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Available online at: http://www.iajps.com *A Case Report*

RETROAREOLAR EPIDERMAL INCLUSION CYST; A UNIQUE PRESENTATION IN AN ADULT FEMALE

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Abstract:
Triple assessment approach for a breast mass may not always provide a definite diagnosis. Herein, we report a case of an unusual retro-areolar intramammary lump in a 47-year-old female, proved on excisional biopsy as an epidermal inclusion cyst (EIC). Deep-seated EIC rarely occurs in the breast and can cause diagnostic confusion even though a meticulous triple assessment approach and require a lumpectomy procedure both to establish a definitive diagnosis and relieve the patients' anxiety.

Keywords: *Epidermoid inclusion cyst, Breast cyst, Breast lump, Triple assessment approach, Lumpectomy, one-stop breast clinic.*

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INTRODUCTION:

Epidermal inclusion cysts (EICs) are the most frequently encountered cutaneous cysts in healthcare, it forms a benign skin lesion, formed by a cystic mass containing keratin. [(1)] These cysts classically present as nodules or masses directly below the affected skin, often have a visible central punctum, can occur anywhere on the body and are usually mobile. EIC range in size from a few millimetres to several centimetres and can progress over time or stay stable. [(2)] There is no method to predict if an EIC will become larger, inflamed or remain quiescent, However, once infected it tends to become larger, erythematous or painful, so it gets the patient's attention. [(2)] This type of cyst usually prevails in young and middle-aged adults, without gender predilection. Generally, EICs form when a build-up of bacteria and keratin occurs in a hair follicular orifice, leading to its cystic expansion and entrapment of keratin debris. [(3)] EICs found in the breast parenchyma are a rare entity, possess a different pathophysiologic aetiology, and when they do occur in the breast, they can cause a diagnostic dilemma until proved histologically at excisional biopsy. [(4)]

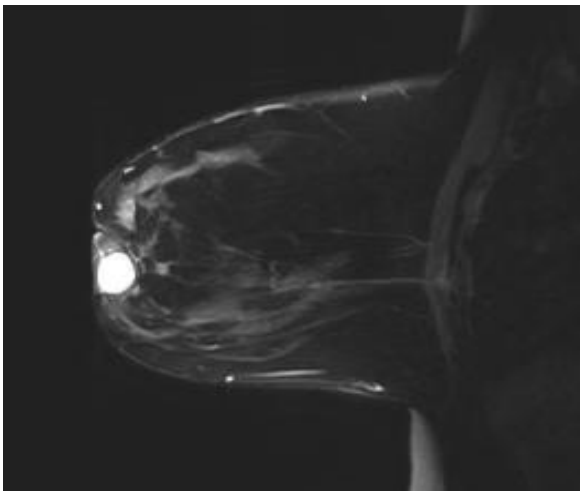
CASE PRESENTATION:

A previously healthy 47-year-old female, who is married with 2 children born by caesarean section, both were breastfed and she had no medical co-morbidities or another surgical history, presented to the one-stop breast clinic in King Hamad University Hospital with a two months' history of a mildly painful right breast lump behind the upper part of the nipple-areola complex. The lump was rapidly increasing in size, mobile and becomes more prominent before her monthly menstrual cycle. No associated nipple changes or discharge was perceived and neither past medical/surgical history nor positive family history of breast cancer was found. Her menarche was at age eleven. She had never been on oral contraceptive pills or hormone replacement therapy.

On general physical examination, she looked healthy and of normal weight; breast examination revealed a 20x20 mm, palpable, well-circumscribed, peri-areolar right breast oval lump. It was freely movable, not associated with any skin or nipple changes and no attachment to underlying structures is seen.

Mammography and U/S scan done and showed: Right retroareolar oval shaped parallel isoechoic hypovascular mass lesion with few internal calcific foci and posterior acoustic enhancement, the possibilty of intraductal papillary lesion can't be excluded. (BIRADS 0)

Mildly suspicious small Right 7 o'clock mass lesion as described above BIRADS 4A. After discussing the case with the radiologist, a bilateral breast MRI was advised which was preformed and showed: Right retroareolar (looking to be intra-ductal) circumscribed oval shaped mass lesion measuring about 25 x 20 mm, displaying rim enhancement. It displays low T1 signal, high T2 & STIR signal, restricted diffusion of the cavity, the enhancing wall shows type 1 rising kinetic curve suggestive of a possible complicated cyst (infected focal duct ectasia?) rather than remote possibility of papilloma (BIRADS 3). Very few tiny cysts seen scattered in both breasts (BIRADS 2). [fig.1 & 2]



