

Received: 2018.12.03
Accepted: 2019.02.22
Published: 2019.04.24

Clinical and Imaging Features of a Ruptured Epidermal Inclusion Cyst in the Subareolar Area: A Case Report

Authors' Contribution:
Study Design A
Data Collection B
Statistical Analysis C
Data Interpretation D
Manuscript Preparation E
Literature Search F
Funds Collection G

ABDEF 1 **Suk Jung Kim**
BD 2 **Woo Gyeong Kim**

1 Department of Radiology, Haeundae Paik Hospital, Inje University College of Medicine, Busan, South Korea
2 Department of Pathology, Haeundae Paik Hospital, Inje University College of Medicine, Busan, South Korea

Corresponding Author: Suk Jung Kim, e-mail: pure0815@hanmail.net
Conflict of interest: None declared

Patient: Female, 58
Final Diagnosis: Epidermal inclusion cyst
Symptoms: Bloody nipple discharge
Medication: —
Clinical Procedure: —
Specialty: Radiology

Objective: Rare disease





Background: Epidermal inclusion cysts rarely develop in the breast. The cysts that do develop within the breast typically present as cutaneous or subcutaneous cysts. They more rarely present in a subareolar location or in a ruptured state. Thus far, 5 cases of ruptured epidermal inclusion cysts in subareolar locations have been reported in the English literature. Furthermore, clinical presentation of nipple discharge is rare in epidermal inclusion cysts of the breast; only 4 such cases has been reported.

Case Report: A 58-year-old female presented with a 1-month history of bloody discharge from her left nipple. Mammography showed focal asymmetry in the left subareolar region; sonography showed a left subareolar mass with irregular shape, indistinct margin, heterogeneous echogenicity, and posterior enhancement. The mass was surgically excised; a pathological diagnosis of ruptured epidermal inclusion cyst with foreign body reaction and abscess formation was established. In this case, the clinical presentation of bloody nipple discharge was peculiar; furthermore, mammographic and sonographic features were indistinguishable from breast malignancy or typical breast abscess.

Conclusions: A ruptured epidermal inclusion cyst can present in an unusual subareolar location, combined with bloody nipple discharge; importantly, this can radiologically resemble breast malignancy.

MeSH Keywords: Breast • Epidermal Inclusion Cyst • Mammography • Ultrasonography, Mammary

Full-text PDF: <https://www.amjcaserep.com/abstract/index/idArt/914412>

 1419  1  3  26



Background

Epidermal inclusion cyst is a benign cutaneous or subcutaneous lesion lined by stratified squamous epithelium and filled with keratin debris [1]. This type of cyst develops as a result of the proliferation and implantation of epidermal elements within a circumscribed space in the dermis [2]. Its occurrence in the breast is rare; when it does affect the breast, this type of cyst is primarily localized in the skin layer and periareolar region [2]. There have been 15 cases reported in which an epidermal inclusion cyst occurred in the sub- or peri-areolar area [3]. This report describes a ruptured epidermal inclusion cyst of the breast with an unusual location within the subareolar area, as well as its unusual clinical presentation with bloody nipple discharge. Furthermore, this report describes the mammographic and sonographic features of this cyst, which mimicked those of breast malignancies.

Case Report

A 58-year-old female presented with left bloody nipple discharge that had occurred for 1 month. Her medical history included medications for diabetes mellitus and hypertension. She had no family history of breast cancer. There was no associated history of trauma. Upon clinical examination, erythematous change was noted in the skin of the left nipple-areolar complex. Mammography showed left subareolar focal asymmetry (Figure 1A, 1B). No microcalcifications were observed alongside the focal asymmetry on mammography examination. Sonography showed a 1.8-cm subareolar mass with irregular

shape, indistinct margin, heterogeneous echogenicity, and posterior enhancement (Figure 2A, 2B). Color Doppler sonogram revealed increased vascularity within the mass and adjacent tissue (Figure 2C). The clinical feature of the bloody nipple discharge, mammographic feature of focal asymmetry in the subareolar region, and sonographic feature of an irregular indistinct subareolar mass with increased vascularity indicated a subareolar abscess or breast malignancies; based on these findings, the cyst was classified as category 4b according to the Breast Imaging Reporting and Data System (BI-RADS) classification. The mass was surgically excised, which led to a pathological diagnosis of ruptured epidermal inclusion cyst with foreign body reaction and abscess formation (Figure 3A, 3B).

