



Contents lists available at [ScienceDirect](https://www.sciencedirect.com)

JPRAS Open

journal homepage: www.elsevier.com/locate/jpra



Severe Gestational Gigantomastia: from Mastectomy to Staged Autologous Breast Reconstruction. A Case Report

Noureddine Hassayoune, MD^a, Mina Mhallem Gziri, MD, PhD^b,
Audrey Lentini, MD^a, Theodoros Chrelias, MD^a,
Jennifer Hammer, MD^a, Martine Berlière, MD, PhD^c,
Benoît Lengelé, MD, PhD, FRCS^a, Maude Coyette, MD^{a,*}

^a Department of Plastic and Reconstructive Surgery, Cliniques Universitaires Saint-Luc, UCLouvain, Avenue Hippocrate 10, 1200, Brussels, Belgium

^b Department of Obstetrics, Cliniques Universitaires Saint-Luc, UCLouvain, Avenue Hippocrate 10, 1200, Brussels, Belgium

^c Department of Gynecology, Cliniques Universitaires Saint-Luc, UCLouvain, Avenue Hippocrate 10, 1200, Brussels, Belgium

ARTICLE INFO

Article history:

Received 16 March 2021

Accepted 29 April 2021

Available online 18 May 2021

Keywords:

Gestational Gigantomastia

Mastectomy

Delayed Breast Reconstruction

Tissue expander

DIEP flap

nipple-areola complex

ABSTRACT

Gestational gigantomastia (GGM) is a rare condition characterized by a massive overgrowth of breast tissue during pregnancy. Surgical sanction may be required when conservative measures fail. In this study, we report the case of a 29-year-old woman who presented with an evolutive GGM responsible for physical and emotional distress, despite medical treatment. A multidisciplinary decision was made to induce delivery at 32 weeks. In the post-delivery period, the patient developed breast wounds, complicated with septic cardiomyopathy. An emergency bilateral mastectomy was then carried out, together with banking of both nipple-areola complexes. Thereafter, delayed bilateral 2-stage breast reconstruction was started at 12 months with subcutaneous tissue expanders, later on followed by implants removal and autologous reconstruc-

* Corresponding author: Maude Coyette, MD, Department of Plastic and Reconstructive Surgery, Cliniques Universitaires Saint-Luc, UCLouvain, Avenue Hippocrate 10, 1200 Brussels, Belgium, Phone: (+32) 02/7641403 Fax: (+32) 02/7626284.

E-mail address: maude.coyette@uclouvain.be (M. Coyette).

tion with bilateral deep inferior epigastric artery perforator flaps and bilateral nipple replantation.

© 2021 The Author(s). Published by Elsevier Ltd on behalf of British Association of Plastic, Reconstructive and Aesthetic Surgeons.

This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>)

Case report

A 29-year-old patient, 15-weeks pregnant, presented to the obstetrics department with major breast enlargement, associated with erythema and pain. Blood tests showed a light inflammatory syndrome and normal prolactin levels. Ultrasonography, along with full thickness skin biopsies, confirmed the diagnosis of mastitis and an oral antibiotic treatment was initiated. Further investigation with magnetic resonance imaging showed edema and skin thickening. Breast biopsies emphasized pseudo-angiomatous stromal hyperplasia, without malignancy evidence. The patient was diagnosed with gestational gigantomastia. Conservative medical treatment was initiated with bromocriptine 2.5 mg twice daily.

At 23 weeks of gestation, clinical examination showed the worsening of breast enlargement, several painful skin ulcerations, and development of an important superficial venous network (Figure 1). The patient experienced increased discomfort with dyspnea and thoracic oppression. Nevertheless, her vital parameters remained stable. The multidisciplinary team decided to delay surgery until after delivery. The situation continued to deteriorate, with bleeding episodes and mental exhaustion. Fetal lung maturation by oral steroid was started at 29 weeks, and the elective delivery of a healthy baby took place by cesarean section at 32 weeks.



