

Primary hydatid cyst of the breast: A case report and mini-review of the literature

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Abstract. Hydatid disease is a zoonotic infection with a diverse range of clinical presentations. Breast involvement is an extremely rare occurrence. The present study describes the case of an elderly female patient who presented with a longstanding breast hydatid cyst (HC) masquerading as a painless mass. A 79-year-old female patient presented with a slowly enlarging lump in her left breast for the past 20 years. A mammography revealed a circumscribed, hyperdense mass. The lump was surgically excised. A histopathological examination revealed benign breast ducts and lobules in a fibrous stroma containing a well-defined cyst composed of lamellated, chitinous layers of a HC. In addition, a review of 11 cases of breast HC in females identified in the literature revealed that the disease frequently presented as a painless, slowly growing breast mass. The disease duration prior to presentation ranged from 2 to 36 months. The cysts were located in the left breast in 8 cases, in the right breast in 2 cases, and there was bilateral involvement in 1 case. The majority of these cysts were localized to the upper outer quadrant. On the whole, as demonstrated herein, breast HC can remain asymptomatic for a long period of time and may masquerade as a gradually enlarging, painless breast mass.

Introduction

Hydatid disease (HD), caused by the larval stage of the tapeworm *Echinococcus granulosus*, is a zoonotic infection with

a diverse range of clinical presentations (1). Dogs are the definitive hosts of *Echinococcus granulosus*, while livestock and rodents serve as intermediate hosts. Humans can become incidental hosts when they are infected by ingesting the eggs of *Echinococcus* species (1,2). The lungs and liver are the most commonly affected organs. However, extrapulmonary involvement can occur in various locations due to the ability of the parasite to disseminate through the hematogenous route. Uncommon sites of involvement include the kidneys, pancreas, heart, brain, bones, muscles, orbits, peritoneal cavity, chest wall, urinary bladder, neck, thyroid and parotids (3-7). The breasts are an exceptionally rare site for HD. Breast hydatid cysts (BHCs) are unusual even in regions endemic for *Echinococcosis*, where they represent only 0.27% of all HC cases (1,2). Due to its low prevalence and mimicry of more frequent breast pathologies, the diagnosis of BHC is challenging (8).

In accordance with the CaReL guidelines (9), the present study describes the case of an elderly female patient who presented with a longstanding BHC masquerading as a slowly enlarging, painless mass.

Case report

Patient information. A 79-year-old female patient presented to Smart Health Tower, Sulaymaniyah, Iraq) with a palpable, painless, slowly enlarging lump in her left breast that had been present for the past 20 years. She was multiparous with no history of smoking, and her past medical and surgical histories were negative. She had no fever, weight loss, night sweats, or chest pain. She resided in an urban area, but had a history of having a pastoral lifestyle for a number of years (>10 years) with daily close contact with various farm animals, including sheep and cattle.

Clinical findings. Upon a physical examination, a palpable, non-tender, well-defined, freely mobile mass was found in the lateral central part of the left breast, ~40 mm in diameter. The physical examination did not reveal any other notable findings.

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Diagnostic assessment. A breast ultrasound (U/S) revealed (images not available) a heterogeneous mass with internal layering and calcification, but no vascularity. A mammography (MMG) revealed a circumscribed, hyperdense mass, measuring 35x23 mm, in the lateral central part of the left breast. The radiological features were suggestive of a calcified HC. The mammogram classified the mass as BI-RAD 2 (Fig. 1). Based on radiological findings and to minimize costs for the patient, magnetic resonance imaging (MRI) and serology were not performed.

Therapeutic intervention. The patient underwent surgery to excise the mass. The skin overlying the mass was marked for an elliptical incision. The mass was then identified and carefully dissected free from the surrounding breast tissue. Great care was taken to ensure the complete excision of the mass, while avoiding the rupture of the cyst wall. Hemostasis was achieved throughout the dissection using electrocautery. The entire mass was removed intact without the spillage of the contents of the cyst. The surgical cavity was irrigated with sterile saline to remove residual debris (Fig. 2A). A drain was deemed unnecessary due to the absence of visible bleeding and a clean surgical field. The wound was closed in layers with absorbable sutures, followed by the closure of the skin incision with subcuticular sutures for an optimal cosmetic outcome. The cyst was ruptured during post-resection manipulation (Fig. 2B). A histopathological examination was performed on 5- μ m-thick sections which were paraffin-embedded. The sections were then fixed in 10% neutral-buffered formalin at room temperature for 24 h and stained with hematoxylin and eosin (H&E; Bio Optica Co, Italy) for 1-2 min at room temperature. The sections were then examined under a light microscope (Leica Microsystems GmbH). This examination revealed benign breast ducts and lobules in a fibrous stroma surrounding a well-defined cyst composed of an outer fibrous layer and an inner lamellated, chitinous layer with calcification and mixed inflammatory cell infiltration. This confirmed the diagnosis of a calcified BHC (Fig. 3).

