Received: 3 December 2023 | Revised: 12 January 2024 | Accepted: 31 January 2024

DOI: 10.1002/ccr3.8607

CASE REPORT

Clinical Case Reports Open Access WILEY

Gestational macromastia complicating pregnancy: A case report of unusual bilateral giant breasts in a patient who had no such problem in her previous pregnancies

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Funding information
International Cancer Institute (Kenya)

Key Clinical Message

Gestational macromastia is a rare disorder involving excessive breast tissue enlargement during pregnancy, potentially threatening the fetus and the mother. Our patient's previous pregnancies were more physiological, without any associated symptoms.

Abstract

Moderate bilateral enlargement of the breasts is a normal finding during pregnancy and lactation. Occasionally, there is a continuity from this physiological hypertrophy to massive breast hyperplasia or gigantomastia, causing complications that threaten the wellbeing of the fetus and the mother or raising alarm for a malignant disease. We present a case of a 28-year-old G3P2L2 with a gestational age of 29 weeks complaining of massive bilateral breast enlargement for 6 months, accompanied by a threatened abortion. The masses began gradually after conception and increased gradually over time. Notably, the breasts had been unremarkable in her two previous pregnancies. The breast ultrasonography and mammography findings pointed to a benign neoplasm. Histopathology of the lesions reported bilateral ductal hyperplasia. Conservative management and close follow-up were initiated. A cesarean section was performed due to cord prolapse, and a 1.3 kg male baby was extracted. Unfortunately, the neonate succumbed after 3 days due to apnea of prematurity. The patients' breast size subsided considerably with time. Unusually large tumors can cause alarm for other pathologies, such as breast cancer. The radiological tests should reassure the attending practitioner, and the histological examination should confirm the diagnosis. An understanding of the typical and atypical clinico-pathologic characteristics of breast lesions occurring in pregnancy and lactation is essential for appropriate patient care.

KEYWORDS

bilateral, breast, breast hypertrophy, ductal hyperplasia, gestational macromastia, pregnancy

1 | INTRODUCTION

Gestational macromastia, also known as gigantomastia in pregnancy, is a massive breast enlargement during pregnancy.[1–3] This extremely rare condition presents as giant ductal hyperplasia and may have lactational histologic alterations during pregnancy, while prolactin, progesterone, and estrogen levels might be high, encouraging the development of tubuloalveolar structures and ductal expansion as the cause of the noticeable expansion during this time. The benign breast lesions typically affect women in their second and third decades of life, and the clinical implications of the diagnosis of atypical ductal hyperplasia (ADH) and ductal carcinoma in situ (DCIS) are very different. Yet there are "borderline" breast lesions that have characteristics of both ADH and DCIS, and nevertheless, 0.5%–2% of these lesions will advance quickly.[4] Greater than 5 cm or 500 g of giant ductal hyperplasia can cause considerable deformity, raising the possibility of cancer and necessitating surgical excision. About 0.1% of women under the age of 20 will develop primary breast cancer, and there have been cases of in situ ductal carcinoma and neoplasms in fibroadenomas. Women of African or Asian descent are the most frequently affected by these benign tumors. Although the specific cause of bilateral giant ductal hyperplasia is unknown, its increased incidence throughout puberty and susceptibility to pregnancy, oral contraceptives, and cyclic hormones suggest that it is most likely an aberrant reaction to estrogen and may complicate pregnancy.[4] We describe an unusual case of pregnancy-related bilateral giant ductal hyperplasia in a patient who had two previous term pregnancies without this condition, and we provide a brief review of the literature.

2 | CASE HISTORY

A 28-year-old female G3P2L2 presented with chief complaints of bilateral massive breast masses for 6 months. She was pregnant at a gestational age of 29 weeks + 6 days. The breast masses started gradually since conception and were equally progressively increasing with time. They were associated with itching, back pain, skin changes, on/off difficulty breathing while lying flat, as well as difficulty walking. There was no history of trauma, nipple discharge, bleeding, or retraction. There was no history of using local herbal medication or similar illnesses in the family. Additionally, she presented with a gradual onset of lower abdominal pain for 1 day, which worsened progressively with radiation to the lower back. She attended an antenatal clinic at 12 weeks, where she received all her supplements. The malaria rapid test was negative, and the venereal disease research laboratory was non-reactive. Her past medical and socio-family histories were essentially normal. Her breast size increases during previous pregnancies were not as dramatic and were more physiological without other associated symptoms. She exclusively breastfed her last infant for the first 6 months, supplemented with other foods, and kept breastfeeding until the child was 2 years old. At least 3 years have passed since she gave birth to her last infant, and she is no longer nursing her last infant.

