CASE REPORT

Pre-operative diagnosis of hydatid cyst in the breast: A case report of a rare entity and review of literature

Sidrah Mahmood[1], Rabiaa Mahmood[2]

Abstract

Hydatid cysts are mainly found in the liver and lungs. Only few cases have been reported in the breast and majority of these cases have been diagnosed post-operatively. We report a case of hydatid cyst of the breast diagnosed pre-operatively by fine needle aspiration and cytology (FNAC), without any complication. A 50 years old female presented with a slowly growing mobile lump in the left breast for 3 years. Mammography showed a circumscribed opacity in left breast. High resolution ultrasound displayed a unilocular cyst with some echogenic specks settling to the gravity dependent portion. An aspiration was planned and, as the cyst volume decreased, membranes appeared within the residual fluid and started settling down. Cytology of aspirated fluid revealed a hydatid cyst. Hydatid cysts in the breast are rare but possible, particularly in endemic regions. Clinical and radiological aspects of hydatid disease along with review of literature are presented in this paper.

Keywords: Hydatid cyst, Mammography, High resolution Ultrasound, unilocular cyst, Fine needle aspiration cytology.

DOI: https://doi.org/10.47391/JPMA.7085

Submission completion date: 10-08-2022

Acceptance date: 05-01-2023

Introduction

Hydatid cysts are caused by parasitic infection and can occur in any part of body, with the more common locations being in the lungs and liver. Involvement of breast in hydatid cyst is rare, with its incidence being 0.27%.[1] Gharbi[2] and WHO[3] have described several ultrasound stages in the pathophysiology of hydatid cysts- many features among them are nonspecific on ultrasound but a few features are typical of hydatid disease. These include floating internal membranes (Gharbi stage II, WHO CE3A), presence of recognizable matrix with embedded membranes (Gharbi stage IV, WHO CE4)[4] and sometimes the presence of daughter cysts giving a soap bubble or spoke-wheel appearance.

[1]Department of Breast Imaging, Multan Institute of Nuclear Medicine and Radiotherapy Cancer Hospital, Multan, Pakistan; [2]Department of Radiology, Mukhtar A Sheikh Hospital, Multan, Pakistan.

Correspondence: Sidrah Mahmood. e-mail: sidrahibm@hotmail.com
ORCID ID. 0000-0001-5909-2702

Hydatid cyst in breast has been reported to affect women between 30 and 50 years of age, who usually present with a clinically non malignant mass. It is very rare for hydatid disease to present with the breast as a primary site because the breast is more commonly involved as part of systemic dissemination. Definite diagnosis is difficult to make pre-operatively as hydatid cyst in breast can be confused clinically with other breast lesions and due to its rare occurrence, it is usually not included in the differential diagnosis of breast lumps. Mammography and ultrasonography (USG) features are also non-specific and it may be confused with simple or complicated breast cysts, unless the typical features of a hydatid cyst are present Only 131 cases of hydatid cyst in the breast have been published in literature[5] and in the majority of cases, diagnosis was only possible post-operatively.[6] The diagnosis of breast hydatid cyst by fine needle aspiration has been reported with no urticarial or anaphylactic reactions, which are documented complications of this procedure. Therefore, FNAC can act as an economical, minimally invasive and safe procedure for pre-operative diagnosis of hydatid cyst.[7] We report a case of hydatid cyst of the breast diagnosed preoperatively with no complication along with review of the literature.

Case Report

A 50 years old female visited breast imaging department of MINAR cancer hospital Multan in May 2020. She complained of a slowly growing, painless lump in the left breast for 3 years with no fever or axillary lymphadenopathy. On examination, there was a 5cmx4cm, firm, mobile, non-tender lump in the left breast in the upper and outer quadrant. Her mammogram showed a well circumscribed opacity in the upper and outer quadrant of left breast (Figure-1). High resolution breast ultrasound showed a 5cmx2.5cm unilocular cyst in the left breast at 2 o'clock position. The cyst had a moderate amount of echogenic sludge-like material that settled to the dependent location, on moving the patient. Ultrasound guided aspiration yielded watery fluid and caused membranes to appear in the residual cyst fluid (Figure-2). Cytology of the fluid showed retractile hooklets, granular debris and multinucleated giant cells. These findings were consistent with the diagnosis of hydatid cyst. Abdominal ultrasound was done subsequently, which

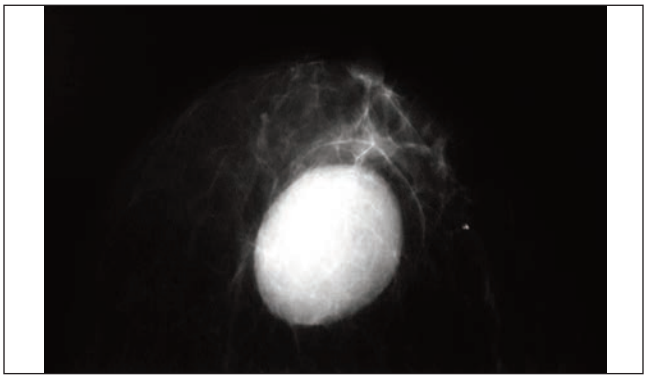

Figure-1: Mammography showed a radio-opaque, well circumscribed, opacity in the left breast.