Follow-up and outcome. The post-operative period was uneventful, and the patient reported no wound complications (Fig. 4). A U/S examination at the 4-month follow-up (images not available) revealed no evidence of recurrence.

Discussion

Echinococcus is a genus of tapeworms that cause a parasitic disease in humans and animals known as echinococcosis. Cystic echinococcosis is the most common of the four main types of the disease, also known as HD. The larval stage of *Echinococcus granulosus* is responsible for causing HD (8). It is an endemic disease in a number of areas, including the Mediterranean, South America, North and East Africa, the Middle East, China, Australia and Russia (10). It is primarily observed in places where sheep and cattle are raised (11).

The *Echinococcus* life cycle requires a definitive host and an intermediate host. Humans can become intermediate hosts (8) when they become accidentally infected by ingesting the eggs of the parasite, which are shed in the feces of infected dogs (12). This can occur through close contact with infected

dogs, particularly when handwashing is omitted following direct contact or when contaminated food or water is ingested. The ingested eggs travel to the human intestines, where they hatch and release oncospheres (1). These embryos then migrate through the bloodstream to different organs, most commonly the liver (in 75% of cases) and lungs (in 15% of cases), followed by other organs (in 10% of cases) (3,13). The embryos develop into HCs in these organs, which can grow slowly over the years. The most probable route for disseminating HD is the bowel lymphatics into the systemic bloodstream (14).

The lymphatic pathways between the liver and breast (Gerota's pathway) are a possible route for breast involvement by HD (14). Herein, following a literature review, it was found that the reported cases mostly occurred between the ages of 30 and 50 years, usually presenting with a history of a painless, slowly enlarging breast mass (3,12,13). A literature review was conducted to identify relevant reports on BHC, filtered by the well-known predatory lists (15). The process involved a Google Scholar search employing the 'breast hydatid cyst' as a key word. In total, 13 reports on BHCs were identified from 2021 to 2024, of which 11 reports were summarized in the present study (2,8,16-24). Among these, rural residency was reported in 3 cases, with an age range of 18 to 75 years and a mean age of 37.6 years. The majority of these cases (90.9%) presented with a breast lump (Table I). In their systematic review, Mutafchiyski *et al* (14) reported that in a total of 52 cases of BHC, the duration of the presentation ranged from 4 months to 19 years. The majority of cases involved a single cyst, with only 2 cases having multiple cysts (14). In a case series, Tavakoli *et al* (10) described 6 cases with symptom durations ranging from 8 months to 3 years, while Koc *et al* (13) reported a case with a longstanding history before presentation, a finding similar to the case presented herein. Among the reviewed cases that specified the duration of presentation, this ranged from 2 to 36 months, with a mean of 15.4 months. The left breast was the most frequently affected side (72.7%), and the upper quadrant was the most commonly involved region (54.5%). In the patient in the present case report, the lesion was located in the lateral central portion of the left breast.

BHC can mimic a simple cyst, fibroadenoma, phyllodes tumor, chronic abscess, or cancer (8,12). Fibroadenomas have an excellent prognosis with minimal malignant potential and may even regress spontaneously (25). BHC also exhibits a benign course following resection; however, follow-up is recommended to monitor for recurrence (26). By contrast, breast carcinoma has a stage-dependent prognosis, with the 5-year survival >99% in localized disease, yet decreasing to ~87% with regional spread and ~32% with metastasis (27).

Generally, HC is an essential consideration in the differential diagnosis of a palpable breast mass, particularly in individuals from regions where HD is endemic (2). Therefore, a triple assessment is valuable for excluding malignancy in any breast mass, including a comprehensive history and physical examination, radiological imaging and histopathological analysis (28). Clinically, HC appears as a firm, mobile, often painless lump of variable size with a regular border (1). Imaging may be helpful in the diagnosis of breast HD, although it is usually not conclusive (1,12). Breast U/S is the method of choice for evaluating this type of cystic lesion. It has a sensitivity of 88-98% and a specificity of 95-100% (8,10). Breast