Figure 1. Massive breast overgrowth with subsequent skin ulcerations hidden under the dressings and major enlargement of the subcutaneous venous network.

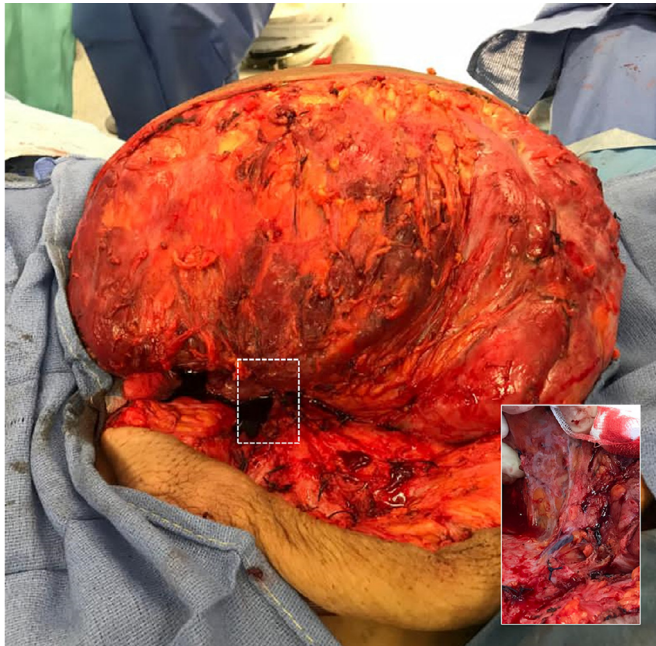


Figure 2. Peroperative view during the mastectomy showing the enlarged perforating vascular pedicles supplying the deep aspect of the mammary gland.

In the early postpartum period, the patient developed a severe *Pseudomonas Aeruginosa* sepsis consecutive to secondary infection of breast ulcerations. We then decided to perform an emergency bilateral skin-sparing mastectomy. The surgery was conducted on both sides at the same time, which aimed to reduce the duration of the procedure. First, the nipple-areola complexes (NACs) were harvested as full thickness skin grafts and transferred as composite grafts to the inner side of the arms. We then proceeded to both mastectomies. To reduce blood loss, we used the LigaSure™ Exact Dissector (Covidien, Boulder, CO, USA) coupled with multiple 2.0 absorbable sutures for larger perforators arising from the chest muscles (Figure 2). The mastectomy weight was 9,315 g on the right side and 6,430 g on the left, with an extra 140 g left accessory axillary gland. The residual skin was redraped and allowed tension-free suture. The postoperative course was uneventful.

Reconstruction began 12 months later with prepectoral placement of 450 cc temporary tissue expanders (Mentor CPX™ 4 Tissue Expander with Suture Tabs, Medium Height, Style 9200, Johnson & Johnson Medical BV). Once the expansion was completed and stable (Figure 3), we performed a bilateral autologous breast reconstruction using deep inferior epigastric artery perforator (DIEP) flaps, after implant removal and complete capsulectomy. Six months later, the patient underwent revision surgery with lipofilling and free nipple graft. Finally, the reconstruction was completed with areola tattooing and offered a good esthetic result (Figure 4).

DISCUSSION

Gigantomastia is a rare condition characterized by breast overgrowth, which leads to physical and emotional distress. H.Dafydd *et al.*¹ proposed to define it as a breast tissue excess that contributes 3% or more to the patient's total body weight. However, this definition is based on the weight of mastectomy or breast reduction specimens and therefore, can only be applied retrospectively. Other authors² proposed a classification system based on the etiology (idiopathic, hormonal imbalance, or drug induced) of hyperplasia.

