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SHORT COMMUNICATION

Isolated primary hydatid disease of the breast masquerading a breast tumor: report of a case and review of the literature

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Abstract Hydatid disease commonly affects liver and lungs. The rare breast involvement is usually a part of generalized hydatid disease. Primary breast hydatidosis is exceptionally rare even in endemic area. A case of isolated primary hydatid disease of the breast is reported in an elderly Indian women presenting with a painless breast lump. On clinical and imaging evaluation, the lump masqueraded a breast tumor. Fine needle aspiration cytology and core needle biopsy of the mass were inconclusive for the tumor. Complete excision of the mass was performed, which upon histopathological examination confirmed the diagnosis of hydatid cyst. We reviewed the relevant literature with a brief discussion on management challenges. Hydatid cyst should always constitute a differential while evaluating a breast lump especially in endemic area.

Keywords Cystic echinococcosis · Hydatid disease · Primary hydatidosis · Unusual location

Introduction

Hydatidosis is a zoonotic disease, caused by the larvae stage of tapeworm *Echinococcus granulosus*. Dogs are definitive host. Infection is transmitted when *Echinococcus* eggs, shed through the stool of definitive host, are ingested by humans or other animals like sheep and goat. Human beings are paratenic host. Hydatid disease is endemic in certain part of world including Indian subcontinent and is a significant public health concern. *Echinococcos granulosus* larvae can produce fluid filled cysts in almost every organ of the body however, liver (75%) and lungs (15%) are the most common organs affected. Involvement of breast is rare and usually a part of generalized hydatidosis. Primary breast hydatid is exceptionally rare even in endemic area with reported incidence only 0.27% (Vega et al. 1994). The usual presentation of the breast hydatidosis is painless palpable lump that poses a challenge to differentiate it from other common breast lump of benign or malignant nature. The current report described one such case masqueraded breast tumor, with a brief discussion on management challenges.

Case report

A 55-year-old otherwise healthy Indian women presented to our out-patient department with a 4-year history of lump in the right breast. She reported the lump was gradually increasing in size with associated heaviness but denied pain, nipple discharge, fever, recent weight loss or swelling elsewhere in body. She also denied history of trauma, tuberculosis, or family history of breast cancer. Patient was a strict vegetarian from lower socio-economic class. On physical examination, a firm, non-tender, oval shaped lump, measuring 5×4 cm, free from the skin and underlying tissue, slightly mobile but fixed to the breast tissue, was palpable in the upper inner quadrant of the right breast. There was no axillary or supraclavicular adenopathy and contralateral breast was normal. Rest of her physical examination was unremarkable.

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Evaluation with X-ray mammography showed a partially marginated oval shaped radio-opaque lesion in upper-inner quadrant along chest wall with margins obscured by pectoralis muscle shadow. No obvious internal microcalcification or surrounding architectural distortion seen. Sono-mammography demonstrated the lesion predominantly being cystic with few thick septae within, measuring 4.7 × 3.1 cm. No obvious solid component or internal vascularity noted on doppler study. The lesion was consistent with BIRADS IV-A (Fig. 1). Contralateral breast and axilla was normal. Chest radiograph and abdominal USG was unremarkable. Her laboratory investigations were reported normal. Based on clinical and imaging evaluation, a diagnosis of cystic neoplasm of breast was made. Fine needle aspiration yielded clear transparent, thin watery, paucicellular fluid showing few macrophages and epithelial cells, scattered turbid material with RBCs, few clusters of apocrine cells and was reported inconclusive. Repeat aspiration showed mixed inflammatory cell infiltrates including scattered eosinophils. Core needle biopsy from cyst wall showed similar finding and inconclusive for malignancy. The patient underwent complete excision of the mass taking wide margins without spillage of their contents. On sectioning the excised mass postoperatively, we suspected it a hydatid cyst. Wound washing was done with scolicidal agent, hypertonic saline thereafter. Histopathological evaluation revealed a cystic mass measuring 5.0 × 4.0 × 3.0 cm. Cut section yielded creamy white to brown fluidy material. Cut surface had pearly white cystic membrane with adjacent greyish brown to haemorrhagic area. On microscopy, cystic lesion displayed outermost skeletal muscle, dense mixed inflammatory cell infiltrate comprising of lymphocytes, plasma cells and frequent eosinophills. The deeper portion of parenchyma was seen in continuation with inflammation with giant cell reaction, pallisading granulomas. Lamellated membrane of parasite was noted. Endocyst portion was showing scolices of parasites with hooklets. These histological features were consistent with hydatid disease (Fig. 2). Tablet albendazole was advised for four weeks. Post operative recovery was uneventful. Follow up 6 months, the patient remains asymptomatic and showed no evidence of recurrence.

