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Bilateral Gravidic Gigantomastia at Souro Sanou University Hospital, Bobo Dioulasso : A Case Report and Review of the Literature

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ABSTRACT

Introduction: *Gestational gigantomastia is a rare benign pathology, with a variable incidence of around 1/100,000 pregnancies. Its pathophysiology is poorly understood. It causes local trophic problems and makes pregnancy difficult. The authors report a rare case of bilateral gigantomastia in a normo-evolving pregnancy of 28 weeks' amenorrhea (SA), managed at the Sourô Sanou University Hospital in Bobo-Dioulasso.*

The Case: *This 37-year-old patient, 5th gesture, 4th pare, with 4 living children, was admitted to the CHUSS of Bobo-Dioulasso for rapid and painful breast enlargement after 28 weeks' amenorrhea. A diagnosis of bilateral gigantomastia in pregnancy was hypothesized. We continued to monitor the pregnancy until 37 weeks' amenorrhea. Management was essentially medical and surgical by Caesarean section. Post-operative management was straightforward.*

Conclusion: *Bilateral gravidic gigantomastia is a rare pathology. The etiology is unknown. Management is multidisciplinary, and radical treatment is based on bilateral mastectomy.*

Keywords
Gigantomastia gravidarum, Bobo-Dioulasso, Burkina Faso.

Introduction
Gestational gigantomastia is defined as a disabling increase in the size of both breasts, exceeding 1500 cm3 in volume, occurring during pregnancy [1-3]. It is a rare benign condition. Since its first description by Palmuth 372 years ago (1648), fewer than 125 cases have been reported in the literature in 2020 [4-6]. The incidence of gigantomastia gravidarum is variable, hovering around 1/100,000 pregnancies depending on the author [7-10]. It is due to hormonal phenomena in poorly defined areas of mastopathy [11]. The clinical picture is noisy, and the patient's breasts enlarge rapidly to become inflamed and painful, with a weight that is difficult to bear, making everyday gestures and pregnancy painful [12]. Few cases have been reported in black women [3]. In Niger this rarity is not documented [6]; in Senegal until 2015, three (3) cases were documented [11]. We report this unique case of bilateral gravidic gigantomastia described and managed at the Sourô Sanou University Hospital in Bobo-Dioulasso.

Observation
Anamnesis
37-year-old patient, 5th gesture, 4th pare, 4 living children, menarche at 13, admitted to the maternity ward of the CHUSS in Bobo-Dioulasso for rapid and painful breast enlargement after 28 weeks amenorrhea. She was fatigued and had difficulty performing daily activities. The evolution was marked by the appearance of bilateral ulceration of the breasts, which was very haemorrhagic, prompting consultation at the Sourô Sanou University Hospital

maternity ward for better management.

Clinical examination on admission revealed good general condition WHO stage 2, clear consciousness, pale anicteric conjunctivae. Blood pressure 120/70 mmHg, pulse 89 pls/mns, temperature 37°c.

Obstetrically, the breasts were large and rounded, with shiny orange peel skin, an ulceration measuring approximately 3x4 cm on the right breast and 3x3 cm on the left breast, highly hemorrhagic, and collateral venous circulation on the chest. The lymph nodes were free (Figure 1). Uterine height was measured at 24cm, no uterine contractions, fetal heart sounds at 140 beats/mns perceived with the Pinard stethoscope. Speculum examination revealed a healthy vaginal wall and cervix. The cervix was purplish. On vaginal touch combined with abdominal palpation, the cervix was mid-length, centered, firm and closed. The cul-de-sacs appeared free. The fingertips were soiled with physiological leucorrhoea.

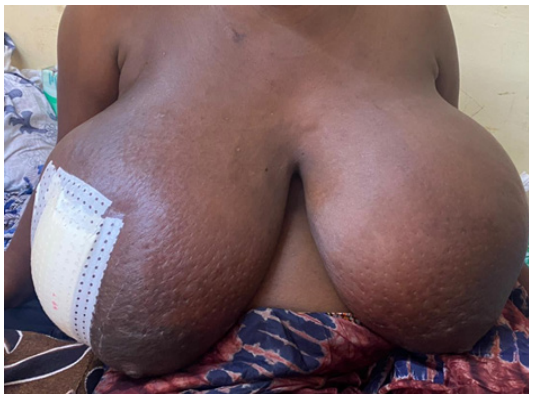

Figure 1: Bilateral gigantomastia in a progressive pregnancy of 28 weeks' gestation.

Additional Tests

Obstetrical ultrasound revealed a normo-evolving intrauterine mono-fetal pregnancy at 24 weeks' amenorrhea plus 3 days, with no morphological abnormalities.

Biological workup showed hemoglobin at 7g/dl, white blood cells at 7.5 .103/ul, platelets at 368. 103/ul. Rhesus A blood group positive. Creatinine level 42.6 µmol/l. FSH < 1mIU/ml, estrogen E2: >3000pg/mL.

Mammary ultrasound showed extensive diffuse infiltration of the mammary glands and cutaneous-subcutaneous tissue, with an "orange peel" appearance and no abscessed collection or identified mass, a priori classified as suspicious ACR4.