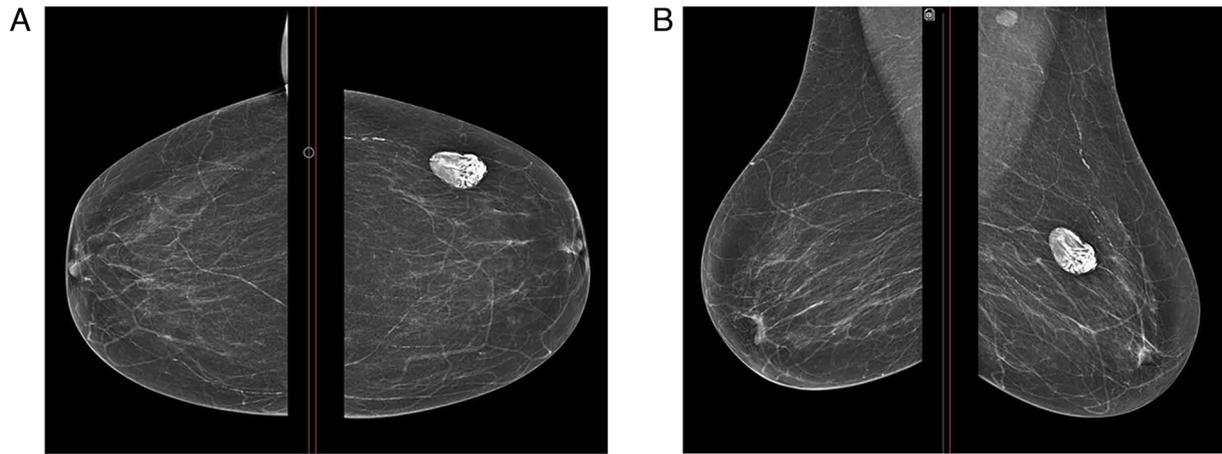


Figure 1. Mammography imaging illustrating an oval, hyperdense, layering calcified lesion in the upper outer part of the left breast, suggestive of a calcified hydatid cyst. (A) Craniocaudal view, (B) mediolateral oblique view.

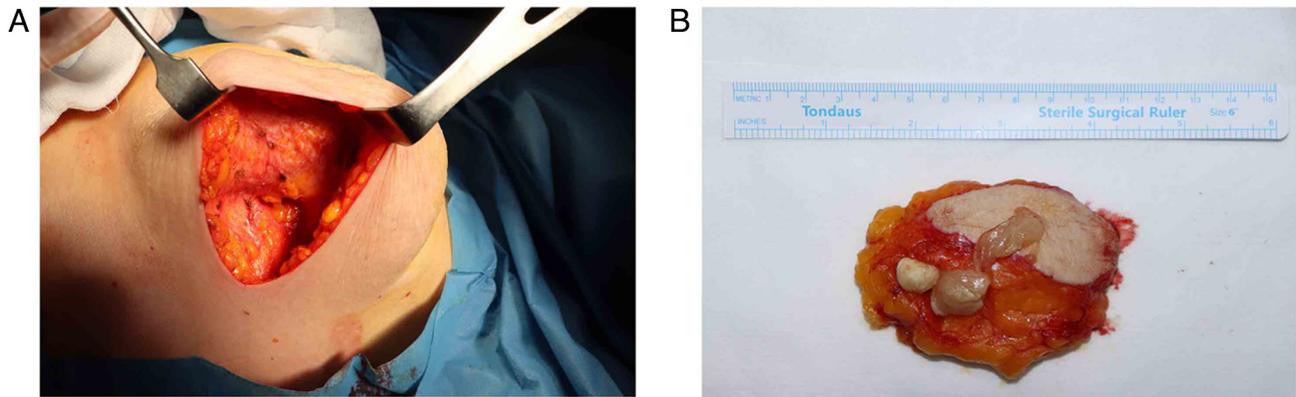


Figure 2. (A) The surgical bed following excision and irrigation with normal saline. (B) The ruptured cystic mass due to post-resection manipulation.



Figure 3. Histopathological examination illustrating benign breast ducts lined by bland epithelial cells (yellow arrows) surrounding a cyst that has an outer fibrous wall and an inner chitinous layer characteristic of a hydatid cyst (black arrows) (hematoxylin and eosin staining; magnification, x40).



Figure 4. Clinical appearance at the 3-week post-operative follow-up.

U/S can indicate a well-defined, sometimes lobulated mass with mixed internal echoes, potentially containing both cystic and solid areas. According to the classification presented in the

study by Gharbi *et al* (29), five ultrasonographic features have been described: An uninoculated pure fluid collection (type I); a fluid collection with a split wall (type II); a multi-vesicular,

Table I. Summary of 11 case reports on breast hydatid cysts in females from 2021 to 2024 identified in the literature.