Discussion

We have presented, herein, a case of intramammary hydatid disease. Although hydatid disease is not uncommon in Indian subcontinent, it presenting as breast lump is exceptional. Our case had isolated hydatid cyst in the breast hence, the primary site. These patients remain asymptomatic for years as slow growing lump present late, secondary to large size of the cyst and pressure effect. A

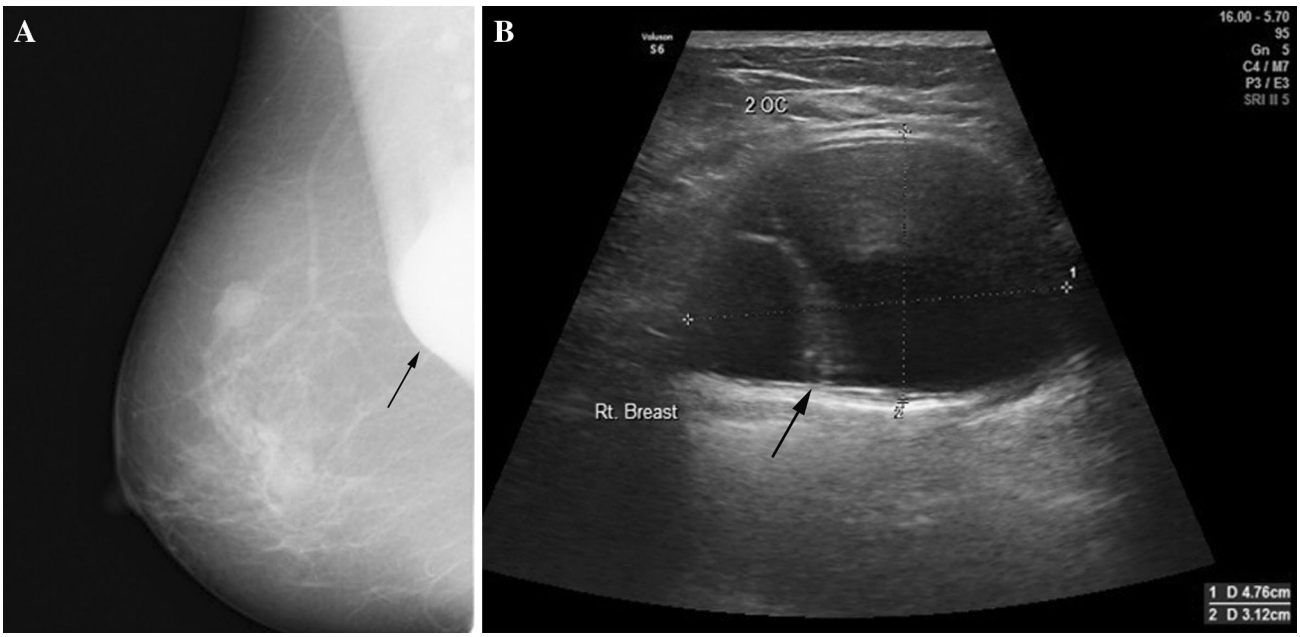

Fig. 1 **a**, **b** (mammography panel): X-ray mammography of the right breast showing a partially marginated oval shaped radioopaque lesion in upper-inner quadrant along the chest wall with margins are obscured by pectoralis muscle shadow. No obvious internal microcalcification or surrounding architectural distortion seen. There also showed a well marginated, oval shaped, radio-opaque lesion consistent with fibroadenoma. Sono-mammography showing a well defined oval shaped predominantly cystic lesion with few thick septations within. No obvious solid component or internal vascularity seen. Findings consistent with cystic neoplasm BIRADS IV-A