Microscopically, the cystic lesion was lined with benign stratified squamous epithelium filled with abundant lamellated basket-weave keratin. Adjacent tissue exhibited inflammatory infiltrate cells with clusters of multinucleated giant cells; this was indicative of a foreign body reaction.

Discussion

Epidermal inclusion cysts are formed by inclusion of keratinizing squamous epithelium within the dermis [4]. Epidermal inclusion cysts have also been described by other terms, such as follicular infundibular cysts, epidermal cysts, and epidermoid cysts [5]. These cysts are commonly cutaneous or subcutaneous, and present in hairy body areas such as scalp, neck, and trunk [5–7]. They rarely occur in the breast; Menville et al. reported the first case of epidermal inclusion cyst of the breast in 1900 [2,8].

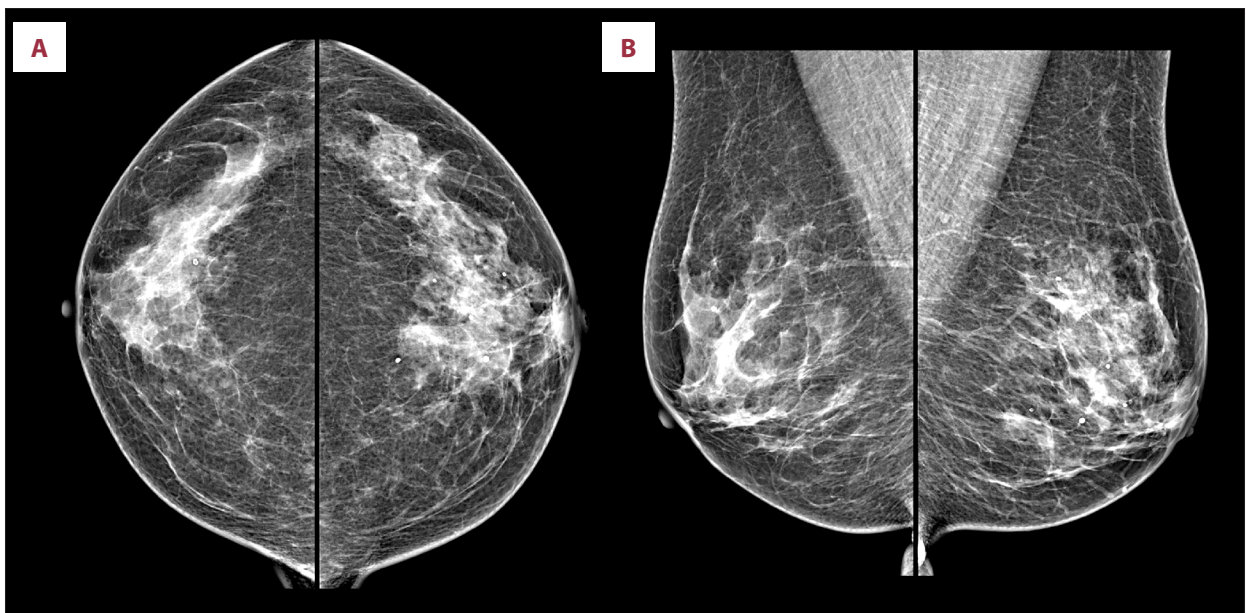


Figure 1. (A) Craniocaudal and (B) mediolateral mammograms show a focal asymmetry in the left subareolar area. No microcalcifications are associated with the focal asymmetry.