First author	Country, year of publication	Age, years	Laterality	Presentation	Duration (months)	Residency	Radiology	Location	Size (cm)	Pre-operative assessment				Management	Follow-up	Outcome	(Refs.)
										Axillary lymphadenopathy	Biopsy	Other involved organs	Biopsy				
Kassahun Tadele	Ethiopia, 2022	18	Left	Painless breast lump	24	Rural	U/S	UQ	5.5	No	Granulomatous inflammation	No	Surgery and post-operative albendazole	6 months	No recurrence	(2)	
Al Sharei	Jordan, 2023	38	Bilateral	Painless breast lump	2	Rural	U/S, MMG	UOQ, upper central	2.3	No	No	Liver	Pre-operative albendazole and surgery	3 months	No recurrence	(8)	
Mesfin	Ethiopia, 2023	28	Left	Breast pain	12	NA	U/S	LOQ	3.8	NA	No	No	Pre-operative albendazole and surgery	NA	NA	(16)	
Samsami	Iran, 2021	31	Left	Painless breast lump	24	NA	U/S	Axillary tail	5	NA	No	No	Surgery and post-operative albendazole	One year	No recurrence	(17)	
Alareqi	Yemen, 2021	23	Left	Breast lump	3	NA	U/S, MMG	UOQ	2.9	Yes	Hydatid cyst	No	Albendazole and antibiotics	NA	NA	(18)	
Abu-Mandeel	Jordan, 2023	38	Left	Painless breast lump	7	NA	U/S, MMG	UOQ	3.2	No	No	Liver	Surgery and post-operative albendazole	NA	No recurrence	(19)	
Dattal	India, 2023	75	Right	Painless breast lump	NA	NA	U/S, MMG	Retro-areolar	4	No	Cellular debris	No	Surgical excision	NA	NA	(20)	
Ines	Tunisia, 2022	50	Left	Painless breast lump	Few years ^a	Rural	U/S, MMG, MRI	LQ	7	No	No	No	Pre-operative albendazole and surgery	6 months	No recurrence	(21)	
Assefa	Ethiopia, 2022	28	Left	Painless breast lump	NA	NA	U/S	UOQ	3.4	No	Hydatid cyst	No	Surgical excision	NA	NA	(22)	
Mahmood	Pakistan, 2023	50	Left	Painless breast lump	36	NA	U/S, MMG	UOQ	3.5	No	Hydatid cyst	Liver	Albendazole	Lost to follow-up	NA	(23)	

Table I. Continued.

First author	Country, year of publication	Age, years	Laterality	Presentation	Duration (months)	Residency	Radiology	Location	Size (cm)	Pre-operative assessment			Management	Follow-up	Outcome	(Refs.)
										Axillary lymphadenopathy	Biopsy	Other involved organs				
Sharma	India, 2021	35	Right	Painless breast lump	NA	NA	U/S, MMG	NA	4.2	No	Simple cystic lesion	No	Surgical excision	NA	NA	(24)

*Exact duration not mentioned. NA, not available; UOQ, upper outer quadrant; UQ, upper quadrant; LOQ, lower outer quadrant; LQ, lower quadrant; U/S, ultrasound; MMG, mammography; MRI, magnetic resonance imaging.

multiseptated cyst with daughter cysts (type III); a mass with a heterogeneous echo pattern (type IV); and a mass with reflecting thick walls (type V). Types II and III HC have more specific diagnostic imaging features than the other types. MMG may illustrate a non-specific, well-circumscribed, round, or oval-shaped mass with internal ring-shaped structures (1). Calcifications within the cyst wall or daughter cysts may be visualized (20). Upon imaging, the case in the present study was found to have type IV features (a mass with a heterogeneous echo pattern) according to the classification presented in the study by Gharbi *et al* (29). However, fibroadenomas typically appear as well-circumscribed, oval, or lobulated masses with smooth, sharp margins on mammography and as uniformly hypoechoic, circumscribed lesions on ultrasound (25). By contrast, invasive breast carcinomas typically present as irregular or spiculated high-density masses (often accompanied by microcalcifications) on MMG and as angular, non-parallel hypoechoic lesions with posterior acoustic shadowing on ultrasound (30,31).

