Hydatid Cyst of Breast: Unwelcome Guest at Unusual Site; A Case Report and Review of Literature

Nisha Meshram[1], Fatema Kamal[2], Rasika Gadkari[3]

ABSTRACT

Introduction: Hydatid cyst is a parasitic disease caused by tapeworm echinococcus granulosus. Echinococcosis is endemic in sheep and cattle raising communities. Human is accidental intermediate host. Hydatid cysts are commonly seen in lung and liver. Breast involvement is very rare.

Case report: We report a case of primary isolated hydatid cyst of the breast in a 21-year-old female, diagnosed on fine needle aspiration cytology along with a review of the literature. The FNAC smears from breast aspirate showed scolices of echinococcus granulosus in various stages of development, along with refractile hooklets in a proteinaceous background. Radiological examination done subsequently endorsed the cytodiagnosis of Hydatid cyst and did not show involvement of any other organ or site. Histopathological examination of lumpectomy specimen confirmed the diagnosis of Hydatid cyst. This case is unique as all stages of lifecycle of echinococcus granulosus could be seen on FNAC.

Conclusion: We like to emphasize that infective etiology like Hydatid disease should be considered as one of the differential diagnosis in case of breast lump with fluid aspirate. FNAC though feared with possibility of anaphylactic reaction, is safe procedure and provides diagnosis in clinically unsuspected cases.

Keywords: Hydatid Cyst, Echinococcusgranulosus, Scolices, Fine Needle Aspiration Cytology

INTRODUCTION

Hydatidosis is an ancient disease known since the time of Hippocrates and most commonly seen in sheep & cattle raising countries.[1] It is a parasitic disease caused by larval cystode (tapeworm) Echinococcusgranulosus. There are two forms of the disease echinococcusunilocularis & multilocularis. The definitive host is dog, fox etc while intermediate host are usually ruminant animals like sheep, cattles. Humans are accidental intermediate hosts, infected by ingestion of food contaminated with eggs shed by the dogs.[2,3] Hydatid disease (HD) is endemic in many parts of the world such as the Middle East Africa, South America, Central Asia, New Zealand, Australia, Iran,Iraq Turkey, and Southern Europe. In India the highest prevalence is reported in Andhra Pradesh, Tamil Nadu, and Jammu and Kashmir.[4–6] Common location of hydatid cysts are liver(75%) and lungs(15%) while unusual locations like kidney, pancreas, bladder, spleen, ovary, brain, heart, thyroid, bone and muscle includes the remaining 10% cases.[7–9] Breast is a very rare site for Hydatid disease, accounting for 0.27% of cases.[10,11] Patients of hydatid breast disease usually present with a painless palpable breast lump. Very few case reports of confirmed preoperative diagnosis of hydatid disease of breast are available. Herein, we report a cytologically diagnosed, case of primary isolated breast hydatid cyst showing almost all the stages of their lifecyle along with review of literature.

CASE REPORT

A 21-year-old married, nulligravida female presented with painless lump in the right breast since one year. It was static in size, not associated with pain, nipple discharge or skin discoloration, also no association with menstrual cycle was present. She had no history of trauma to the breast, or family history of breast cancer. Breast examination revealed 5 cm × 5 cm lump in upper outer quadrant of right breast. The mass was firm in consistency, freely mobile & non-tender. Overlying skin and nipple areola complex appeared normal. There was no axillary lymphadenopathy and contralateral breast was normal. Systemic examination findings were unremarkable. Routine laboratory investigations including complete blood count and biochemistry were within normal limits. In view of clinical diagnosis of Fibroadenoma of breast, fine-needle aspiration cytology (FNAC) was advised. FNAC was performed under palpation by trained Cytopathologistusing a 23-guage needle and 15 ml of clear watery fluid was retrieved. Swelling partially decreased in size.There were no post-procedure complications like urticaria or hypersensitivity reactions. The fluid was centrifuged and smears were prepared from the sediment and stained by May-Grünwald Giemsa (MGG), haematoxylin & eosin (H&E) and Papanicolaou (PAP) stains. Few unstained smears were preserved for any special stain if needed. Microscopic examination of the centrifuged cytosmears showed numerous scolices with attached hooklets, [Figure 1], scolices with rostellum, protoscolices with calcareous corpuscles, flame cells [Figure 2] along with multiple scattered hookletsin a clean background.Small fragments of acellular material representing remnants of laminated membranes was also seen.These cytological findings suggested the diagnosis

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How to cite this article: Meshram N, Kamal F, Gadkari R. Hydatid cyst of breast: unwelcome guest at unusual site; a case report and review of literature. International Journal of Contemporary Medical Research 2021;8(1):A12-A16.