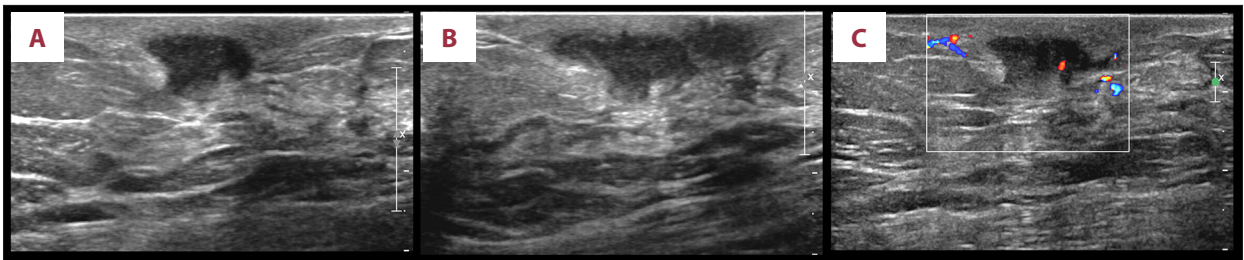


Figure 2. (A) Transverse and (B) longitudinal gray-scale sonograms show an irregular indistinct heterogeneous hypoechoic mass in the left subareolar area. Note the posterior enhancement deep to the mass. (C) Color Doppler sonogram shows a degree of increased vascularity within the mass and adjacent tissue.

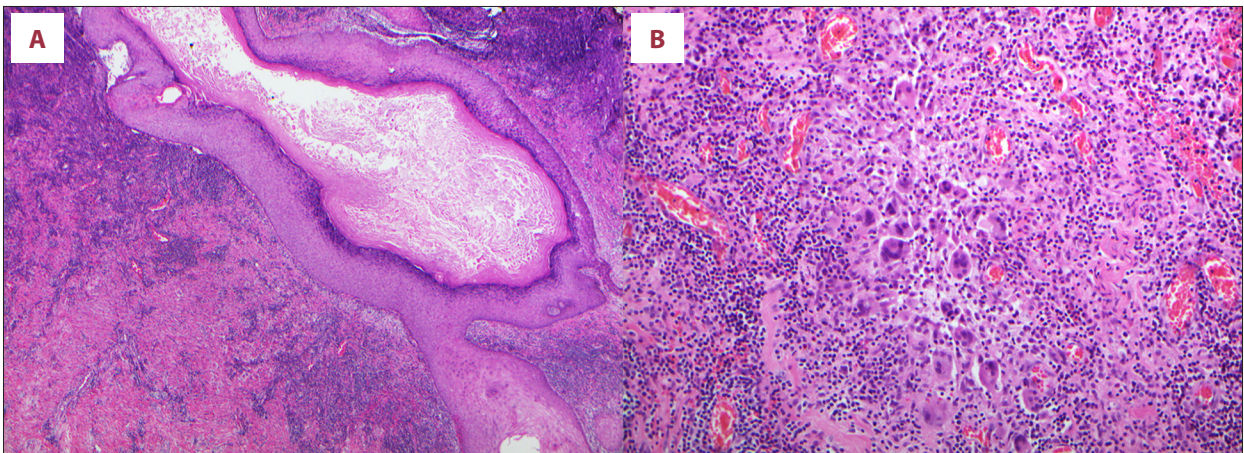


Figure 3. Microscopic findings of the epidermal inclusion cyst. (A) A cystic lesion is lined by benign stratified squamous epithelium filled with abundant lamellated basket weave keratin (hematoxylin and eosin staining 40 \times). (B) Adjacent tissue exhibits inflammatory infiltrate cells with clusters of multinucleated giant cells, indicative of a foreign body reaction (hematoxylin and eosin staining 200 \times).

