Case Reports

EMERGENCY MASTECTOMY IN GIGANTOMASTIA OF PREGNANCY: A CASE REPORT AND LITERATURE REVIEW

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Enlargement of the breast during puberty and gestational periods is a normal expected physiological process. Massive diffuse enlargement of the breast during the gestational period is a rare condition and referred to as gigantomastia, as suggested by Strombeck[1] in 1964. Gigantomastia was first described by Palmuth[2] in the German literature in 1648 and by Simpson[3] in the English literature in 1920. The subject was elegantly reviewed by Beischer et al.[4] in 1989, where they described three cases and reviewed 17 cases in the literature.

We present an usual case of gigantomastia in a 36-year-old, gravida 9, with a total breast weight of 15,155 grams, complicated by ulceration and subsequent hemorrhage, which required emergency mastectomy. This was followed by right axillary breast enlargement during the postpartum period, requiring the excision of 995 grams of the accessory breast tissue.

Case History

A 36-year-old Saudi was admitted on November 6, 1995, complaining of huge enlargement of the breasts, causing her dragging pain and distress, and limiting her mobility. The patient was gravida 9, para 7 + 1, and on admission she was 17 weeks pregnant. All her previous pregnancies had been normal, apart from the last pregnancy, when she noted some degree of enlargement of her breasts, which started at 32 weeks' gestation and was well tolerated until delivery. After delivery, her breasts returned to normal size. During the current pregnancy, the patient noticed enlargement of both breasts during the first trimester, which continued increasing rapidly as the pregnancy progressed.

The patient had been diagnosed with secondary infertility due to hyperprolactinemia after her last pregnancy. Therefore she was put on several courses of bromocriptine until she became pregnant. There was no significant personal medical or family history.

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While admitted, the patient was treated conservatively with cold compresses and a supportive bra. During her stay in the hospital, her breasts continued to increase in size and blisters erupted on the left areola, with eventual ulcer formation. There were multiple ulcers measuring 2-3 cm in diameter surrounding the area of the nipple complex. They were very painful, with occasional yellowish discharge. A swab taken for culture and sensitivity yielded a light growth of *Acinobacter* species. The patient was started on antibiotics and daily dressings of the ulcers. Surgery was consulted for reduction mammoplasty. There was also a question of terminating the pregnancy. This was declined by both the surgeons and the family.

The patient had a complete laboratory work-up, including a complete blood count, chemistry, liver function tests, coagulation screen and hormonal assay. Her hemoglobin was 101 g/L and WBC was 5.6×10^9 L. All hormonal studies were normal except prolactin, which was noted to be raised on two occasions to 51.6 and 46.5 μg/L (0-25 μg/L).

Because of increase in prolactin, bromocriptine 2.5 mg was given twice daily. The breasts initially responded and became softer, but still progressive enlargement was noted. A small right axillary breast was noted; the size was approximately 4 x 5 cm. The patient's disability became apparent as the ulcerations became more prominent, with increasing venous engorgement.

Five weeks after admission the patient started to bleed profusely from the left breast, when the ulceration eroded into a major vein, leading to considerable blood loss. The bleeding was stopped by applying pressure and suturing the ulcers. The decision was then made to take the patient immediately to surgery for a bilateral mastectomy. This was done with immediate reimplantation of the areola-nipple complex of both breasts. The procedure was considered to be life-saving. A considerable amount of bleeding was encountered during the procedure and the patient required eight units of packed RBCs together with six units of fresh frozen plasma. There was no attempt to excise the right axillary breast, as this was of a small size. During the postoperative period, the patient had a minor wound infection, but this was controlled conservatively. Subsequently the patient made a good recovery.

Ultrasound of the pelvis revealed a viable, progressing, normal fetus.

The patient was discharged home three weeks later and was advised to attend both obstetric and surgical outpatient clinics. Later the patient progressed to a spontaneous, normal, vaginal delivery with a viable, healthy baby. During the postpartum period, the patient started to complain of pain and swelling in the right axillary breast, which had increased considerably, reaching a size of 12 x 10 cm. Therefore the patient was put on bromocriptine to reduce the size of the axillary breast.

Subsequent follow-up in the outpatient clinic revealed that the axillary breast had continued to increase considerably in size, therefore interfering with the normal activity of the patient's right arm. The problem was discussed with the patient and she was advised to have an excision of the axillary breast. The patient underwent the surgical excision of her axillary breast and her postoperative period was uneventful.

From the pathological point of view, the right breast weighed 8 kg and the left breast weighed 7.155 kg (Figure 1). The accessory breast weighed 956 g. Histologically, sections of the breast tissue showed extensive proliferation of medium-sized ducts, with a tendency to aggregate around large distended ducts. The stroma was severely edematous. There was no evidence of duct epithelial hyperplasia, and features were consistent with gigantomastia.