DOI: http://dx.doi.org/10.21276/ijcmr.2021.8.1.31

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of hydatid cyst. Following the diagnosis USG breast was performed which demonstrated well defined double walled cyst in upper outer quadrant of right breast, suggestive of Hydatid Cyst (Type I Gharbi'sclassification). Clinical and radiological examination performed subsequently for evidence of hydatid disease in other organs, did not show involvement of any other organ or site. The patient also did not give any past history of hydatid cyst in any other organ. So final diagnosis of primary isolated Hydatid cyst of breast was made. As typical features of Hydatid disease were present on cytology and later substantiated by radiology, serological tests were not done. Patient received adjuvant antihelminthic

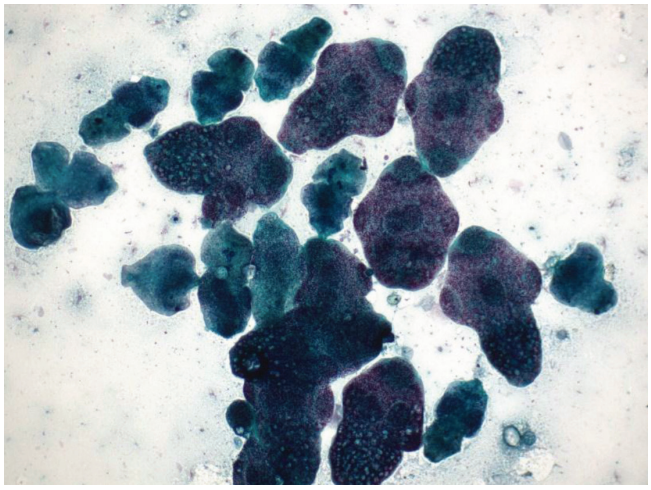

Figure-1: Multiple scolices with attached hooklets (Masson Trichrome stain, 10X)

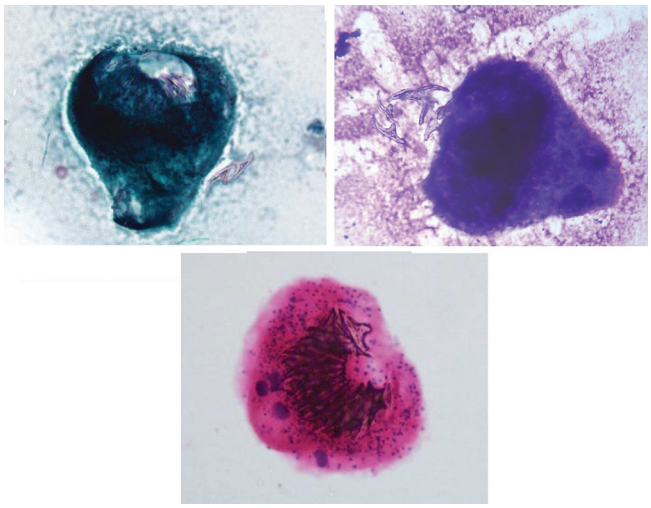

Figure-2: Scolex with rostellum (MGG, MT & HE stain, 40X)

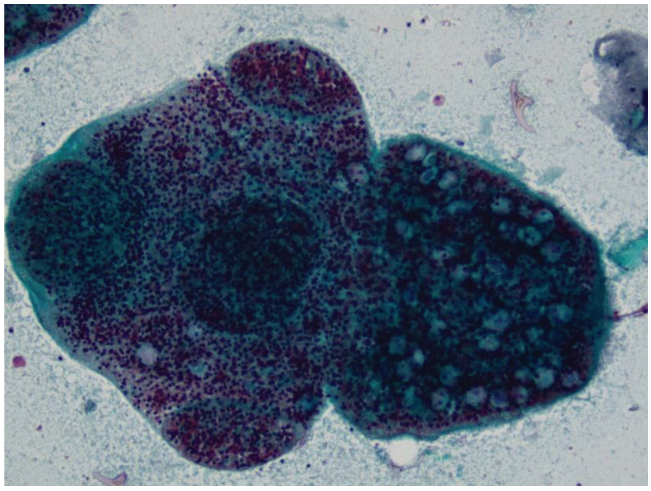

Figure-3: Protoscolex with calcereal bodies (MT stain, 40X)