A breast MRI can be a helpful tool for the diagnosis of HC in the breast (13), as it can provide more detailed information about the size, location and characteristics of the cyst. A well-defined cystic lesion with a smooth wall, perilesional edema (fluid build-up around the cyst), daughter cysts (smaller cysts within the larger cyst), T1 hypo-intensity (dark signal on T1-weighted images), T2 hyperintensity (bright signal on T2-weighted images) and peripheral rim enhancement (increased uptake of contrast dye along the rim of the cyst) are the MRI findings that are suggestive of an HC (10,20). Notably, these findings are not specific to HCs and can also be observed in other breast lesions (13). The results of a breast MRI need to be interpreted in conjunction with other clinical information, such as the medical history of the patient and the results of the physical examination, in order to ensure an accurate diagnosis. In their systematic review, Mutafchiyski *et al* (14) reported that MRI was used as a diagnostic modality in only a few cases. Among the 11 reviewed cases in the present study, only 1 case had undergone a breast MRI (21).

Fine needle aspiration cytology was previously considered controversial in the pre-operative diagnosis of HC in the case that the U/S examination suggested the disease, as it may lead to spillage and anaphylactic reaction (10,24). Recent literature, however, has concluded that the procedure may be safe, fast and inexpensive (2,12,32). The microscopic identification of the scolices or hooklets in the fluid is required for the diagnosis (12,14). Serological tests can be used for diagnosis, screening and post-operative follow-up, including enzyme-linked immunosorbent assay (ELISA), hydatid immunoelectrophoresis, latex agglutination and an indirect hemagglutination test. The sensitivity of ELISA ranges from 80-100%, and its specificity ranges from 88-96% for hepatic cysts. The sensitivity of the test ranges from 50 to 56% for lung HD and from 25 to 65% for HD of other organs (4). Since serological tests demonstrate variable sensitivity in diagnosing HCs at different anatomical sites, the development of a rapid and reliable serological assay for extrapulmonary HC remains necessary (33). Therefore, U/S, MRI or MMG may help rule out the presence of HC in the breast (13,21), as in the case in the present study.

Surgical intervention, puncture aspiration injection and re-aspiration, a 'watch and wait' approach, and chemotherapy are the main treatment options for HC (4,8). However, complete surgical excision is the best diagnostic and therapeutic approach (14). By contrast, fibroadenomas are usually observed or removed by local excision if large or symptomatic, while invasive carcinomas require oncologic surgery, often with additional radiotherapy, chemotherapy, or hormonal therapy depending on stage and tumor biology (25,34).

A definitive diagnosis of HC is often achieved through a post-operative histopathological examination (8). HCs have a multilayered wall composed of a laminated membrane and a germinal layer that produces protoscoleces and daughter vesicles, typically accompanied by surrounding fibrosis and inflammation (26). However, fibroadenomas are benign, well-circumscribed tumors of stromal and ductal elements with intact myoepithelial lining and no atypia (25). Breast carcinomas exhibit malignant invasion beyond the basement membrane, characterized by nuclear pleomorphism, increased mitotic activity, and disorganized growth patterns (34). Preoperative chemotherapeutic agents, such as albendazole, can reduce the postoperative recurrence rate (2,3), which ranges from 2 to 25% (3). However, among the cases reviewed herein, only 3 cases used pre-operative albendazole (8,16,21). The mortality rate due to echinococcosis is very low, ranging between 0.29 to 0.6% (4). Complete surgical excision with intact borders followed by a course of albendazole was the favored treatment in many studies (1,2,8,25). Of the cases reviewed herein, 4 cases were administered post-operative albendazole (2,17-19). The current case underwent the complete surgical excision of the cyst without postoperative complications. Histopathological examination confirmed BHC. Following a 4-month follow-up, no recurrence was reported. Further studies with larger sample sizes and more robust designs are warranted to elucidate the mechanisms underlying BHC occurrence.

In conclusion, BHC can remain asymptomatic for a long period of time and may masquerade as a gradually enlarging, painless breast mass.

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Availability of data and materials

The data generated in the present study may be requested from the corresponding author.

Authors' contributions

AMS and SHH were major contributors to the conception of the study, as well as to the literature search for related studies. HOAb, SL and BOH were involved in the literature review, in the conception of the study and in the writing of the manuscript. HOAl, KAA, RMA, FHF and SMA were involved in

the literature review, in the design of the study, in the critical revision of the manuscript, and in the processing of the table and figures. LRAP was the radiologist who performed the assessment of the case. AMA was the histopathologist who performed the diagnosis of the case. AMS, SL, BOH and HOAl were involved in the management and monitoring of the case. AMS and SHH confirm the authenticity of all the raw data. All authors have read and approved the final manuscript.

Ethics approval and consent to participate

Written informed consent was obtained from the patient for her participation in the present study.

Patient consent for publication

Written informed consent was obtained from the patient for the publication of the present and any accompanying images.

Competing interests

The authors declare that they have no competing interests.

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