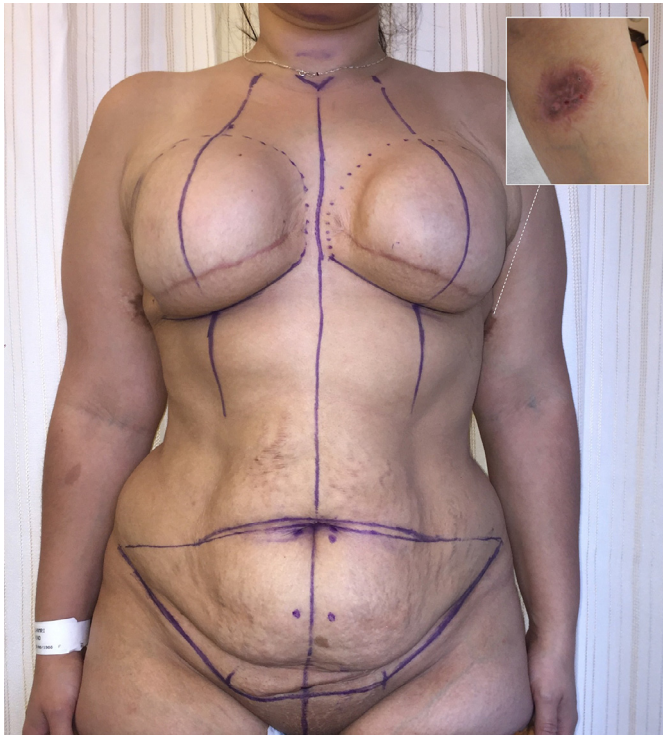


Figure 3. Preoperative markings for bilateral DIEP flap reconstruction. Abdominal stretch marks illustrated in this picture motivated the use of temporary expanders shown here in full expansion. The nipple areola complexes are visible on the inner side of the arms.

Gestational gigantomastia (GGM) incidence is reported to be between 1 in 28 000-100 000 pregnancies. It seems to occur more commonly in multiparous women, with the onset of unusual breast enlargement in the first trimester.³ The etiology and pathogenesis of GGM are not straightforward. Hormone abnormalities are believed to play a major role. However, breast tissue receptor sensitivity, malignancy, and/or autoimmune mechanisms may also be involved.⁴ This might explain why the course and the severity of the disease, as well as the response to medical treatment, are patient dependent. Conservative approaches with diverse drugs that include testosterone derivatives, estrogens, progestins, tamoxifen, hydrocortisone, diuretics, and prolactin suppressors have been attempted with mixed results.² Bromocriptine, a dopamine agonist that might stop the progression or slightly reverse breast overgrowth, is commonly advocated as the first line of treatment.^{2,4} However, results are inconsistent and regression to initial breast size has not been shown.³

Surgical approach seems to offer a more effective solution when medical management fails. Reduction mammoplasty offers a good esthetic result while breastfeeding ability is maintained. However, this option exposes the patient to a more hemorrhagic procedure and a 100% recurrence rate for subsequent pregnancies.² Bilateral mastectomy, on the other hand, ensures the absence of GGM recurrence.³ Nevertheless, it is a mutilating procedure that requires several reconstructive steps. The timing of the surgery should always be weighed against the risk of maternal and fetal complications. When the life of the mother and/or the baby is at stake, a mastectomy should be performed urgently.⁵ It is a fast procedure that limits blood loss and reduces the fetus exposure to anesthetic drugs. Given the high risk of inducing premature delivery, preliminary oral steroids administration to promote fetal lung maturation is mandatory.⁶ Nonetheless, it is always better to postpone surgery until the un-



Figure 4. Final good esthetic results after lipofilling, banked nipples grafting, and areola tattooing.

born child is viable, and whenever possible, after delivery itself. This safest strategy was successfully demonstrated, despite early dramatic clinical presentation.

Immediate breast reconstruction has been described in GGM surgical management.^{4,7-9} Nevertheless, skin retraction and wound healing issues are difficult to predict and could lead to poorer aesthetic results, or even to reconstruction failure. Therefore, a delayed approach is often offered to minimize these potential problems.^{2,6} Our patient was eligible for a secondary autologous reconstruction but did not want her major abdominal stretch marks transferred on her reconstructed breasts. We then decided on a two-stage procedure. First, prepectoral expanders placement allowed a progressive expansion of the remaining thoracic skin. During the second stage, a complete capsulectomy was performed to position the DIEP flaps properly and promote their adhesion to surrounding tissues. The prior expansion allowed us to obtain sufficiently large pockets to entirely bury both flaps, leaving no visible skin paddles. Moreover, the prepectoral location of the expanders avoided the need for secondary repositioning and anchoring of the pectoralis major.

