

: Bilateral complex lactating adenosis: clinical presentation and management in Sudan

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Abstract

lactating adenosis are benign breasts neoplasm usually seen during pregnancy and lactation. We present a rare case of bilateral complex benign lactating adenosis in 19 year old female patient presented with lactating adenosis mimic cancer growth after giving birth. Histopathology confirm diagnosis and Simple mastectomy was the treatment of choice.

Bilateral complex lactating adenosis: clinical presentation and management in Sudan

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Clinical Key Message:

1. Menstruation and pregnancy in young females can be followed by an aggressive type of lactating adenosis mimic cancer growth.
2. A simple mastectomy can be treatment of choice for complex lactating adenosis
3. Little is known about lactating adenosis clinical presentation and management in Sudan.

Abstract:

Background: Lactating adenoses are benign breast tumors that are usually seen both during pregnancy and lactation period. There is not yet clear information about the exact pathophysiology and management of such cases in the literature. We report a case of bilateral complex growth lactating adenomas which clinically and pathologically mimic inflammatory breast carcinoma in a 19-year-old lactating woman.

Case Presentation: A 19-year-old female, primigravida presented 3 months after giving birth to a healthy baby, with huge multiple bilateral breast masses started to grow two years prior to the presentation (age

17). Clinical assessment, biopsy and Immunohistochemistry cannot rule out malignancy completely. The main diagnosis was complex bilateral lactating adenosis. A simple mastectomy of the left mass with active surveillance of the right mass has been carried out as a treatment of choice for this case.

Conclusion: Menstruation and pregnancy in young females can be followed by an aggressive type of lactating adenosis with a complex macro-histological picture that mimics cancer growth. Moreover, under the influence of puerperal hormones, bilateral breast involvement can be inevitable. A simple mastectomy can be treatment of choice for complex benign lactating adenosis when malignancy cannot be rule out.

Keywords: breast cancer, lactating adenosis, fibroadenoma

Background:

Lactating adenomas are benign tumors of the breast and are most commonly found during pregnancy and lactation [1]. They are mostly small (less than 3 cm) and slow-growing, although some cases of larger lactating adenomas have been reported in the literature as rare entities [2]. This is a very complex case of benign lactating adenosis to be reported in Sudan. However, little is known about lactating adenosis and other benign breast tumors. The origin of lactating adenomas is disputed. However, it is believed to be either De Novo or a modification of a pre-existing lesion such as fibroadenoma or tubular adenoma, reflecting the changes resulting from the physiological state of pregnancy induced mainly by hormones [1,3,4]. Nevertheless, the exact physiological mechanism of the role of hormones in such growth remains controversial; some studies have linked the rapid growth with the concentration of prolactin [5]. The heterogeneity in the glandular component of lactating adenomas may be encountered, hence the difficulty in distinguishing between lactating adenosis, benign phyllodes, and adenocarcinoma of the breast may arise [6]. Overall, lactating adenosis known to have benign behavior and does not carry a high risk of malignant transformation [7]. Lactating adenosis has got very little interest in Sudanese literature, all focus is on the malignant neoplasm of the breast, however the complex cases of benign breast neoplasm usually add some clinical complexities and management difficulties for many Sudanese clinicians which may associated with negative impact on some patients. This article aims to discuss a case of bilateral complex lactating adenosis mimic cancer features in young Sudanese female as well as review in literature and analyses the clinical practice of such kinds of complex benign neoplasms of the breast.

