simple mastectomy.



Fig. 2. Post-surgery image of patient with GG.



Fig. 3. Patient after recovery from surgery.



Fig. 4. First visit after bilateral nipple reconstruction.

gigantomastia (GG). She reported breast enlargement since the beginning of her pregnancy, which was complicated with skin ulcerations and discharge. According to her medical records, there were no other medical conditions aside from the GG symptoms upon admission. Obstetric data also indicated that she is gravida 3 para 2, and previous children were delivered via spontaneous vaginal delivery (SVD). Previous pregnancies went without uncommon complications, and noted no family history with breast cancer or similar conditions. Moreover, she was on contraceptive patches for about 3 years. Conservative management of GG symptoms was attempted through topical and intravenous antibiotics, as well as bromocriptine. Patient was subsequently discharged.

The patient returned the following week, exhibiting worse GG symptoms of skin ulcerations involving the nipples. Due to the increasing pain and discomfort, she opted for a bilateral skin sparing mastectomy procedure, despite knowing that she will not be able to breastfeed afterwards (Figs. 1–4).

During the surgery, it is worth noting that the skin was stiff and the veins were very prominent. A significant amount of bleeding was also encountered despite careful hemostasis. The extracted part of the right breast weighed 5.08 kg while the left weighed 4.490 kg.

Immediate reconstruction by pre-pectoral implant and Acellular dermal matrix (ADM) was done as a first time for gigantomastia cases. Histopathology of breast tissues revealed marked stromal fibrosis associated with pseudo angiomatous stromal hyperplasia

(PASH). Additionally, immunostaining showed a positive result for CD34 and negative results for CD31, ER and PR.

Three days post-operation, the patient developed a small seroma at the upper right region of the breast which was confirmed via ultrasound and the observed epidermolysis. She was administered amoxicillin/clavulanic acid intravenously for cellulitis and infection, awaiting the wound swab result. After 4 days, she was operated on for right breast wound debridement. She was then discharged after recovery.

Twelve days later, the patient came back again to the ER showing an exposed left implant and pus draining from the right breast wound. Emergency operation was performed; bilateral implants were removed and pus was drained from her wound. Results of her wound cultures revealed that her right breast wound is infected by *Serratia marcescens* while her left breast wound is mainly of *S. marcescens* and *Candida albicans* populations. She was discharged after 3 days and remained under meropenem for 2 weeks. Throughout the process since the first admission, her OB-GYN was notified of all the procedures she had undergone. Given the circumstance, she was admitted for labor induction on her 39th week and successfully delivered through SVD a baby boy weighing 2.58 kg.

After labor, the patient was insisting to go for reconstruction by implant again but her breast skin flap was so thin and cannot accommodate an implant, so fat grafting was offered to address this issue and get a thicker breast flap before proceeding with implant to avoid any further complications. The amount of fat aspirated from her abdomen and flanks was 300 cc, 140 cc of which allocated to the left breast and 160 cc for the right breast.

Months later, the patient came back asking for autogenous reconstruction to get more natural consistency than implant. The options were discussed with the patient and after proper examination; she decided to go for LD flap. She underwent reconstruction by bilateral latissimus dorsi flap with implants sized 225. She was initially doing well aside from blisters with erythema in the back donor site, which were treated conservatively. Twenty days post-op, a small amount of discharge from the back donor site, and a

small area of wound dehiscence were presented. Culture results revealed mainly Klebsiella populations. The appropriate antibiotics and wound dressing were administered and she is currently doing well without issues. The patient then underwent bilateral nipple reconstruction and upsizing of the implants to 375 cc.

3. Discussion

The information about gestational gigantomastia in literature is limited, which could be the consequence of its rarity [4]. Rezai et al. [5] in 2015 listed some relevant observations about the condition. GG occurs at slightly higher rates in multiparous women and those of Caucasian origin. It can manifest in any pregnancy, and there is a higher risk of recurrence in subsequent pregnancies. Having a history of GG is also a risk factor. Additionally, it has been observed that patients undergoing breast reduction procedures are exposed to an increased risk of recurrence than those undergoing total mastectomy, which could be related to the remaining hypertrophic tissues after the procedure.