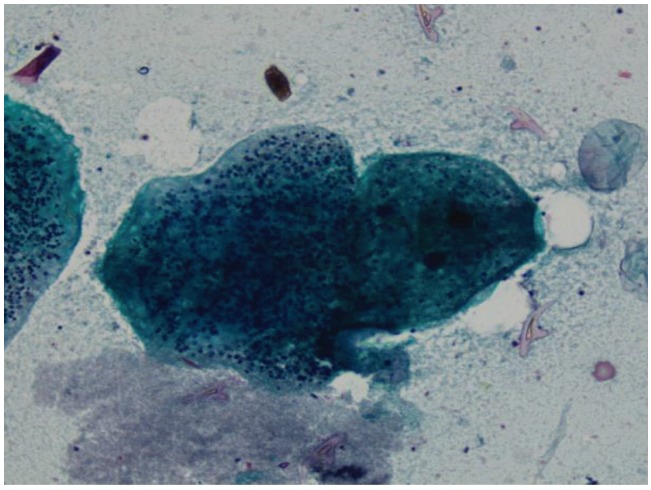

Figure-4: Flame cell (MT stain, 40X)

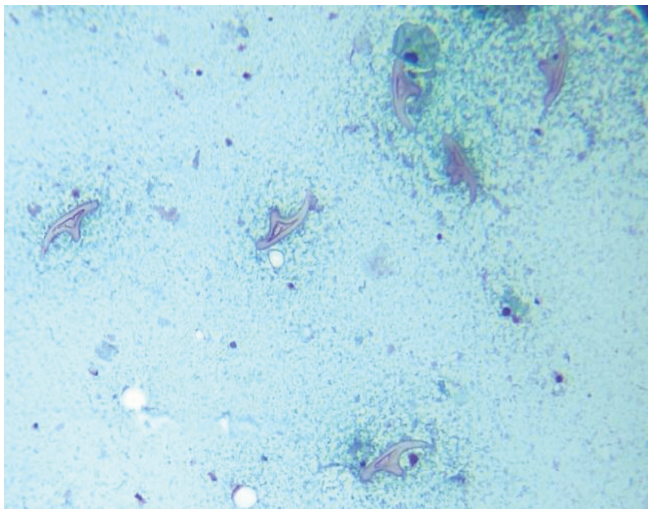

Figure-5: Multiple scattered hooklets (MT stain & MGG stain, 40X)

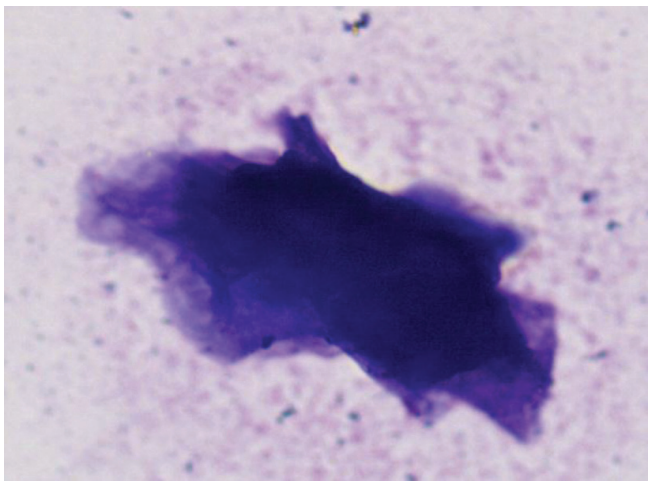

Figure-5: Multiple scattered hooklets (MT stain & MGG stain, 40X)

Organism Name	Membrane s	Scolices	Hooklets	Suckers
Echinoccoccus Granulosus	Thick &lamellated	Multiple, Small, oval	2 rows, 22-44um	Present, 4 in no
Cysticercous	Thin, membranous, Nucleated	Single, Large, 1mm	2 rows, large & small 130-170 um	Present, 4 in no
Coenuruscerebralis		Multiple	Present, 40-175um	
sparganosis	Absent	Absent	Absent	Absent

Table-1: Distinguishing features of echinococcus from other cestodes[18,16]