Case Presentation:

We report a case of a 19-year-old female, from east Sudan presented 3 months after giving birth of her first healthy baby, with huge multiple bilateral breast masses. She was very anxious about this bilateral tumor growth in her breasts and kept thinking about cancer and death according to that fast rapid growth, especially she is very young. This tumor started to grow two years prior to the presentation (Age 17). Initially the masses appeared in the right breast and shortly involved the left one. Lumps showed a gradual course initially, but a dramatic rapid growth was noticed during pregnancy. There was severe discomfort especially in the left breast, due to rapid growth and inflammation. No past history of breast trauma or family history of breast cancer. The patient started to menstruate at age of 15 year, without reporting any abnormal events in her breasts. She was of good internal and external hygiene, no history of smoking, drinking or any bad habits and no allergies. Both breasts had multiple lumps, the largest right breast mass was measuring about 7cm x 7cm, hard and nodular with no skin changes or evidence of deep structure involvement by clinical palpation. The largest left breast mass was about 7cm X 5 cm with other smaller ones occupying all 4 quadrants. There was large area of skin changes surrounds the left large lump with a large irregular inflamed ulcer below and lateral to the Left nipple with no elevated edges. Both Breasts were lactating and milk discharge from the nipple was observed with normal color.

Ultrasounds report of left breast showed multiple bilateral echogenic breast enlargements, the largest one was about 7 cm x 7.55 cm. Also, there was a 7cm x 4 cm well defined, turbid and cystic collection noted beneath the ulcerated area in the left breast [figure 1]. Dilated right breast mammary ducts were noticed. The nipple and overlying skin were intact. Multiple enlarged bilateral axillary lymph nodes (LN) were identified. The cytology report confirmed area of hyalinization with no malignant changes. Patient completed six months of

conservative management locally without any signs of improvement. The size of the tumor steadily growing, inflammation, ulceration and lymph node enlargement were getting worse with time. Consequently, left simple mastectomy had been done base on failure of conservative management and infiltration of all quadrants left breast with very huge ulcerated tumor with active surveillance of right mass. The excised specimen's histopathological report revealed macroscopically an exophytic growth measuring 7cm x 6 cm lateral to normal looking nipple with skin ulceration. Cross sectioning of the specimen showed no breast tissue, only lobulated firm mass with central cyst behind the nipple measuring 10cm x 7cm x 5 cm in diameter. Microscopically sections showed enlarged lobules, with complex glands composed of inner actively secreting epithelial cells with vacuolated cytoplasm and apical cytoplasmic blebs, and outer myoepithelial layer [Figure 2, 3]. There was no cytological atypia and stroma was scanty. There were 2 reactive isolated axillary lymph nodes identified during histology, the right breast showed same histological features [Figure 4, 5]. P63 and S100 were positive. The immunohistochemistry confirmed a diffuse tumefactive lactating adenosis with very complicated histological picture. Eventually, Six month follow up was arranged and overall prognosis after surgery was satisfied.

Discussion:

The histogenesis of lactating adenoma is somewhat controversial. In some studies, the lactating adenoma is considered to be a pure lesion that is morphologically distinguishable from tubular and fibroadenoma and is associated with either pregnancy or lactation. The tumors may not exhibit lactational secretions [7,8]. However, they do exhibit secretory changes that mimic the physiological changes manifested in pregnancy; that is why they proposed to name them “tumors of pregnancy”. Scholars such as Hertel et al. [9] believed that lactating adenomas have precursors, including fibroadenoma and tubular adenomas. These lesions then undergo secretory changes associated with physiological states of pregnancy. These give rise to controversies that may belong to different nature of the growth which is influenced by the effect of hormones on the breast both during pregnancy and lactation period. However, World Health Organization recognizes both views in their latest book on breast tumors [10]. About 3% of breast cancers are diagnosed during pregnancy [11]. And due to the physiological changes that occur in the breast during pregnancy, the possibility of the presence of or the coexistence of malignant lesions should not be neglected [12].

Many cases of lactating adenoma that have been mentioned in the literature were of atypical presentation. Some of them are even with an aggressive presentation. Though lactating adenomas are rendered to benign behavior, the literature has mentioned the coexistence of benign and malignant tumors in the same patient [12,9,13], others in the same anatomical site [14]. Hence, caution must be taken while dealing with lactating adenoma when the patient comes with atypical presentation or with signs and symptoms suspicious of carcinoma as in this case, where she was presented with features that mimic inflammatory breast carcinoma.