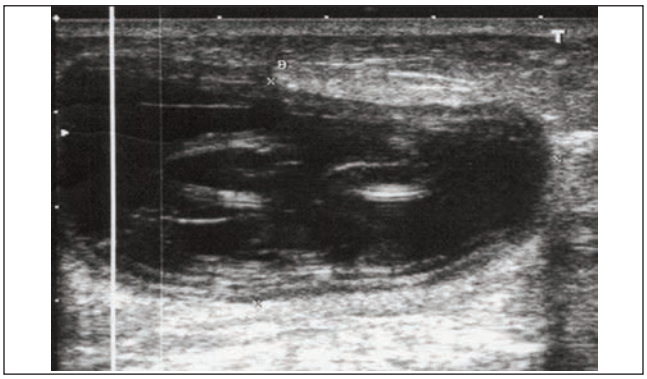

Figure-2: USG Breast: Aspiration of watery fluid caused membranes to appear in the cyst fluid.

revealed a cyst in the liver. Her chest X-ray, and blood chemistry was normal. She was put on 3 cycles of tablet Albendazole 400mg twice a day for 28 days, followed by 14 days of medicine free interval. Written consent was taken from the patient to publish this case. On follow up, scans of the breast showed cysts becoming progressively smaller, reaching a size of 3.2cmx1.8 cm over the next year; after which the patient was lost to follow-up.

Discussion

Hydatid disease is a parasitic disease caused by Echinococcus granulosus. Hydatid disease is found to have the highest prevalence in Mediterranean countries, the Russian Federation and its adjacent states, China, North and East Africa, Australia and South America. In Pakistan, close association of individuals with sheep and dogs and the unavailability of clean water have caused this disease to become endemic in the region.

Hydatid cysts may be found in any part of the body. The liver acts as the first filter and accounts for about 75% of all cases; while lungs, the second filter, make up about 10%; 2.5% of the cysts are found in the kidney, 2.5% in heart, 2% in bone, 1.5% in spleen, 1% in muscle and 0.5% in the brain.[1] The breast is a very rare site of infestation, as it constitutes only 0.27% of all infections. The breast can be a primary site or present as part of a systemic disease.[7]

The largest reported series in literature are 20 cases of breast hydatid cysts, reported by Quedraogo in a retrospective study (between 1969-1982).[8] Sagin et al. found 54 breast hydatid cysts which were reported until 1994 and Arikan found 10 more reports up till 2004.[9] A total of 121 cases of breast hydatid cyst were reported in the literature up to October 2018.[5] We found 10 more cases of hydatid cyst in the breast till date on PubMed search, making a total of 131 cases. Our case of hydatid cyst in the breast would add upto 132 recorded cases, according to the best of the authors' knowledge.

Breast lump is evaluated pre-operatively by the triple assessment protocol that includes clinical examination, imaging studies and cytological evaluation.[10] Hydatid cyst generally presents in the form of a slowly growing, painless and palpable mass with no signs and symptoms of malignancy. Diagnosis of hydatid cyst in breast is frequently missed clinically due to its rare occurance.

Mammographic signs of hydatid cyst are non-specific and show a homogeneous, circumscribed opacity with no suspicious micro calcifications. In over penetrated views, a ring-shaped structure may be seen inside the mass that is typical of daughter cysts inside the main cyst.[11] Since hydatid disease of the breast is extremely rare, it is usually not included in the differential diagnoses for circumscribed breast lesions, as seen on mammogram. Differentials of circumscribed opacities on mammogram include cyst, fibroadenoma, phyllodes tumour, hamartoma, intra mammary lymph node and circumscribed carcinoma.[12]

High resolution breast ultrasound is more specific than mammography in identifying a hydatid cyst. The recent consensus statement published by WHO Informal Working Group on Echinococcosis (WHO-IWGE) recommended ultrasound examination as a very useful screening tool ,having a sensitivity and specificity of 93-98% and 88-90% in endemic areas.[5] Gharbiet al. have described five types of ultrasound findings for hydatid disease: a purely cystic lesion, cyst with split wall (detached membrane giving water lily sign)[4] fluid collection with septa (daughter cysts), and a completely solid appearance and reflecting walls with calcifications.[2] Differentials for hydatid cyst on ultrasound include simple cyst, complex cyst, solid mass, intra cystic carcinoma, inflammatory carcinoma and mastitis.[13] Surgery is the recommended treatment for hydatid cyst. According to the American College of Gastroenterology Guidelines, this option is reserved for complicated hydatid cysts, large cysts (>10cm) with multiple daughter cysts, or a superficially located cyst with risk of rupture or secondary infection.[14]