Paliotta et al. identified a total of 91 patients affected by epidermal inclusion cysts of the breast through a search of the literature in 2014 [2], and thereafter, sporadic case reports of epidermal inclusion cysts have been published until recently [7,9–15]. Epidermal inclusion cysts of the breast typically develop during the fifth decade of life [2], and are usually located in the skin; however, they can develop in unusual locations, such as cutaneously on the nipple or areola or intra-parenchymally in the sub- or peri-areolar regions. Through a search of the literature, we identified a total of 21 cases where epidermal inclusion cysts developed in these unusual locations; 15 in sub- or peri-areolar region [1,3,5,12,16–19], 4 on the nipple [11,13,14,20], and 2 on the areola [10,11] (Table 1). Furthermore, we found presentations of nipple discharge in a total of 4 cases in the literature; 3 cases in sub- or peri-areolar regions [3,12,16] and 1 case on the nipple [14]. The nature of nipple discharge in these cases was not specifically described; however, it was briefly mentioned as non-bloody in 1 case [12], purulent in 1 case [16], yellowish in 1 case [3], and infected in 1 case (*Staphylococcus aureus* isolated from discharge culture) [14]. Similar to the current case, all these previous cases showing nipple discharge were associated with complications such as rupture [3,12,16],

infection [14], inflammation [3], or abscess [16]. To our knowledge, the current case is the fifth presentation of nipple discharge (and the first one presenting as a bloody nature) with an epidermal inclusion cyst of the breast and the 16th report of an epidermal inclusion cyst with sub- or peri-areolar location in the breast.

When epidermal inclusion cysts affect the breast, their clinical appearance commonly comprises that of palpable lumps (79%); less commonly, symptoms include local discomfort (67%), inflammation (33%), spontaneous rupture (12%), and ulceration (4%) [2,21].

Actual pathogenesis in the formation of epidermal inclusion cysts is not completely understood. However, several etiologies may be involved [1,5,22]. First, they are assumed to be congenital in the majority of cases [2,5]. Second, they can result from obstructed hair follicles [1,2,22]. Third, they can be derived from implanted epidermal fragments within breast tissue due to trauma such as reduction mammoplasty or needle biopsy [2,6,19,23,24]. Fourth, inflamed pilosebaceous structures can result in a cystic reaction in the dermis [2,5]. Fifth, they can

Table 1. Previous reports of epidermal inclusion cysts involving the nipple-areola or sub- or peri-areolar region.

Case no.	Authors	Published year	Age	Sex	Location	Size	Duration	Clinical presentations	Nipple discharge	Trauma history	Complication at presentation	Sonographic features
1	Amrani et al. [11]	2018	9 months	M	Areola	1 cm	Since birth	White, dome-shaped, soft, fluctuant, non-tender	Absent	Absent	Absent	Not available
2	Amrani et al. [11]	2018	3 months	M	Nipple	2 cm	Since birth	White, dome-shaped, freely movable, nontender, smooth surfaced	Absent	Absent	Absent	Not available
3	Ben Naftali et al. [10]	2018	44 years	F	Areola	4 cm	A few months	A solid polypoid irregular mass	Absent	Absent	Present (chronic inflammation)	A mass with local edema and an axillary lymph node with a thick cortex (BIRADS IVa)
4	Martin et al. [12]	2014	42 years	F	Subareolar	3.75 cm	Not available	Nipple discharge	Present (non-bloody)	Present (reduction mammaplasty)	Present (rupture)	A solid, well circumscribed ovoid mass
5	Marchesi et al. [13]	2014	39 years	M	Nipple	1.3 cm	5 months	An enlarging cutaneous lesion (exophytic, polypoid protuberance)	Absent	Absent	Absent	Cystic lesion
6	Dilek et al. [14]	2014	27 years	F	Nipple	0.4 cm	2 years	Painful, white, soft, immobile, firm, smooth surfaced	Present (isolation of staphylococcus from the discharge)	Absent	Present (infection, staphylococcus isolated from nipple discharge)	Well circumscribed, central hyperechoic mass
7	Jain et al. [20]	2012	15 months	F	Nipple	0.8 cm	2 months	Well circumscribed, pearly white, dome-shaped, soft, non-tender	Absent	Present (squeezing the breast during neonatal period to express the witch's milk)	Absent	Not available
8	Singh et al. [16]	2012	60 years	F	Peri-areolar	1 cm	15 days	Firm, mobile non-tender, partially adhered to the skin	Absent	Absent	Absent	Not available
9	Singh et al. [16]	2012	30 years	F	Peri-areolar	1 cm	6–7 years	Firm, mobile, mildly tender, partially adhered to the skin	Absent	Absent	Absent	Not available

Table 1 continued. Previous reports of epidermal inclusion cysts involving the nipple-areola or sub- or peri-areolar region.