The relation between the effect of females' hormones and breast tumors is not well understood and many studies have subjugated this complex relation to intense investigations. Gill S et, al. concluded in their study that a high prolactin concentration has a positive correlation to female breast tumors [5]. However, the exact mechanism remains very sophisticated [5,15]. In the case of our patient, the complicated growth picture, enlarged LN, and the presence of ulceration may be attributed to the hormonal changes during pregnancy and lactation, especially the growth picture in this case seems against the classical histology of lactating adenosis that appears as well-circumscribed, lobulated, solitary or multiple, gray with areas of necrosis; microscopically, they are seen as cuboidal cells with actively secreting, closely packed glands [16]. There has been a strong recommendation for using triple assessment in such cases in order to differentiate between adenoma and carcinoma of the breast [16,17], but even though with such complex growth the distinguishable point from cancer clinically remains very difficult and only histology can give clear differentiation. The detection of positive P63 in the histopathology has been linked to underlying ductal carcinoma in situ and some cases of invasive ductal carcinoma, and this add sounds of doubts for future behavior of this benign tumor [17], a simple mastectomy is the safest option for huge ulcerated lactating adenoma which is resistant to conservative management.

Conclusion: Menstruation and pregnancy in young females can be followed by an aggressive type of lactating

adenosis with a complex macro-histological picture that mimics cancer growth. Moreover, under the influences of puerperal hormones, bilateral breast involvement can be inevitable. The complex growth of giant lactating adenosis can be missed as cancer even with usual triple assessment, and the immunohistochemistry is the mainstay of differentiation, and with the suspicion of underlying DCIS, the safest option of treatment of such cases with complex growth is simple mastectomy which can give much promising prognosis. Eventually, this case sheds light on the complex growth of benign breast tumors and the effect of hormonal changes during both pregnancy and lactation, and it is the effect on the pathogenesis of tumors and bilateral complex growth.

Lactating adenomas are benign tumors of the breast and are most commonly found during pregnancy and lactation. This is a very complex case of benign lactating adenosis reported in Sudan. However, Fibroadenoma is one of the commonest benign breast tumors in young females in Sudan, but little is known about lactating adenosis and other benign breast tumors.

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Figures



Figure 1: growth section of the left breast mass after simple mastectomy, showing ulcerated huge lactating adenoma not responding to conservative management.