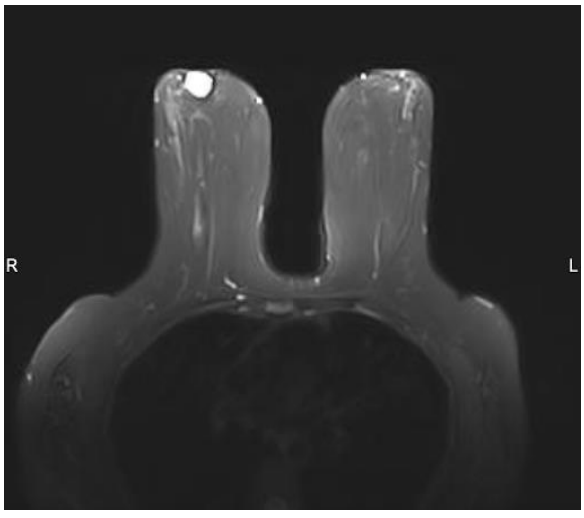


Figure 1 & 2. Right breast MRI showing retroareolar (looking to be intraductal) circumscribed oval shaped mass lesion measuring about 25 x 20 mm, displaying rim enhancement.

U/S guided Tru-Cut biopsy was taken and the pathology findings were those of a [B2] benign mammary tissue with chronically inflamed dense fibrocollagenous stroma, mixed periductal lymphoplasmacytosis and excess stromal polymorph eosinophils. Part of inflamed ductal epithelium is found in one of the three cores and was associated foamy histiocytic/macrophages tissue reaction.

A subsequent excisional biopsy was performed. [fig. 3] The gross histopathological examination of the excised breast lump measured 30 x 20 x 10 mm and the cut surface revealed a 10 x 10 x 8 mm cystic lesion with luminal creamy white paste. Microscopic examination revealed a completely excised, ruptured epidermal inclusion cyst with associated florid epithelioid foreign-body type histiocytic granulomatous reaction toward spilled luminal keratinous content [fig. 4 & 5]. No abscess formation or evidence of specific infection is seen. Of particular interest, was also the presence of a nearby duct ectasia with inspissated thick granular luminal content and minimal periductal stromal fibrosis with mild chronic periductal inflammation [fig. 6]. No ductal epithelial hyperplasia was found. The associated findings of adjacent duct ectasia helped us to confirm the hypothesised link of such deep seated EIC developmental aetiology.

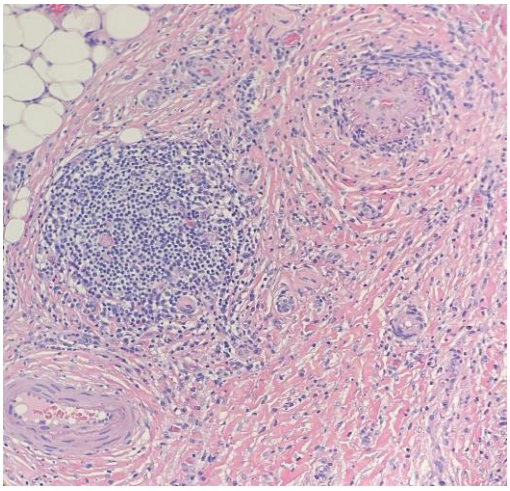


Figure 3. Intra-operative image of the retro-areolar mass lesion

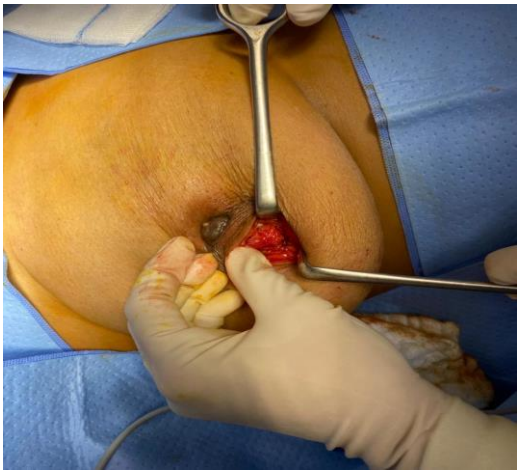


Figure 4. Mammary Excisional Biopsy [inflamed epidermal inclusion cyst with residual stratified squamous epithelium. (Low Power Field, H & E Stain).

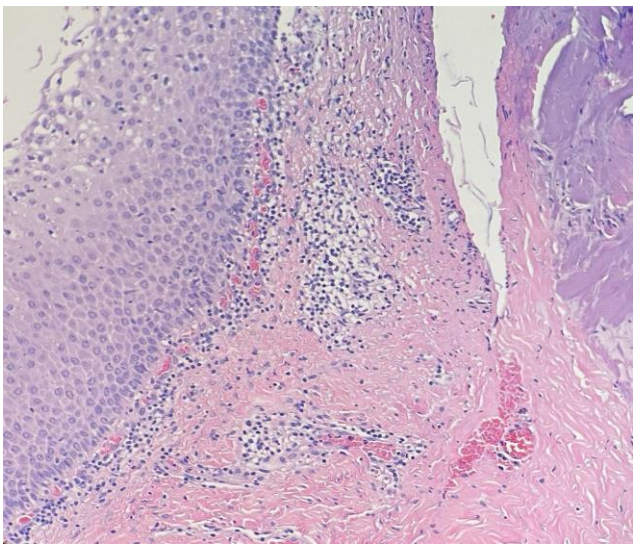


Figure 5. Mammary Excisional Biopsy [ruptured epidermal inclusion cyst with associated florid epithelioid foreign-body type histiocytic granulomatous reaction] (High Power Field, H & E Stain).