It is important to point out that the condition, although the symptoms may be that of a malignancy, is benign. Rutherford et al. [6] in 2019 noted in his case report about a unilateral GG patient with benign breast tissues, and exhibiting a higher rate of collagen formation and stromal fibrosis. No malignancy or any cell abnormality was observed.

There were also similar cases of breast enlargement attributed to *D*-penicillamine, a drug used for treating autoimmune rheumatic diseases such as scleroderma and rheumatoid arthritis [7,8]. Other rare complications include galactorrhea and hyperprolactinaemia [9]. Most interventions for this type of gigantomastia rely on surgical removal/reduction of breast tissues.

As of the moment, there are also limited options in the medical management of gestational gigantomastia. Bromocriptine is used as the main pharmacological intervention for GG [10]. It is known to halt the excessive and fast growth rate of the breasts, but further studies need to be conducted to determine if it can actually cause regression. Some drugs that have been employed to no avail include hormonal treatments of androgen, progesterone, estrogen; 2 Br-alpha ergocryptine, tamoxifen, norethindrone and stilbesterol [11].

The most promising interventions are mainly surgical procedures: reduction mammoplasty and total mastectomy [12]. Though, in reduction mammoplasty, there is an increased risk of recurrence due to the remaining tissues that may exhibit hyperplasia in subsequent pregnancies, it still preserves the breastfeeding capability of the mother post-operation [11]. On the other hand, total mastectomy minimizes the risk of recurrence. This is the reason why total mastectomy is preferred several months after delivery (when the baby can wean off breastfeeding) and reduction mammoplasty is performed weeks or months before delivery. In very rare cases, GG underwent complete resolution after pregnancy even without medical interventions [13].

In this case of GG, several complications were encountered, and were treated as emergency due to the increased risk associated with pregnancy. Any delay or failure to treat these complications may lead to septicemia, which is dangerous for the patient and her child.

4. Conclusion

Gestational gigantomastia is a rare medical condition occurring in pregnant women. The etiology of the disease is not yet established, though several medical observations point towards surgery as the definitive solution. Total mastectomy will be of greater benefit for women who are desirous of pregnancies in the future due to the less risk of recurrence. On the other hand, reduction mammoplasty may be performed to retain the breastfeeding func-

tion of the mother. Complications associated with GG and surgery should be managed appropriately through follow-up consultations post-operation. This is the first published case report of gestational gigantomastia in Saudi Arabia that consider the pre-pectoral implant with ADM as an option for reconstruction.

Conflicts of interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

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Ethical approval

Ethical consideration was approved by the ethical committee of the surgery department of King Faisal Specialist Hospital and Research Centre.

Consent

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Author contribution

1. Dr. Foad hashem: Surgeon performing the operation, final approval of the manuscript and follow up.
2. Dr. Moraya Alqahtani: Surgeon performing the operation, final approval of the manuscript and follow up.
3. Nehal mahabbat: Writing the manuscript, final approval of the manuscript and follow up.
4. Asma Abdulla: Writing the manuscript, final approval of the manuscript and follow up.
5. Fares Alsufayan: Writing the manuscript, final approval of the manuscript and follow up.
6. Ahmed Alarbi: Writing the manuscript, final approval of the manuscript and follow up.
7. Atif Rafique: literature review, final approval of the manuscript.

Registration of research studies

1. Name of the registry: Gestational gigantomastia on a Saudi woman: A case report on surgical removal and reconstruction by pre-pectoral implant with ADM
2. Unique identifying number or registration ID: researchregistry6149
3. <https://www.researchregistry.com/browse-the-registry#home/registrationdetails/5f91a9bbde5c3a00167b6529/>

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