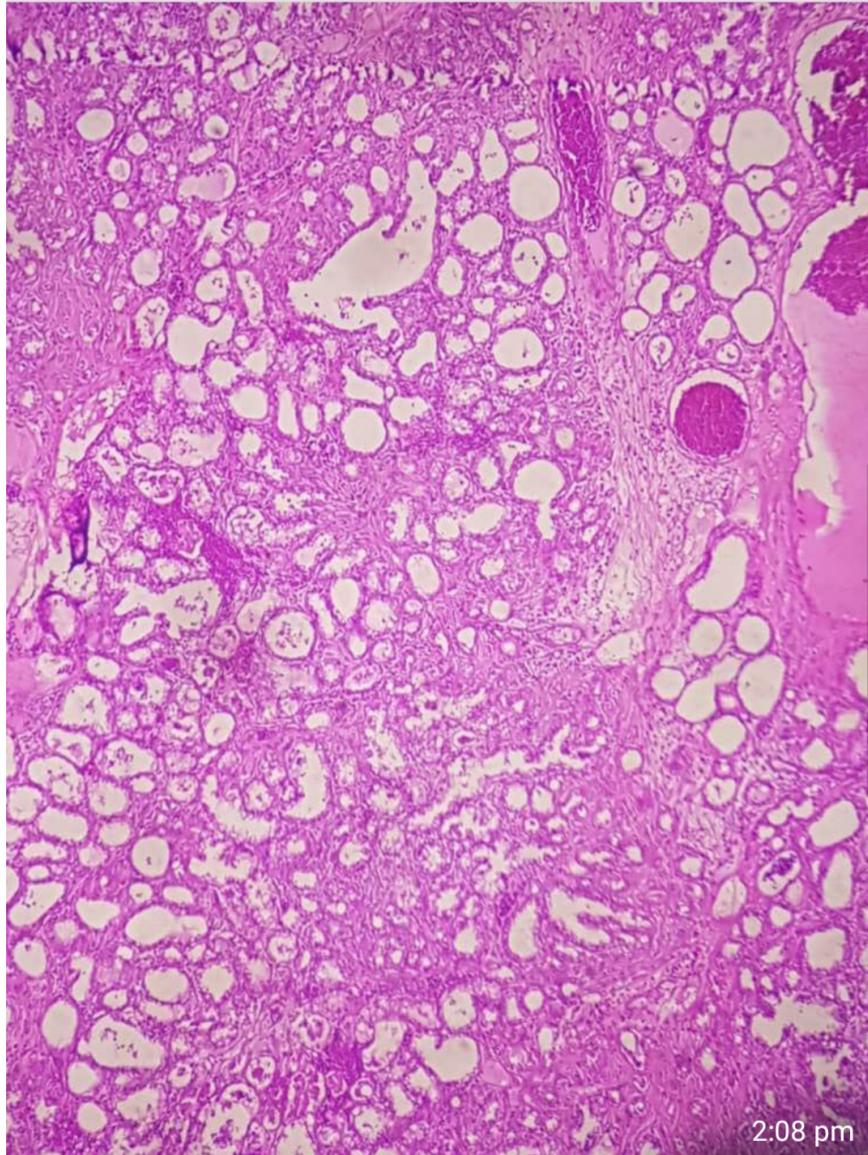


Figure 2: left breast medium power view blebbing and secretory changes in complex glandular architecture