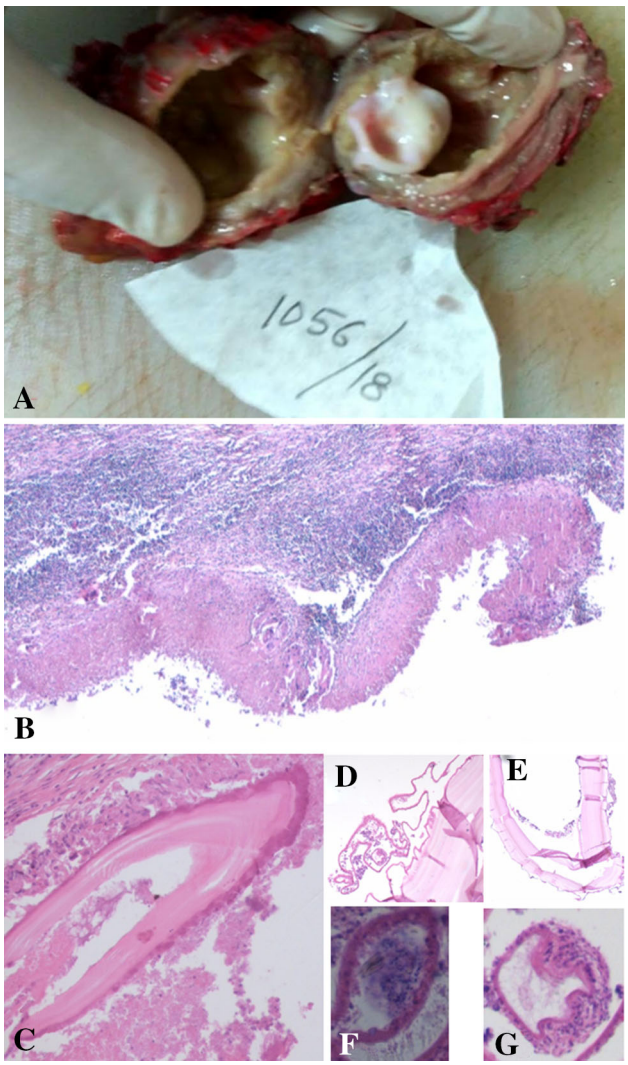

Fig. 2 **a**–**g** (HPE panel): Cystic breast lump displaying whitish cyst membrane (**a**); Pallisading necrosis and dense lympho-mononuclear infiltrate seen in the wall (**h** and **e** stain × 10) (**b**); Laminated membrane and daughter cysts (**h** and **e** stain × 10) (**c**–**e**), Scolices and hooklets of parasite (**h** and **e** stain × 20) (**f** and **g**)

breast lump is evaluated preoperatively through triple assessment, utilising clinical evaluation, imaging studies and cytological examination (Russel et al. 2004). Owing to the rare site of the disease and no disease specific sign on assessment, the diagnosis of breast hydatid poses a challenge, as lump often mistaken for other more common breast pathologies like benign cyst, chronic abscess, fibroadenoma, phylloids tumour or even carcinoma especially in elderly women (Yaghan 1999). Our assessment, in present case, had mistaken the lump as cystic neoplasm of breast. Fragment of paracytic cyst wall, scolices or hooklet were absent in cytology material examined on both occasions The diagnosis of hydatid disease of breast was established postoperatively on histopathological examination of excised mass as it revealed characteristic laminated membrane, scolices and hooklets of parasite.

On X-ray mammography, hydatid cyst appear non-specific homogenous, smooth, well defined lesion (Gharbi et al. 1981). In overpenetrated view characteristic ring shaped structure may be seen due to differential density between cyst wall and daughter cysts (Vega et al. 1994). Sonographic features of hydatid cyst have been described under five categories that can range from simple cyst to completely solid appearance based on cyst evolution and complications (Gharbi et al. 1981). Present case had predominantly cystic lesion with thick septae within, consistent with type-3 cyst. A ''congealed water lily sign'' which is considered pathognomonic of hydatid disease seen as solid thing arises due to trapping of germinal membrane between echogenic fluid (Tutar et al. 2006). In view of liver and lung being the most common sites of hydatid cyst, abdomen sonography and chest radiograph have an important role in excluding generalized hydatidosis. Breast hydatid has been reported through characteristic findings on MRI. These findings on imaging study however, often missed if hydatid disease is not kept as a differential (Sinha et al. 2008). FNAC showing clear watery fluid on aspiration should raise suspicion of hydatid fluid but reports are often inconclusive due to inadequate material not showing characteristic features (Afroz et al. 2014). FNAC diagnosis of musculoskeletal hydatid disease is described by the author previously. It is important to highlight that cytology is limited material therefore diagnosis of hydatid disease may be missed during aspiration and histological evaluation is important for confirmation (Hui et al. 2015). Although, in our case, FNAC had suggested benign nature of cyst, we went ahead with a wide excision of the lump. This was done to clear the fear of uncertainty for lesion being malignant and to obtain a confirmatory diagnosis. Careful, complete, cysto-pericystectomy without spillage of contents is the curative treatment. When hydatid disease is a differential, FNAC or biopsy however, are not indicated as to prevent the risk of spillage of the contents, causing anaphylactic reaction and dissemination (Jakubowski and Barnard 1971). In such cases, a course of antihelminthic agents like albendazole and praziquantel are recommended preoperatively. Intraoperatively, hypertonic saline can be useful to kill daughter cysts and to prevent anaphylactic reaction and further spread. Delay in diagnosis or mistaken diagnosis may lead to inadequate treatment or complication at surgery or future recurrence. Raised eosinophilic counts can be of helpful in making preoperatively diagnosis of parasitic infestation, however, the eosinophilic counts are not increased in more than 90% of reported cases. Serological tests like indirect haemagglutination, enzyme-linked immunosorbent assay (ELISA), are widely used method to detect antibodies against

Echinococcus and follow up but sensitivity and specificity is far less compared to the other common sites (Jakubowski and Barnard 1971; Brunetti et al. 2010)

In conclusion, primary hydatid disease of the breast is exceptionally rare. Preoperative diagnosis is challenging and pose a serious concern as lump may mimic breast tumour, thus, highlighting the need for greater awareness of the entity. Hydatid cyst should always constitute a differential while evaluating a breast lump especially in endemic area.

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Compliance with ethical standards

Conflict of interest All authors declare that they have no conflict of interest.

Informed consent Informed and written consent to the contents of the manuscript to be published for medical, scientific and educational purposes without revealing the identity has been obtained from the patient.

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