Two major options are possible for NAC management at the time of the mastectomy. They can be harvested as full thickness skin grafts and either be transferred directly on the mastectomy flaps^{5-7,9} or banked for further use.¹⁰ However, in the case of immediate replantation, the risk of partial or full necrosis and malposition must be taken into account. Therefore, and given a mycotic infection of the groin regions, we decided to temporarily bank both NACs on the inner face of the arms. Unfortunately, because of scar contraction and subsequent deformity of both NACs, we decided to replant only the nipples from the banking sites onto the reconstructed breasts. The final result, however, after tattooing was favorable.

The diagnosis of GGM should be confirmed after a full work-up is completed to rule out any malignancy or underlying disease. Bromocriptine is the first line therapy, but surgery is often required to

solve the problem. There is an increased recurrence risk following reduction mammoplasty in case of subsequent pregnancy. Consequently, bilateral mastectomy, with secondary autologous breast reconstruction offers ideal and durable esthetic results. The multidisciplinary management of these young patients should be made on a case-by-case individual approach, particularly regarding the optimal timing of surgery versus delivery and considering the safest balanced vital and esthetic outcomes for the mother and her child.

Conflict of Interest

None.

Funding

None.

Ethical Approval

N/A.

Patient consent

Confirmed that written consent was obtained for publication.

Supplementary materials

Supplementary material associated with this article can be found, in the online version, at doi:[10.1016/j.jpra.2021.04.011](https://doi.org/10.1016/j.jpra.2021.04.011).

References

1. Dafydd H, Roehl KR, Phillips LG, et al. Redefining gigantomastia. *J Plast Reconstr Aesthet Surg*. 2011;64:160–163.
2. Dancey A, Khan M, Dawson J, et al. Gigantomastia – a classification and review of the literature. *J Plast Reconstr Aesthet Surg*. 2008;61:493–502.
3. Swelstad MR, Swelstad BB, Rao VK, et al. Management of gestational gigantomastia. *Breast*. 2006;118:840–848.
4. Shoma A, Elbassiony L, Amin M, et al. Gestational gigantomastia": A review article and case presentation of a new surgical management option. *Surg Innov*. 2011;18:94–100.
5. El Boghdadly S, Pitkanen J, Hassonah M, Al Saghier M. Emergency mastectomy in gigantomastia of pregnancy: A case report and literature review. *Ann Saudi Med*. 1997 Mar;17(2):220–222.
6. Lapid O. Breast Reconstruction After Mastectomy for Gestational Gigantomastia. *Aesth Plast Surg*. 2013;37:388–391.
7. Nail-Barthelemy R, Burin des Roziers B, Daoud G, et al. Breast reconstruction for gigantomastia complicating pregnancy. A case report. *Ann Chir Plast Esthet*. 2015 Aug;60(4):330–335.
8. Mahabbat N, Abdulla A, Alsafayan F, Alharbi A, Rafique A, Alqahtani M, Hashem F. Gestational gigantomastia on a Saudi woman: A case report on surgical removal and reconstruction and management of complications, KFSH&RC. *Int J Surg Case Rep*. 2020 Oct 28;77:157–160.
9. Qin F, Si L, Zhang H, Zhang M, Zeng A, Long F, Yu N, Liu Z, Wang X. Management of gestational gigantomastia with breast reconstruction after mastectomy: case report and literature review. *J Int Med Res*. 2020 Jun;48(6).
10. Ahmed AK, Hahn DE, Hage JJ, Bleiker EM, Woerdeman LA. Temporary banking of the nipple-areola complex in 97 skin-sparing mastectomies. *Plast Reconstr Surg*. 2011 Feb;127(2):531–539.