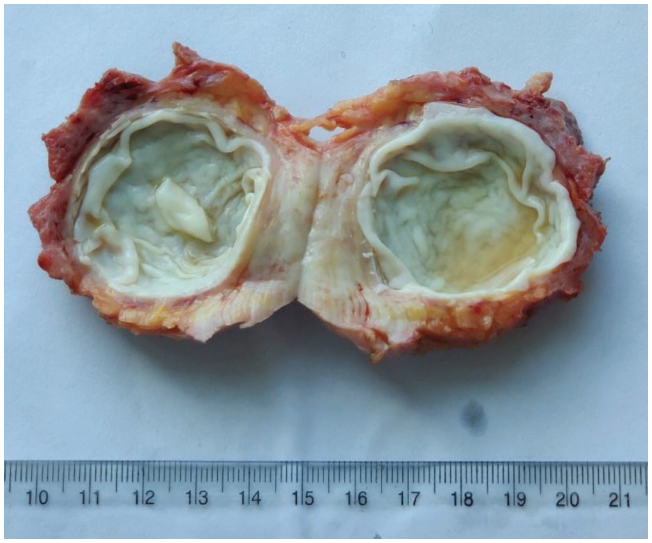

Figure-7: Gross Appearance of cyst (C/s, Tender coconut)

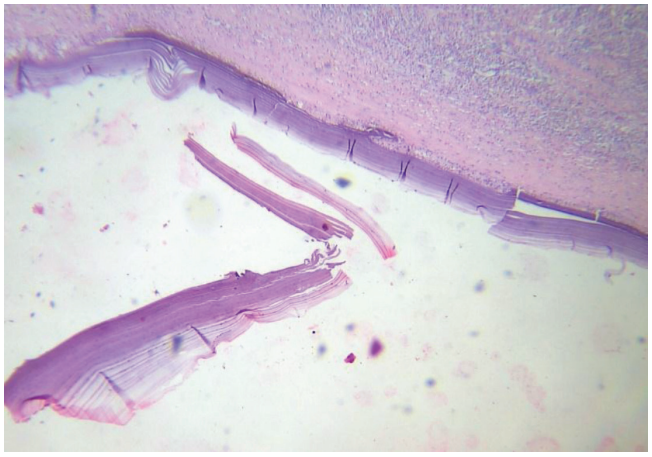

Figure-8:

treatment with albendazole for two weeks.

After two-week course, lumpectomy was done. Lumpectomy specimen was a well-circumscribed cystic mass with thick fibrous wall &measured 4 cm × 4 cm × 2 cm. Cut section showed a unilocular, thick walled (1.5cm) cystic structure, whitish in colour with a semi translucent, shiny inner surface (tender coconut) (Fig 7). Histopathology revealed acellular laminated cyst wall surrounded by dense inflammation comprising of eosinophils and lymphocytes along with thick fibrous pericyst surrounded by breast parenchyma(fig 8)

Histopathological findings confirmed the cytological diagnosis of hydatid cyst of breast. Postoperative recovery was uneventful. The patient was discharged on the 2nd postoperative day without any complication.

DISCUSSION

The adult E. granulosuslives attached to the mucosa of the small intestines of dogs and related animals. It is a small worm measuring 2 to 9 mm in length. It consists of three or four segments and has a globular scolex of 0.3 mm in diameter containing a rostellum and four cuplike oval suckers. The rostellum is armed with a double crown of large and small hooklets. The majority of protoscoleces have two rows of hooks with an equal number of large and small hooks. The final hosts of E. granulosusare dogs, wolves, jackals, dingoes, coyotes and foxes, while the common intermediate hosts are many domesticated mammals such as sheep, cattle, pigs, goats and camels. Sheepharbors the most fertile hydatid cysts, and is the most important intermediate host. Animal hosts differ from area to area.[1]

Echinococcus infection in humans occurs after accidental ingestion of tapeworm ova from dog faeces in contaminated food. The hexacanth larva hatches in the gastrointestinal tract, penetrates the intestinal wall with the help of the hooklets and enter the venous circulation of the mesentery and through portal vein reach the liver, where most of them are caught in the hepatic sinusoids. Few may pass through the liver (first filter) and reach the lung (second filter) and systemic circulation, causing hydatid disease in other organs and sites. Dissemination through lymphatic channels has also been suggested as a possible mechanism and accounting for cases with solitary cysts in uncommon sites. Another mechanism may be direct spread from adjacent sites.[3] HC may occur in almost any of the organs, however, in 80% cases the liver and lung bear the main burden.[12]

Hydatid disease of the breast is rare, and is seen in only 0.27% of the cases.The breast involvement can be primary or as part of disseminated disease. Despite

its rareness, primary breast involvement might constitute an important differential diagnosis of breast lumps in areas endemic for hydatid disease. The usual presentation is painless breast lump, with slow growth or increase in size. (imranamasroor)2 commonly women between 30–50 years of age are affected, but a wider age range (26–74 years) is also reported.[13]