Case no.	Authors	Published year	Age	Sex	Location	Size	Duration	Clinical presentations	Nipple discharge	Trauma history	Complication at presentation	Sonographic features
10	Singh et al. [16]	2012	38 years	F	Peri-areolar	1.5 cm	2 months	Firm, mobile, non-tender	Absent	Absent	Absent	Not available
11	Singh et al. [16]	2012	32 years	F	Peri-areolar	1.5 cm	2 months	Firm, partially mobile, tender, pus-discharging sinus	Present (purulent)	Absent	Present (rupture, infection)	Not available
12	Singh et al. [16]	2012	32 years	M	Peri-areolar	1 cm	1 months	Firm, mobile, non-tender, partially adhered to the skin	Absent	Absent	Absent	Not available
13	Singh et al. [16]	2012	25 years	M	Peri-areolar	1 cm	2 months	Firm, mobile	Absent	Absent	Absent	Not available
14	Lee et al. [5]	2012	47 years	F	Subareolar	> 8 cm	6 months	Firm, well circumscribed, attached to the skin	Absent	Absent	Present (micro-rupture)	Solid, hypoechoic mass with heterogeneous echoes and well demarcated border
15	Debanath et al. [17]	2012	69 years	F	Subareolar	8.5 cm	3 months	Painful, firm, nipple retraction	Absent	Present (traffic accident)	Present (inflammation)	Solid mass extending into the ductal system (highly suspicious for malignancy)
16	Whang et al. [3]	2007	50 years	F	Subareolar	1.8 cm	1 week	Subareolar pain, palpable mass	Absent	Not available	Present (rupture)	Ill-defined, heterogeneous, hypoechoic, irregular shaped mass
17	Whang et al. [3]	2007	44 years	F	Subareolar	2.2 cm	Several months	Periareolar pain	Present (yellowish)	Not available	Present (rupture, inflammation)	Ill-defined mass with irregular shape, heterogeneous echogenicity, posterior enhancement
18	Kwak et al. [1]	2004	23 years	F	Subareolar	4.4 cm	4 years	Painful, palpable mass	Absent	Absent	Absent	Well-defined heterogeneous echoic mass, no blood flow within the mass
19	Stephenson et al. [18]	1987	52 years	F	Subareolar	0.8 cm	Not available	Unremarkable	Absent	Absent	Present (Paget's disease)	Not available

Table 1 continued. Previous reports of epidermal inclusion cysts involving the nipple-areola or sub- or peri-areolar region.

Case no.	Authors	Published year	Age	Sex	Location	Size	Duration	Clinical presentations	Nipple discharge	Trauma history	Complication at presentation	Sonographic features
20	Gerlock [19]	1974	41 years	F	Subareolar	2.5 cm	2 years	Painful, Hard, sharply marginated	Absent	Present (needle biopsy)	Absent	Not available
21	Gerlock [19]	1974	62 years	F	Subareolar	1.5 cm	5 years	Painful, firmly affected to the skin	Absent	Present (needle biopsy)	Absent	Not available
22	Current	2019	58 years	F	Subareolar	1.8 cm	1 month	Bloody nipple discharge	Present (bloody)	Absent	Present (rupture, abscess)	Irregular, indistinct, heterogeneous, posterior enhancement

M – Male; F – Female.

result from a squamous metaplastic transformation of normal columnar cells of the breast within an ectatic duct in fibrocystic disease or fibroadenoma [2,25,26]. Among the 15 previously reported cases of epidermal inclusion cyst in sub- or peri-areolar regions, 4 cases were associated with trauma [12,19]; 1 case with reduction mammoplasty [12], 1 case with a traffic accident [17], and 2 cases with needle biopsy [19]. Furthermore, 6 cases were presumed to have resulted from obstruction of a hair follicle [16]. However, possible mechanisms were not proposed in the remaining 5 cases [1,3,5,18].