A biopsy showed no malignant features.

In view of the painful bilateral exuberant breast swelling in a pregnant patient, and given the results of the anatomopathological examination and ultrasound scan, we accepted the diagnosis of gigantomastia gravidarum.

Treatment

We opted to continue monitoring the pregnancy until 37 days' gestation (Figure 2). Her breasts were bandaged. With antibiotic treatment and local care, the ulcerations healed after three weeks. Anemia was treated by polytransfusion of red blood cells (approx. 1600 ml), iron supplementation with folic acid and maternal and fetal monitoring. Fetal extraction by Caesarean section was indicated and performed at 37 days' gestation under spinal anaesthesia, resulting in the extraction of a live female newborn, Apgar score 9-10-10, birth weight 2500g, head circumference 33 cm, chest circumference 30 cm, height 49 cm, placental weight 400g, cord length 48 cm. Breast-feeding was indicated.

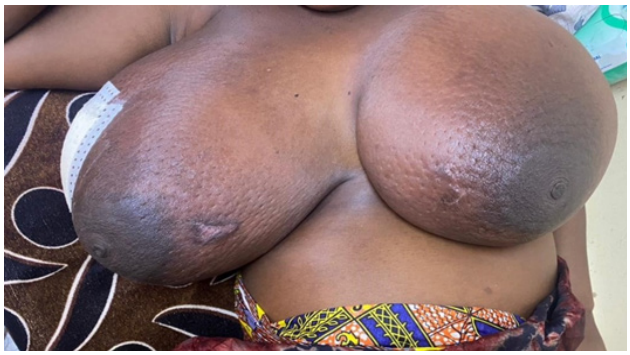

Figure 2: Bilateral gigantomastia in a progressive pregnancy of 37 weeks' gestation.

Post-operative Care

Post-operative care consisted mainly of an analgesic infusion of paracetamol 1g combined with tramadol every 6 hours for 24 hours. Anemia was corrected with an additional transfusion of 800cc of packed red blood cells. Transit had resumed by the 2nd postoperative day. After an 8-day stay to correct the anemia, she was discharged. The first dressing performed on 9-day confirmed a healed skin wound. The woman was seen again at the postoperative consultation on day 42, and the evolution was marked by a progressive, bilateral regression of breast volume in the post-partum period.

Discussion

A rare pathology, gigantomastia is an exuberant form of breast hypertrophy [3]. The etiology is not fully understood, but the usual forms of this exceptional entity are the juvenile form, which affects girls in puberty and is rarely part of Cowden's syndrome, the gravidic form, which manifests itself during the first weeks of pregnancy, and more rarely, gigantomastia may be iatrogenic, secondary to medication or associated with leukemia or lymphoma. Idiopathic gigantomastia is even more exceptional, affecting adult women over 20 years of age outside pregnancy [4,13,14]. It occurred in our patient with a progressive pregnancy of 28 weeks' amenorrhea. According to Shoma [15], in 97% of cases pregnancy appears to be the only factor responsible for gigantomastia gravidarum, which was the case in our observation. The patient's only cause was pregnancy. The mean age of onset was 26.8 years in Shoma's review [15] of 46 cases of gigantomastia gravidarum.

Our patient was 37 years old, which could be explained by the fact that she was in her 5th pregnancy. According to Chavoin [16], the age of onset is not specific, but it is frequent in multiparous women after normal first pregnancies.

Gestational gigantomastia occurs unexpectedly on previously normal breasts, during pregnancy, which it renders difficult because it is disabling [11,17]. In our case, the patient had no history of mastopathy. Gigantomastia gravidarum is most often bilateral and rarely unilateral [15,18], which was the case in our observation. Diagnosis is based on a number of factors. Imaging is poor. Ultrasound is the most effective routine examination in gravid and inflammatory situations [3]. Histology shows a florid pluristratified epithelial proliferation with papillary structures, without atypia; the epithelial cells are vacuolated, testifying to significant secretory activity [3,19]. In our case, histology ruled out malignancy and ultrasonography was poor.

Treatment is not well codified. Medical, obstetrical and surgical treatments have been proposed [19,20]. We opted for a medical treatment consisting in the management of local complications (highly hemorrhagic ulcerations) with local dressings and blood transfusions, combined with antibiotic therapy and surgical management of the pregnancy once full term. Indeed, beyond the first trimester, surveillance is recommended until the egg matures for surgical extraction [3].

Our patient had not undergone hormonal treatment; according to Eben [20], hormonal treatments based on estrogen, progesterone and testosterone have not proved effective. The prognosis is essentially local. Breast hygiene and good obstetrical follow-up are essential [11]. The prognosis was favourable in our patient, with a progressive reduction in breast volume from 32nd SA and more significant a few weeks after fetal extraction.

Conclusion

Gestational gigantomastia is a rare pathology characterized by breast enlargement with significant physical and psychological repercussions. The etiology remains unknown, diagnosis is straightforward, but treatment is not well codified. Management must be multidisciplinary.

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