3 | METHODS

On examination, she was alert in pain, mildly pale, dyspneic, not jaundiced, and not cyanosed, with no lower limb edema. She had palpable bilateral axillary lymph nodes. Her vital signs were all stable. There were bilaterally enlarged breasts measuring about 40 × 32 cm each, with hyper-pigmented lesions in some parts and a hard and irregular surface upon palpation. There were no color changes observed; she had a normal body temperature. The breasts were severely tender upon palpation and not associated with nipple discharge, bleeding, or nipple retraction (Figure 1A). Per abdomen, a gravid abdomen was moving with respiration and was noted with a fundal height of 28/40 with contractions. Obstetric ultrasound showed a single live intrauterine pregnancy, longitudinal lying, cephalic presentation, FHR 143 b/min, EGA 31 weeks, and a weight of about 1.2 kg.

Per vaginal examination, the cervix was thin, anterior, and 8 cm dilated, with a spontaneous rupture of membranes (ROM) and a visible cord through the cervix to the vagina, hence cord prolapse. Except for the massive bilateral enlargement of the breast, all other systems were essentially normal. Laboratory FBC results showed HB of 10.5 g/dL, while ALAT, AST, WBC, creatinine, BUN, and serum electrolytes were within the normal ranges. Estrogen was 44.1 pg/mL, and prolactin was 853.3 uIU/mL (extremely high). The chest X-ray and abdominal ultrasound had normal findings. The bilateral breast ultrasound and mammogram results were suggestive of mastitis. Trucut breast

FIGURE 1 Photographs of the patient demonstrating bilaterally massive breast enlargement in pregnancy (A); considerably subsided breasts 1 year after delivery (B).

biopsies were collected for histopathology. A cesarean section was undertaken due to cord prolapse, and a 1.3 kg male baby was extracted with an apgar score of 6 and 8 in the 1st and 5th minutes, respectively.

4 | CONCLUSION AND RESULTS

Thereafter, the neonate was admitted to the pediatric ward and died after 3 days due to apnea of prematurity. In the ward, the patient received bromocriptine (2.5 mg twice daily), post-operative management including antipain drugs such as pethidine and diclofenac, and antibiotics such as metronidazole and ceftriaxone. The histopathology results from the bilateral breast biopsies demonstrated features consistent with ductal hyperplasia (Figures 2A,B). She was then discharged and counseled that the condition may improve with time. To date, 1 year has passed, she is doing well, and her breast size has subsided significantly (Figure 1B).

5 | DISCUSSION

Our report describes a case of unusually massive bilateral breast hyperplasia during pregnancy in a 28-year-old African female who had no such problem in her previous pregnancies. Risk factors for gestational macromastia are not well understood, but their occurrence is more common in Caucasian and multiparous women. Gestational macromastia may happen during any pregnancy. A prior history of macromastia increases the risk of its occurrence in subsequent pregnancies.[1–3] Furthermore, rates of recurrence have historically increased in patients who underwent reduction mammoplasty instead of bilateral total mastectomy, with the recurrence being attributed to retained hypertrophic tissue after mammoplasty. As it was seen in the index case, it is worth noting that the disease can emerge in multiparous women who have never had a problem with previous pregnancies.[5] Young women are more likely to develop ductal hyperplasia, which is a benign neoplasm with epithelial (glandular) and stromal (fibrous) components.[4,6] A large glandular epithelium and enhanced stromal cellularity are described in gigantic fibroadenomas.[7] As in the index case, these lesions frequently develop in the upper outer quadrant, can enlarge to massive size, and may have lactational histologic alterations during pregnancy. High levels of estrogen, progesterone, and prolactin encourage the development of tubuloalveolar structures and ductal expansion. This could be the cause of the noticeable expansion during this time. Breast enlargement can happen in as little as a few weeks, and within 3–6 months, the mass can double in size, growing bigger than the preexisting normal breast tissue. Surgical excision is the normal course of treatment for all gigantic lesions. Although it is uncommon, local recurrences can happen. On imaging, gigantic macromastia show up as solid masses on ultrasound and well-circumscribed masses on mammography.[6,7]

