

Case Report

Spontaneous Excessive Bleeding From a Breast Lactating Adenoma; a First Reported Case

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Abstract: Background: Lactating adenoma is a rare benign breast tumor that occurs during pregnancy or breastfeeding. Excessive bleeding from such lesion during biopsy has been previously reported in several cases. This is the first report of excessive spontaneous bleeding from a lactating adenoma triggered by the onset of labor. **Case Presentation:** We report of a case of a 33 years old African woman pregnant at 38 weeks who presented to us complaining of a rapid enlargement of her right breast for 8 months with a prior history of a diagnosis of an ipsilateral breast fibroadenoma. Physical examination revealed a 15*15cm right breast swelling with a 3*4cm ulcer on the right lower inner quadrant which had indurated margins and a shallow floor, no palpable lymph nodes. Excessive spontaneous intractable bleeding occurred from the mass during labor to necessitate an emergency mastectomy. Histological analysis of the mass confirmed a lactating adenoma. **Conclusion:** Lactating adenomas often run a silent clinical course. Redistribution of blood volume in the breast blood vessels during labor and delivery coupled with tissue necrosis by the tumor can result in excessive spontaneous loss of blood from the affected breast to necessitate emergency mastectomy.

Keywords: Pregnancy, Lactating Adenoma, Mammary Blood Flow, Doppler Velocity, Oxytocin, Case report.

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INTRODUCTION

Pregnant and lactating women are subject to different physiological changes including hormonal induced breast changes. Lactating adenoma; a rare benign breast tumor is usually seen during pregnancy and/or lactation [1]. They are often small in size about 3cm in size, solid, painless, well circumscribed, lobulated mobile nodule which grow slowly owing to their benign nature. Axillary lymph nodes are usually not palpable [2-5]. Lactating adenomas commonly affects women between 20-35 years of age, often the last trimester of pregnancy or during lactation particularly in primigravidae but seldom among multigravidas [6, 7].

Gold standard diagnosis of lactating adenomas is biopsy followed by histological analysis of the tissue sectioned, when it occurs during pregnancy, biopsy should not be delayed however excess bleeding should be anticipated [8]. Our case report is quiet peculiar because it involves a lactating adenoma that not only

had features suggestive of malignancy but also had intractable bleeding during labor and delivery that resulted in a worsening anemia necessitating emergency mastectomy and multiple blood transfusions.

CASE PRESENTATION

A 33-year-old African woman gravida 3 with 2 living children presented to us at 38 weeks of pregnancy complaining of a progressive right breast enlargement for 8 months. She noticed the enlargement during the early weeks of her pregnancy reporting rapid enlargement throughout pregnancy associated with on and off pricking pain. Two years prior to the current complaint, she experienced a right breast lump which increased in size during menstruation with regression post menses. It was a painless lump with no associated breast discharge. Excision of the mass was done with histological analysis revealing fibroadenoma of the breast.

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The current incident is a recurrent in which the painless swelling was first noted in the first trimester rapidly increasing in size as pregnancy advanced. A breast ultrasound at 28 weeks of pregnancy done at a periphery facility reported a complex large breast mass consistent with BIRAD 3 with no axillary lymph node enlargement. The patient was scheduled for Fine needle aspiration cytology however the procedure was not done due to patient's reluctance. Since then, the breast mass was noted to continue increasing in size accompanied with on and off pain. Three weeks prior to the current admission, the swelling spontaneously ulcerated with reported oozing of blood from the superficial vessels seen engorging the swelling. Through the course of current presentation, she denies history of nipple retraction or discharge and no history of fever. Besides the documented moderate anemia noted in her antenatal clinic card, her antenatal history appeared unremarkable. Family history is significant for breast masses during pregnancy as she reports that both her grandmother and mother had history of breast swelling during pregnancy that resolved during breastfeeding.

On examination, the right breast was grossly enlarged with a huge palpable mass of 15 *15 cm, hyperemic with visible distended superficial blood vessels, no peau-de orange appearance, no nipple retraction. A healed surgical scar was noted around the areola. On palpation the breast was firm but mobile with irregular margins and varying areas of mild tenderness. No discharge was noted from the nipple. An ulcer was present on the right lower inner quadrant with indurated margins measuring approximately 4 cm x 3 cm, with a shallow floor (Fig 1 & 2). Axillary lymph nodes were not palpable and the contralateral breast appeared normal. Breast ultrasound revealed a grossly enlarged right breast with areas of nodulation displaying multiple masses within the breast parenchyma with some areas of necrosis seen. Based on the above findings a provision diagnosis a breast tumor in pregnancy with differentials of a Phylloides tumor and breast cancer was suspected and planned for biopsy.