Percutaneous aspiration procedure of suspected hydatid cyst was, for many years, notorious for risk of anaphylactic reaction. However aspiration cytology of fluid should enable a correct diagnosis. In literature, a total of 131 cases of hydatid cysts in the breasts were reported up to February 2021, with aspiration having been done in 28 cases. This aspiration showed the presence of fragments of laminated membrane, scolices, and hooklets, which was diagnostic of hydatid disease, with no allergic reactions noted post-procedure like in our case.[5]

Conclusion

Hydatid cysts in the breast are rare and present with non-specific clinical and imaging findings. Aspiration might help in the diagnosis by revealing post aspiration changes on ultrasound, which may be accompanied by features of hydatidosis on cytological examination of the aspirate without any complication. An index of suspicion of hydatidosis would help evaluate subtle imaging features that might increase specificity.

Disclaimer: None.

Conflict of interest: None.

Funding disclosure: None.

References

1. Kumar A, Kumar A, Gaurav K, Chandra G, Tiwary AK, Bhagat S, et al. A rare case of isolated hydatid cyst of breast. Int J Surg Case Rep. 2015; 7C:115-8. doi: 10.1016/j.ijscr.2014.10.093.
2. Gharbi HA, Hassine W, Brauner MW, Dupuch K. Ultrasound examination of the hydatic liver. Radiol. 1981; 139:459-63. doi: 10.1148/radiology.139.2.7220891.
3. WHO Informal Working Group. International classification of ultrasound images in cystic echinococcosis for application in clinical and field epidemiological settings. Acta Trop. 2003; 85:253-61. doi: 10.1016/s0001-706x(02)00223-1.
4. Durr e S, Sabih Z, Khan AN. "Congealed waterlilly" sign: a new sonographic sign of liver hydatid cyst. J Clin Ultrasound. 1996; 24:297-303. doi: 10.1002/(SICI)1097-0096(199607/08).
5. Mutafchiyski VM, Popivanov GI, Tabakov MS, Vasilev VV, Kjossev KT, Cirocchi R, et al. Cystic Echinococcosis of the Breast - Diagnostic Dilemma or just a Rare Primary Localization. Folia Med (Plovdiv). 2020; 62:23-30. doi: 10.3897/folmed.62.e47740.
6. El Moussaoui K, Lakhdar A, Baidada A, Kherbach A. Hydatid cyst of the breast: case report. Int J Surg Case Rep. 2020; 77:325-8. doi: 10.1016/j.ijscr.2020.10.109.
7. Alamer A, Aldhilan A, Makanjuola D, Alkushi A. Preoperative diagnosis of hydatid cyst of the breast: a case report. Pan Afr Med J. 2013; 14:99. doi: 10.11604/pamj.2013.14.99.2396.
8. Ouedraogo EG. [Hydatid cyst of the breast. 20 cases]. J Gynecol Obstet Biol Reprod (Paris). 1986; 15:187-94.
9. Arikan S, Yucel AF, Barut G, Kocakusak A. Hydatid disease in the breast: a case report. Acta Chir Belg. 2004; 104:473-5.
10. Singh MP, Tandon A, Huda T. Isolated primary hydatid disease of the breast masquerading a breast tumor: report of a case and review of the literature. J Parasit Dis. 2019; 43:333-6. doi: 10.1007/s12639-019-01083-w.
11. Vega A, Ortega E, Cavada A, Garijo F. Hydatid cyst of the breast: mammographic findings. AJR Am J Roentgenol. 1994; 162:825-6. doi: 10.2214/ajr.162.4.8140999.
12. Tutar N, Cakir B, Geyik E, Tarhan NC, Niron EA. Hydatid cysts in breast: mammography and ultrasound findings. Br J Radiol. 2006; 79:e114-6. doi: 10.1259/bjr/33156239.
13. Dagli AF, Ozercan MR, Kocakoc E. Hydatid cyst of the breast mimicking inflammatory carcinoma and mastitis. J Ultrasound Med. 2006; 25:1353-6. doi: 10.7863/jum.2006.25.10.1353.
14. Hassen A. Ultrasound examination of the hydatic liver. Radiology. 1981; 139:459-63. doi: 10.1148/radiology.139.2.7220891.