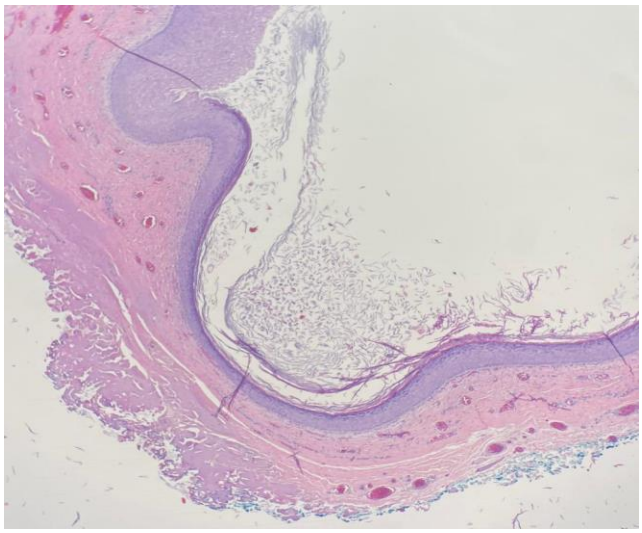


Figure 6. Duct ectasia with inspissated thick granular luminal content and minimal periductal stromal fibrosis with mild chronic periductal inflammation. Note the dilated duct ectasia (right top corner). (High Power Field, H & E Stain).

DISCUSSION:

Epidermal inclusion cysts (EIC) in the breast are a very rare entity. [5] Up and until recently, the international literature has described 90 cases of patients who have been diagnosed with mammary EIC. Furthermore, the first histologically confirmed case of mammary EIC was reported in 1900 at The Johns Hopkins Hospital (Baltimore, MD, USA). [6] From the literature, it can be determined that EIC of the breast mostly affects patients in their fifties, and a few of the cases had male patients who were subsequently diagnosed with EICs. [7] This type of cyst is naturally slow-growing and asymptomatic; however, spontaneous rupture of large cysts was noted in a small proportion of cases, releasing non-absorbable keratin that irritates the surrounding tissue and further leads to secondary granulomatous reactions secondary infection with or without abscess formation. History of trauma or prior local surgery appears to be the main associated most comprehended reason for the development of this

benign cystic lesion in but other aetiologies have been suggested.

EIC of the breast may also develop from obstructed hair follicles, but It is also believed that in many cases, EIC of the breast may be congenital, arising from the remaining cells of the embryonic mammary ridge. Furthermore, EICs has been hypothesised as an end result of squamous metaplasia of normal columnar cells within a dilated duct in the case of chronic inflammatory conditions such as fibrocystic change, duct ectasia as well as in fibroadenoma or phyllodes tumours. [8] When it comes to the diagnosis EICs, physical examination no matter how thorough, is still unreliable. [7] Ultrasonographic imaging consistently achieved an accurate diagnosis [9] whilst mammography achieved 79% accuracy in the reported cases [10] Magnetic resonance imaging was also observed to consistently identify EIC of the breast accurately. [11] It was identified that the association between EIC of the breast and malignant tumours was 12%. Malignant transformation appears to occur more frequently in EIC of the breast as opposed to EIC in other body parts, and this may be associated with squamous metaplasia of the mammary duct epithelium typically lined by columnar epithelium. [5] When the diagnosis of EIC is established, being asymptomatic and small (<2 cm) sized, no necessity for active treatment is indicated. Especially if typical on ultrasonographic findings, however, mammary EIC that is palpable and causing patient physical and psychological discomfort, requires surgical excision with the removal of the entire cyst wall for pathological examination and to prevent recurrence or the remote risk of malignant transformation. [12] EICs have a great prognosis after the complete excision of the whole cystic wall including its contents. [1]

CONCLUSION:

In conclusion, the diagnostic dilemma while dealing with a suspected breast malignancy is not infrequent. In our case, the presence of EIC in a close-by area of duct ectasia and associated chronic inflammation made us consider the metaplastic aetiology for such lesions and not just being a result of an obstructed hair follicle or as a consequence of local trauma, as it is the case for most dermal counterpart.

From a healthcare system perspective, prompt referral, diagnosis and definitive treatment by a specialised breast surgeon for mammary lumps would lead to a single procedure reduce overall healthcare costs and a better level of patient satisfaction with favourable patient outcomes.

Conflict of Interests:

All authors have no conflict of interest relating to the manuscript.

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