Figure 1: Shows the affected right breast with the contralateral normal left breast. Hyperemic changes, incisional scars and distended superficial veins are noted

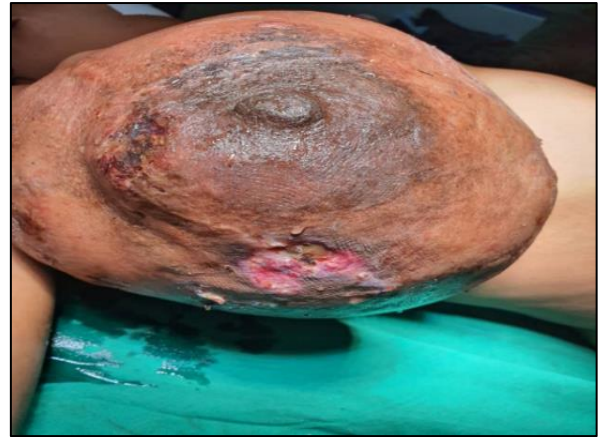


Figure 2: A preoperative image of the breast swelling with an ulcer

Her blood workup showed a hemoglobin of 7.5 g/dl (11.5-16.5); microcytic hypochromic type with normal platelets and leucocytes. The liver enzymes and renal function tests were within normal ranges. The prothrombin time; partial thromboplastin time and the INR were 11.0 seconds [11-13]; 27.3 (25.5-39.6) seconds and 0.91(<1.1) respectively. Fetal doppler scan showed a normal placental blood flow, a normal placenta in echogenicity and contour, fundal placed and anterior. Gestation age by fetal biometry was 37 weeks with an estimated fetal weight of 3.2kg ±472g and a normal amniotic fluid index.

Three units of blood transfusions and iron supplements were given to correct the anemia in preparation for delivery and breast biopsy. The hemoglobin level rose to 10.3 g/dl after a week and at 39 weeks of gestation age the patient was counselled and planned for an elective induction with 3mg of dinoprostone (prostaglandin E2 analogue).

Upon induction, as the labor was progressing, spontaneously bleeding from the superficial vessels of her right breast started. The bleeding was of high pressure and occurred concurrently with each uterine contraction, worsening during the entire active phase of labor losing about 600mls of blood from the bleeding breast vessels. Hemostasis was temporarily achieved with a tight adrenaline compression pack. Labor progressed normal and she managed to deliver a 3.1kg baby boy with an Apgar score of 8 and 10 with no complications. The 3rd stage of labor was completed successfully with an estimated blood loss of 200mls.

Seven hours after delivery, bleeding from the right breast resumed with about 300mls of blood loss estimated. Her vital signs appeared to be a blood pressure of 106/66mmHg and a pulse rate of 100b/m. The initially applied compression pack was fully soaked and was replaced by a new adrenaline compression pack. Hemostasis was temporarily achieved and an additional two units of blood were transfused to the patient. An emergency consultation was done to the

surgeon on call and a decision to prepare the patient for an emergency mastectomy was made; an additional unit of blood was prepared to be available for transfusion once hemostasis was achieved after mastectomy.

Intraoperatively, the supra-areola breast tumor measuring 30 x 30 x 10 cm was dissected and removed en – block. Estimated blood loss during surgery was 250mls. Two more units of blood were transfused and the patient came out of theatre in stable condition. The breast tissue was taken for microscopic histological evaluation and histological diagnosis of lactating adenoma with suppurative inflammation was reached (Fig 3 & 4).

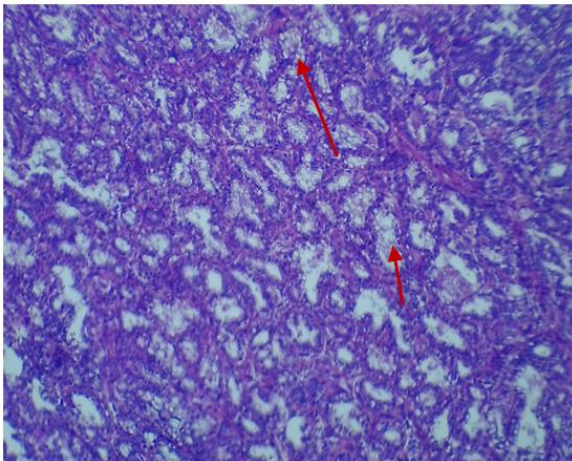


Figure 3: Section of tissue section showing multiple hyperplastic closed packed lobules with both myoepithelial and epithelial lining composed of small round cells with clear vacuolated cytoplasm and some with eosinophilic secretion as pointed in red arrows (x10hpf)

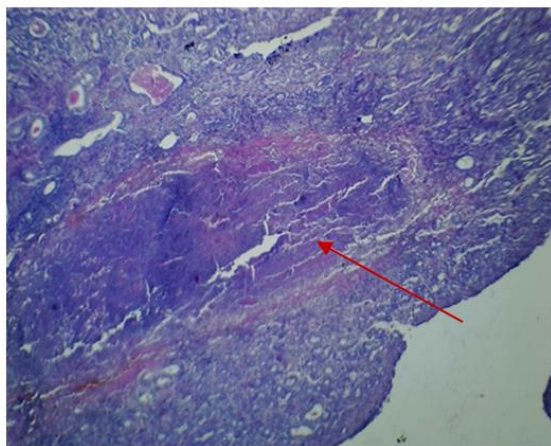


Figure 4: Section of tissue showing area of localized extensive necrosis with inflammatory cells as pointed by red arrows (x4hpf)

Postoperatively, the patient was kept under antibiotics and analgesia. The progress was good and she did not develop any complications. She was discharged after 3 days of mastectomy with her baby to

attend surgical outpatient and post-natal clinic respectively.