Discussion

The cause of gigantomastia remains unknown. Several theories have been discussed to explain the phenomenon. Endocrine imbalance would seem a suitable explanation. However, hormonal assay in our case, as well as in the literature,[5] failed to produce an explanation. An isolated report of hyperprolactinemia[6] similar to our case was observed, but this finding is not confirmed in any other publications.[7] Target organs, hypersensitivity or an increased number of receptors in the breast were considered as a possible explanation; however, no studies have been carried out to measure organ responsiveness. Lafreniere et al.[5] studied estrogen, progesterone, steroid and androgen receptor assays and found that all the receptor assays were normal. A cohesive theory remains to be developed to explain the condition.

Initially there is an increase in the periductal interlobular connective tissue and later, moderate lymphocytic infiltration of the hyaline connective tissue predominates. A considerable degree of interstitial edema accounts for the hugely enlarged breast. The enlargement may be associated with ulceration of the skin[8] followed by sepsis[9] or bleeding from the grossly engorged veins.[10] The sheer size and weight of the breast usually restricts mobility, causes dragging pain and, on occasion, kyphosis. Sloughing of the breast and death have been reported.[3]

As the etiology of the condition is not well understood, controversy exists regarding the therapeutic modality. As some degree of regression occurs after delivery, perhaps conservative measures could be used in anticipation of this regression. Hormonal manipulation by testosterone,[11] progesterone,[10] stilbestrol[12] and hydrocortisone[9] have been used without success. Bromocriptine[13] produced a partial response in a few cases, even with normal prolactin levels, but failed to produce a similar effect in our case, which involved high prolactin levels. Because of the observed partial response of bromocriptine, it would perhaps be worthwhile to give a trial of bromocriptine in early cases.

Therapeutic abortion was considered on the basis that regression is observed in the postpartum period. However, the poor result of therapeutic abortion combined with the unethical principle of sacrificing a life for an organ makes therapeutic abortion an unacceptable option.[5]

Surgical treatment becomes mandatory once complications occur. Infection, ulceration and hemorrhage are absolute indications. In the absence of complications, surgical indication depends on sound surgical judgment, taking into consideration the size of the breast, the degree of disability, the duration of pregnancy and the risk of miscarriage during surgery. Controversy still exists about the best surgical treatment. Reduction mammoplasty, subcutaneous mastectomy, and total mastectomy are the various surgical options. Reduction mammoplasty may be followed by long-term hormonal manipulation to prevent recurrence and will fail if subsequent pregnancies occur, because of the regrowth of the breast.[14] Subsequent mastectomy[5] will be insufficient for large breasts. Simple mastectomy[10] could possibly be the treatment of choice, particularly with immediate reimplantation of the areola-nipple complex. The surgeon must be aware of the possibility of large amounts of blood loss.

Our case exhibited a few unusual features. The patient was a 36-year-old, gravida 9 and is the oldest patient recorded to have gigantomastia. She demonstrated all complications of gigantomastia, including ulceration, sepsis and sudden massive hemorrhage necessitating emergency mastectomy. Both breasts together weighed 15,155 grams. The peculiar feature here is that the axillary breast started to enlarge only after mastectomy. The degree of enlargement was out of proportion to that expected, reaching approximately 995 grams, and required subsequent excision. We were not able to explain this phenomena. Despite hyperprolactinemia and subsequent treatment with bromocriptine, the breast continued to increase in size, making hyperprolactinemia a less likely cause for the gigantomastia.

In conclusion, gigantomastia of pregnancy is a rare condition with ill-understood etiological factors. A short

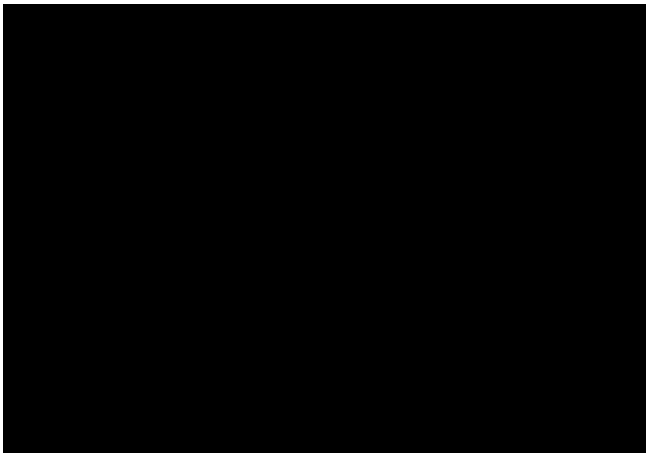

FIGURE 1. Shows the unusually enlarged breast.

trial of bromocriptine may be beneficial in anticipating regression after delivery. Surgical treatment is indicated in complications and in incapacitating conditions. We recommend total mastectomy with immediate implantation of the nipple-areola complex as the optimum surgical treatment.

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