An Isolated Hydatidosis of Breast: A Rare Case Report with Review of Literature

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ABSTRACT
Hydatid cyst disease caused by parasite Echinococcus granulosus is endemic in sheep raising communities and man is an occasional intermediate host. Hydatid cysts are usually located in the liver and the lungs; they are very rarely seen in the breasts. We present an uncommon case of an isolated hydatid cyst of the breast in a 28-year female in whom, preoperative diagnosis was made only after the fine-needle aspiration cytology (FNAC). The cytosapirate was one ml of clear straw colored fluid which after centrifuging revealed refractile hooklets on a background of cellular debris and proteinaceous material. This case emphasizes that preoperative diagnosis of hydatid cyst in breast by FNAC is a difficult one, and is often missed out as echinococcal infection in breast is uncommon, and the breast lump mimic many tumoral conditions of breast.

INTRODUCTION
Breast parasitosis is very rare and is usually caused by larval form of filarial worm and Taenia solium. Hydatid cyst disease of the breast is very uncommon, caused by Echinococcus granulosus and accounting for 0.27% of all cases of hydatidosis even in endemic areas.1 It is a zoonosis and humans are intermediate hosts for the larval form of the parasite.2 Breast hydatidosis most frequently is caused by systemic dissemination and very rarely a primary site of infection. Patients usually present to the hospital with a palpable and painless lump in the breast that gradually increases in size. It is frequently seen in the age group of 30 to 50 years.3 Only few reports of breast hydatidosis are published and majority of the reported cases have been diagnosed postoperatively after histopathological study. Here we present a case of uncommon preoperative diagnosis following fine needle aspiration cytology (FNAC) of isolated hydatid cyst of breast in a 28 year old female.

CASE HISTORY
The patient, a 28 year old house wife presented with a painless lump in the left breast of one year duration. There was no history of injury, discharge from the nipple or family history of breast cancer. Examination revealed a mobile, firm lump measuring 4 cm × 3 cm in the subareolar region of the left breast predominantly in the upper outer quadrant. The left breast and nipple were normal and there was no axillary or cervical lymphadenopathy. Routine investigations were normal except 9% eosinophils in blood differential count. The chest x-ray and abdominal ultrasonography were normal. FNAC was done from breast mass and one ml of clear pale yellow fluid was retrieved. Centrifuged cytosmear showed good number of refractile hooklets in a granular debris and fluid background. A diagnosis of hydatid cyst of breast was made (Fig.1a & b) and the patient was treated with mebendazole. After two-week course, lumpectomy was done from the left breast and the gross specimen was sent for histopathological study which consisted of fibrofatty tissue measuring 3x3 cms. Cut surface showed a unilocular soft thin walled fluid filled cystic structure, whitish in colour and with a semitranslucent, shiny inner surface (Fig.2). Histopathology revealed the cyst wall made of compressed breast tissue, infiltrated intensely by lymphocytes and eosinophils and giant cell reaction with an eosinophilic acellular laminated cyst wall adjacent to it, which confirmed the diagnosis as a case of hydatid cyst of breast (Fig.3). The patient was discharged on the 2 postoperative day without any complication. Mebendazole treatment was continued for three months and now the patient is doing well without any recurrence.

DISCUSSION
Echinococcus granulosus is the most common cause of hydatid cyst which is a tape worm from cestodian class. Humans are the
accidental intermediate hosts (dead end) and animals (herbivores and omnivores) are both intermediate and definitive hosts. The adult E. granulosus is a worm that inhabits small intestine of definitive host and produces eggs containing embryos (oncospheres) expelled in faeces which is ingested by intermediate hosts like cows, sheep, and humans. It enters into their blood system through the intestinal mucosa and migrates to liver and other viscera, where it grows to a multilayer cyst (metacestode) and subsequently development of protoscolices within it. When a definitive host ingests the intermediate host organs the protoscolices grow into adults in the intestinal mucosa and the cycle repeats. According to Barret and Thomas, 60% of the cysts are found in the liver, 30% in lungs, 2.5% in kidneys, 2.5% in heart and pericardium, 2% in bone, 1.5% in spleen, 1% in muscle, and 0.5% in brain.

Fig.1: Gross specimen of hydatid cyst of breast showing a fluid filled whitish globular soft thin walled cystic structure.

Fig 2a & b: Refractile hooklets in a background of granular debris and proteinaceous material. H&E stain and Pap stain x 400

Fig.3: Compressed breast tissue infiltrated by lymphocytes, eosinophils and giant cells with an eosinophilic acellular laminated cyst wall adjacent to it. H&E stain x 400

Hydatid disease of breast is rare and accounts for only 0.27% of all cases. The breast can be a primary site or part of a disseminated hydatidosis. Hydatid cyst consists of 3 layers like outermost adventitia of fibrous tissue (pseudocyst), middle layer of laminated membrane (ectocyst) and innermost layer of germinal epithelium (endocyst) which secretes hydatid fluid internally and laminated layer externally. Typically, the patient presents with painless breast lump, which increases slowly in size without regional lymph node involvement. It generally affects women between 30 and 50 years of age. It might mimic fibroadenoma, phyllodes tumors, chronic abscesses, or even carcinoma. So breast hydatid cyst should be included in differential diagnosis of breast lumps especially in endemic areas. Preoperative diagnosis can be made by fine needle aspiration cytology where scolices, hooklets or laminated membrane can be identified. It is a safe procedure, as no complications were mentioned in the literature. The sonographic appearance of mammary hydatid cysts may be similar to those observed in benign cysts, showing a well-defined, lobulated mass of heterogeneous echogenicity that may contain multiple cystic areas. Hemagglutination tests may be helpful in diagnosis. Due to the rarity of this condition, mammographic and sonographic appearances of breast hydatid disease are frequently missed until an operative diagnosis has been made. Rarely, a preoperative diagnosis can be made using a combination of clinical, imaging and FNAC findings. The treatment of a hydatid cyst of the breast is complete excision. However, recurrent cysts have been reported postoperatively in 10% of patients. It has been reported that pre and post-operative treatment with mebendazole reduces the risk of recurrence.
CONCLUSION
Hydatid cyst of breast is rare and it often goes unnoticed by mammography and ultrasonography. It can be challenging to differentiate it from other tumoral lesions of the breast like fibroadenoma, phylloides tumour and abscess. Thus preoperative diagnosis may be performed using FNAC which minimizes the risk of intraoperative rupture. Cytological diagnosis by FNAC is difficult. Here in this case FNAC revealed presence of hooklets and histopathological examination confirmed it to be a case of hydatid cyst.

REFERENCES

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