DISCUSSION

Lactating adenomas are the most common breast masses during pregnancy and puerperium [9]. Ducto-alveolar proliferation during pregnancy and breastfeeding are influenced by the hormonal milieu and their interplay [10].

Despite being the most prevalent tumor during pregnancy, diagnosing a lactating adenoma may not be straightforward especially in cases which point to a malignant involvement. Our patient's case is among those few because like malignant breast lesions, it presented with a rapid growing breast mass which appeared to be highly vascularized and developed an ulcer [11-13]. Moreover, tumor growth in early pregnancy in a parous woman seen in our patient is rather unique to the usual presentations of Lactating adenoma which are often seen in the third trimester and lactation.

Oxytocin released from the posterior pituitary gland not only acts on the uterus to facilitate uterine contractions during labor and parturition; but is also released during breastfeeding to facilitate milk ejection reflex [14]. In the breast, oxytocin acts on myoepithelial cells surrounding the alveoli ducts causing contraction of these cells to effect milk ejection from the lactiferous ducts.

Earlier studies have showed that oxytocin affects the volume and velocity of the blood vessels supplying the breast [15, 16]. With the contraction of the myoepithelial cells, an increase in pressure occurs within the breast resulting in compression of the vessels penetrating the breast and tumor with a resultant increase in the peripheral vascular resistance to reduce blood flow and volume within the breast via the mammary branch of the lateral thoracic artery. However, this also causes a redirection of blood in the lateral thoracic artery distal to the mammary branch causing the volume of blood in this vessel to increase substantially. Because lactating adenoma is a vascularized tumor [11] as is seen in our patients case, blood vessel dynamics within the breast and surrounding the tumor were subject to this increase in volume. When these changes are coupled with the background of tissue necrosis caused by the tumor, weakening of the vascular connective tissues was inevitable hence making the vessel walls more fragile and liable to breaking. This could possibly explain the emergence of a spontaneous yet excessive bleeding from the tumor that was seen concurrent with uterine contractions felt by the patient during labor, delivery and in the immediate postpartum. We believe that the increased blood volume and peripheral resistance triggered the cascade for bleeding from the lactating adenoma.

Breast conservation surgery was not feasible due to the mass effect brought about by the large size of the tumor and extent of breast parenchyma distortion; thus, a mastectomy was offered to the patient. Also, the aggressiveness of the bleeding which was intractable was another factor that supported the surgeon's decision of mastectomy. In the typical usual presentations, enucleation of the mass is performed [10, 13]. Conversely, bromocriptine therapy can be used preoperatively to shrink the tumor but this was not practicable in this patient because of breastfeeding and its importance especially in low income countries where infant baby formula feeding is not easily achievable. Reeves et al stressed on the beneficial effects of bromocriptine therapy use in a similar case of a patient with a giant lactating adenoma that included 3 times tumor shrinkage and less cosmetic sequela. Despite this, tumor excision could not be avoided due to the residual tumor size left [17].

Malignant potential of lactating adenomas has not been proven [17]. However a unique presentation in cases like ours calls for individualization of patient's in order to provide a patient's tailored follow up care.

CONCLUSION

Excessive bleeding from a lactating adenoma triggered by onset of labor has never been reported before. Though silent features and behaviors of lactating adenomas calls for no course of alarm in the clinical setting of a patient, altered blood flow dynamics during onset of labor together with tumor related breast changes can result in unanticipated excessive bleeding from these tumors. Histological analysis is of paramount importance to differentiate these tumors from another breast swellings because a huge lactating adenoma may clinically resemble a breast cancer.

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Timeline

The patient was admitted on April 2021, with the medical intervention and management initiated immediately. Surgical intervention was done after birth, and the patient was admitted for a total of 12 days. Preparation and completion of the case report took 11 months, with the case presented in a tumor board conference at Bugando Medical Centre and to the ethical committee after obtaining consent.

Authors' Contributions

HH and RK played equal roles in the preparation of this case report. The other co-authors contributed to the management of the patient and the

writing of the case report. All authors read and approved the final manuscript.

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The cost of care offered to this infant was waived by the hospital administration. The cost of preparing this manuscript was covered by the authors and Catholic University of Health And allied Sciences.

Ethics approval and consent to participate

Written informed consent was obtained from the patient for publication of this case report, and ethical clearance was granted by the joint Catholic University of Health and Allied Sciences/ Bugando Medical Centre Research and Ethical review committee.

Consent for publication

Written informed consent was obtained from the patient for publication of this case report and any accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal. Additionally, consent was sought and granted by the Catholic University of Health and Allied Sciences Directorate of Research and Publication to publish this work. A copy of the clearance document is also available for review by the Editor-in-Chief of this journal.

Competing interests: The authors declare that they have no competing interests.

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