Sonographic presentation of epidermal inclusion cysts of breast is generally that of a circumscribed mass with complex cystic and solid or heterogeneous internal echogenicity, as well as posterior enhancement [2,21]. A characteristic “onion-ring” sign, along with alternating concentric hyperechoic and hypoechoic rings corresponding to lamellated keratin have been reported in some epidermal inclusion cysts [4]. Instead of an “onion-ring” sign, a “tram-track” appearance consisting of multiple parallel alternating echogenic and hypoechoic lines was also reported in 1 case [15]. Among the previous 15 cases of epidermal inclusion cysts in the sub- or peri-areolar regions, sonographic features were described in 6 cases [1,3,5,12,17]. Similar to the current case, 3 cases demonstrated suspicious sonographic features [3,17] that were indistinguishable from breast malignancies, whereas the remaining 3 cases demonstrated usual sonographic features favoring benign lesion [1,5,12].

Mammographic presentation of epithelial inclusion cysts of the breast usually includes a circumscribed iso- or hyperdense mass [2,21] and can sometimes be accompanied by microcalcifications [21]. Magnetic resonance imaging features were reported in a few cases, which were circumscribed masses with a fluid-like signal of a variable low-signal component on

T2-weighted images and peripheral rim enhancement [2,5,6,9] or no enhancement [22] on contrast-enhanced images.

Epidermal inclusion cysts of the breast are associated with several complications such as spontaneous rupture, inflammation, and abscess formation [1]. In the case of rupture, the released keratin from the cyst might act as an irritant resulting in a foreign body reaction, granulomatous reaction, and abscess formation [5]. Furthermore, the transformation of epidermal inclusion cyst of the breast to squamous cell carcinoma has been reported; the reported incidence greatly varies from 0.045% to 19% [5]. Through a literature search, Paliotta et al. determined that the overall rate was 12% [2]. The rate of malignant transformation of epidermal inclusion cysts in the breast appears to be more frequent compared to that in other body parts, possibly due to the pathogenesis of squamous metaplasia affecting the mammary duct epithelium [2]. A significant association has been observed between tumor size and malignancy transformation [2]. Specifically, when larger than 5 cm in diameter, epidermal inclusion cysts are classified as giant, which are rare and more likely to develop complications such as malignant transformation [7]. Paget’s disease involving a subareolar epidermal inclusion cyst also has been reported in 1 case [18].

Because of the potential for malignant transformation, surgical excision is recommended, especially when the cyst is large (≥ 2 cm) and palpable, or when it causes the patient discomfort [2]. However, asymptomatic small sized lesions might not require treatment [5].

Conclusions

This report describes a ruptured epidermal inclusion cyst that was observed in an unusual location (subareolar area)

and had an unusual clinical presentation (bloody nipple discharge), which was radiologically indistinguishable from malignancy. Awareness of this unusual manifestation of epidermal inclusion cyst can be useful when assessing subareolar breast pathology.

References:

1. Kwak JY, Park HL, Kim JY et al: Imaging findings in a case of epidermal inclusion cyst arising within the breast parenchyma. *J Clin Ultrasound*, 2004; 32: 141–43
2. Paliotta A, Sapienza P, D'Ermo G et al: Epidermal inclusion cyst of the breast: A literature review. *Oncol Lett*, 2016; 11: 657–60
3. Whang IY, Lee J, Kim JS et al: Ruptured epidermal inclusion cysts in the subareolar area: sonographic findings in two cases. *Korean J Radiol*, 2007; 8: 356–59
4. Crystal P, Shaco-Levy R: Concentric rings within a breast mass on sonography: Lamellated keratin in an epidermal inclusion cyst. *Am J Roentgenol*, 2005; 184: S47–48
5. Lee YA, Park SG: Giant sized epidermal inclusion cyst of the breast initially mimicking a large fibroadenoma or phyllodes tumor. *J Korean Surg Soc*, 2012; 83: 107–10
6. Wynne E, Louie A: Epidermoid cyst of the breast: Mammography, ultrasound, MRI. *Radiol Case Rep*, 2015; 6: 431
7. Mahmud MU, Sheuly SB, Bhuiyan NH et al: Giant epidermoid cyst in the breast: A common benign lesion at a rare site – A case report. *Int J Surg Case Rep*, 2017; 36: 130–32
8. Menville JG: Simple dermoid cysts of the breast. *Ann Surg*, 1936; 103: 49–56
9. Kucuk AI, Kocer B, Turan G et al: A benign rare lesion of the breast: Giant epidermal inclusion cyst. *Cureus*, 2018; 10: e2650
10. Ben Naftali Y, Shoufani A, Krausz J, Hershko D: Unusual presentation of epidermoid cyst mimicking breast cancer involving the areola – case report. *Int J Surg Case Rep*, 2018; 51: 17–20
11. Amrani A, Sahu P, Dayal S, Verma P: Congenital epidermal inclusion cyst on the breast: A case series of a rare entity. *Int J Appl Basic Med Res*, 2018; 8: 54–56
12. Martin C, Bombard T, Payne E et al: Ruptured epidermal inclusion cyst of the breast. *J Women's Health*, 2014; 3: 1–2
13. Marchesi A, Parodi PC, Palitta G et al: A “polypoid” nipple: An unusual case of epidermoid cyst. *J Plast Reconstr Aesthet Surg*, 2014; 67(1): e27–28
14. Dilek N, Dilek AR, Saral Y, Sehitoglu I: Epidermoid cyst on the nipple: A rare location. *Breast J*, 2014; 20: 203–4
15. Rahul K, Panda A, Handa N, Hari S: Epidermal inclusion cyst in a male breast: Parallel linear echoes (tram-track appearance) on sonography as a diagnostic clue. *BMJ Case Rep*, 2015; 2015: pii: bcr2015213045
16. Singh M, Maheshwari B, Khurana N, Jain S: Epidermal inclusion cyst in breast: Is it so rare? *J Cytol*, 2012; 29: 169–72
17. Debnath D, Taribagil S, Al-Janabi KJ, Inwang R: A large epidermoid cyst of breast mimicking carcinoma: A case report and review of literature. *Int J Surg Case Rep*, 2012; 3: 437–40
18. Stephenson TJ, Cotton DW: Paget's disease in an epidermal cyst. *Dermatologica*, 1987; 174: 186–90
19. Gerlock AJ: Epidermal inclusion cyst of the breast associated with needle aspiration biopsy. *Radiology*, 1974; 112: 69–70
20. Jain S, Sarkar R, Garg VK, Khurana N: Epidermal inclusion cyst or giant milium of the nipple. *Indian J Dermatol Venereol Leprol*, 2012; 78: 103–5
21. Denison CM, Ward VL, Lester SC et al: Epidermal inclusion cysts of the breast: Three lesions with calcifications. *Radiology*, 1997; 204: 493–96
22. Taira N, Aogi K, Ohsumi S et al: Epidermal inclusion cyst of the breast. *Breast Cancer*, 2007; 14: 434–37
23. Fajardo LL, Bessen SC: Epidermal inclusion cyst after reduction mammoplasty. *Radiology*, 1993; 186: 103–6
24. Davies JD, Nonni A, D'Costa HF: Mammary epidermoid inclusion cysts after wide-core needle biopsies. *Histopathology*, 1997; 31: 549–51
25. Chantra PK, Tang JT, Stanley TM, Bassett LW: Circumscribed fibrocystic mastopathy with formation of an epidermal cyst. *Am J Roentgenol*, 1994; 163: 831–32
26. Willis RA: Squamous-cell mammary carcinoma of predominantly fibrosarcoma-like structure. *J Pathol Bacteriol*, 1958; 76: 511–15

Conflict of interest

None.