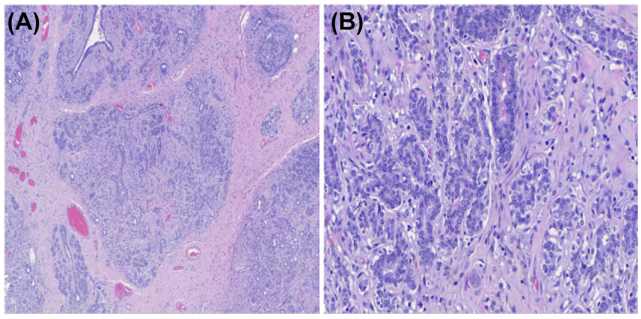

FIGURE 2 Histopathology demonstrating a benign breast ductal proliferative lesion typically characterized by secondary lumens and streaming of the central proliferating cells (A); proliferation of cells of luminal and myoepithelial lineages (B).

Given their benign nature, some obstetricians recommend surgical removal of these tumors prior to conception, while others tend to favor conservative treatment. Other scientists argue that the anti-estrogen tamoxifen decreases the proliferation of breast cancer, although its role in the prevention of the disease is not clear.[8,9] The necessity for tissue diagnosis in these patients is made more pressing by the fact that giant breast hyperplasia may present with malignant features such as ulceration, skin dimpling, nipple inversion, and peau d'orange. Phyllodes tumor, lipoma, abscess, juvenile breast hypertrophy, breast abscess, macrocyst, and pseudo-angiomatous stromal hyperplasia are additional tumor types that could have comparable traits. It is crucial to remember that lymphadenopathy is frequently brought on by phyllodes tumors (both benign and malignant), and this should not be interpreted as a symptom of malignancy in these patients.

Gestational macromastia may result in a psychologically and physically debilitating condition. A thorough workup including endocrinology, histopathology, and hematology must be performed in order to entirely rule out underlying disease processes that can present as massive breast enlargement during pregnancy. Fetal wellbeing should be monitored whenever bromocriptine therapy is used because of the risks of intrauterine growth retardation. Cytoreduction surgery has been suggested for patients who desire future pregnancies, whereas bilateral total mastectomy is suggested for those who do not desire having children since there is an increased risk of recurrence following simple mastectomy or reduction mammoplasty.[10,11]

In conclusion, the breast can be affected by a range of different pathologies during pregnancy, including benign disorders directly associated with physiologic changes,

inflammatory and viral diseases, juvenile papillomatosis, and benign and malignant entities. Thus, in unusual cases of massive breast enlargement, the diagnosis of aggressive diseases such as breast cancer should always be considered the most important differential diagnosis. In such scenarios, the diagnosis of breast cancer may pose a challenge as a result of pregnancy-induced hyperplasia. This may cause a delay in diagnosis and thus result in poor treatment outcomes. An understanding of the typical and atypical clinico-pathological characteristics of breast lesions occurring in pregnancy and lactation is essential for appropriate patient management.

AUTHOR CONTRIBUTIONS

John Lugata: Conceptualization; data curation; methodology; writing – original draft. **Onesmo Mrosso:** Data curation; methodology; writing – review and editing. **Bariki Mchome:** Data curation; methodology; supervision; writing – review and editing. **Alex Mremi:** Conceptualization; data curation; funding acquisition; methodology; writing – review and editing.

ACKNOWLEDGMENTS

The authors thank the patient for allowing us to use her medical information for academic purpose in this publication, as well as the KCMC hospital administration, and staffs of departments of Obstetrics and Gynaecology, Radiology and Pathology for providing information in support of this study.

FUNDING INFORMATION

This work is supported by the International Cancer Institute (Kenya) as part of the Blueprint for Innovative Access to Health care through Takeda.

CONFLICT OF INTEREST STATEMENT

The authors declare no conflicts of interest.

DATA AVAILABILITY STATEMENT

There are no data generated from this study.

ETHICS STATEMENT

The patient provided written informed consent to allow for her de-identified medical information to be used in this publication. A waiver for ethical approval was obtained from the authors' institution review board committee.

CONSENT

Written informed consent for publication of clinical details and images was obtained from the patient. A copy of the consent is available for review by the chief editor of this journal.

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How to cite this article: Lugata J, Mrosso O, Mchome B, Mremi A. Gestational macromastia complicating pregnancy: A case report of unusual bilateral giant breasts in a patient who had no such problem in her previous pregnancies. *Clin Case Rep*. 2024;12:e8607. doi:10.1002/ccr3.8607