In an unsuspected case chest X-ray and ultrasonography may aid in diagnosis. Differential diagnoses of fluid-filled cysts in breast includes, fibroadenoma, carcinoma, metastases, and inflammatory masses.However, CT can provide an accurate diagnosis especially in complicated hydatid cysts.26 Serology can be useful if morphological diagnosis is not available. Common serological tests used for diagnosis of hydatid disease are Casoni intradermal skin test, Weinberg complement fixation (CF) test, indirect hemagglutination

(IHA) test, ELISA, and western blot (WB). A positive serological test for hydatid cysts is significant, however, negative test does not indicate the absence of the disease. The serological tests are complementary to the clinical and radiologic findings and they can also be used in the follow-up of patients after surgical resection.[14]

Antigen detection assay show a sensitivity of 25.7% and a specificity of 98.0% whereas a sensitivity of 94.2% and specificity of 81.6% were found for the antibody detection assay by Sadjjadiet al.[1]

FNAC of the cyst yields mostly watery, clear aspirate but sometimes it can be turbid also. The smears may show scolices, hooklets or membranes. Commonly lamellatedmembrane &hooklets are seen. PAS stain best demonstrates the delicate, parallel laminations of the membrane.[12] Masson Trichrome stain is used for demonstrations of hooklets. The hooklets are about 20-40um in size, semi-translucent, refractile, triangular or sickle-shaped with an inner semi-translucent core and because of their refractile nature can be visualised by lowering the condenser of microscope.8 The scolices appear as long, oval structures about 100µm in diameter with four protruding suckers at one end, some show two rows of characteristic radially arranged hooklets. Scolices without hooklets are named as flame cells. Almost all the scolices show rounded calcareous bodies. Presence of scolices is rare finding, moreover identification of all stages of development of scolices is rarer making this case unique as almost all stages of development could be identified.

Other cystodes causing fluid filled cysts are Coenurus cerebralis (coenurosis), Diphyllobothriumlatum, Spirometra species (sparganosis) and Taeniasolium (cysticercosis).[15]

Cysticercosis caused by the pig tapeworm, *Cysticercuscellulosae* or Tenia Solium is a close differential. Both have a bladder wall, scolices with hooklets, and initiate an inflammatory response, but The bladder wall is thick, acellular and lamellated in hydatid cyst whereas it is thinner, membranous, and shows multiple nuclei in Cysticercus,. Hydatid cysts have multiple daughter cysts within a parent cyst and hence may yield many scolices in an aspirate. The individual scolices are small; the hooklets measure about 22 um and 40 um and can be seen only at higher magnification. In contrast, Cysticercus has only one large scolex, 1 mm diameter, and two rows of alternating large and small hooklets measuring approximately 170 mm and 130 mm, respectively. The scolex is visible to the unaided eye and is easily recognised at scanning magnification.[16] The presence of multiple scolices, membranous laminated structure, and hooklets can help to distinguish HC from cysticercosis.[7,17]

Hooklets and suckers are present in coenuri, but not in spargana. Though coenuri also has multiple protoscolicescytomorphological details of the aspirate help to differentiate these parasites [Table 1].

Preoperative diagnosis is important due to anaphylactic reaction consequent to cyst rupture during the treatment. Contamination of surrounding tissues with the cyst contents may cause local recurrence.[12] Hence complete Surgical excision of the intact hydatid cyst without rupture is the treatment of choice. Puncture of the cyst followed by aspiration of the contents, cyst pouch irrigation with scolicidal solutions, and reaspiration can also be done. Preoperative chemotherapy with albendazoleor Mebendazolemay reduce the recurrence of disease. Also it can be used in selected cases where the surgical excision is not possible or may have risk of spillage of the contents of the cyst.[17] The recurrence rate due to incomplete removal or unidentified cyst is around 2% and 25%.

CONCLUSION

Aspiration of clear fluid with and varying proportions of inflammatory cells consisting of eosinophils, neutrophils, plasma cells, and giant cells in a palpable breast nodule should arouse suspicion of parasitic origin even in the absence of identifiable parasitic fragments.[19]

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Source of Support: Nil; **Conflict of Interest:** None

Submitted: 27-12-2020; **Accepted:** 29-01-2021; **Published:** 31-01-2021