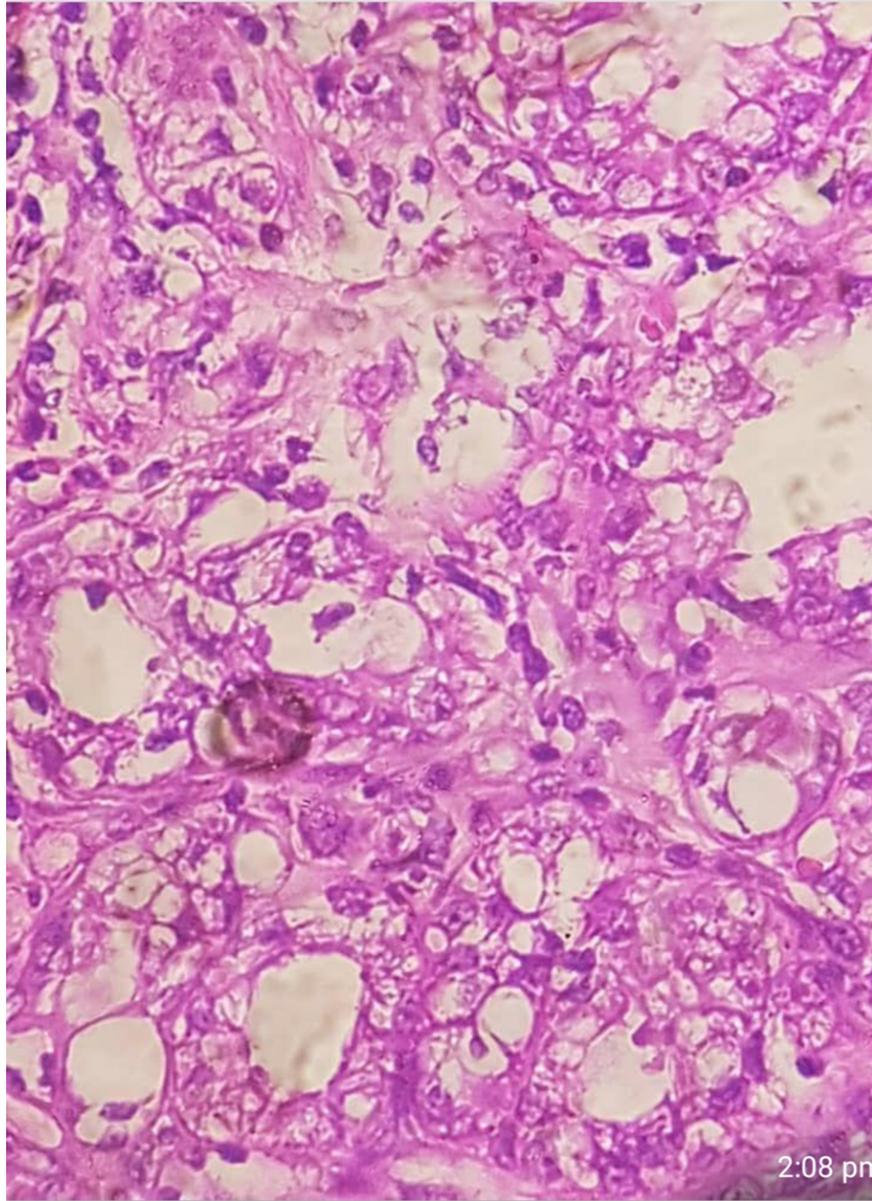


Figure 3: left breast High power view, bland cytological features with cytoplasmic vacuulations and secretory changes.

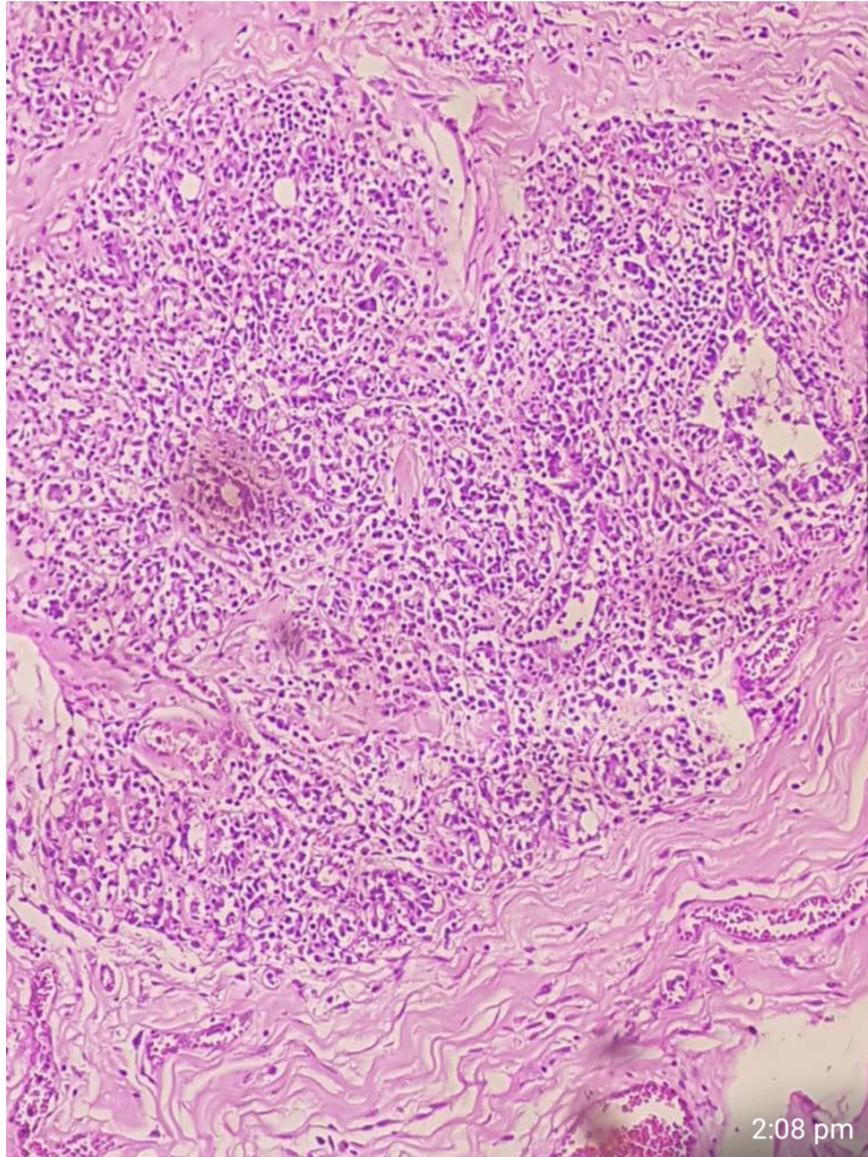


Figure 4: Right breast Medium power view shows an enlarged lobule filled with glands

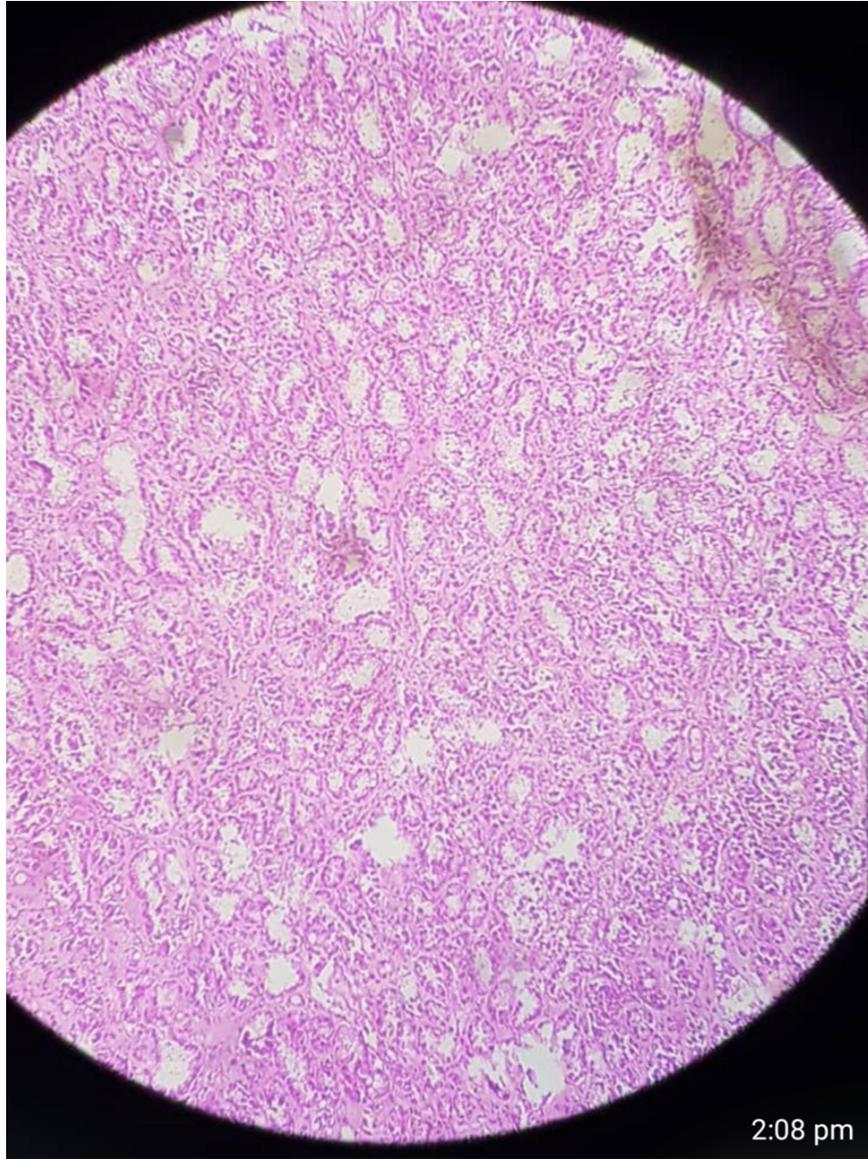


Figure 5: right breast low power view shows a neoplasm with densely packed glands