



Bilateral hydatid cyst of the breast: a case report and review of the literature

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Abstract

Hydatid cyst disease is a parasitic infestation caused by the tapeworm *Echinococcus granulosus*. The incidence of the disease is 1–200 cases per 100 000. The usual presentation is the formation of fluid-filled cysts in the liver or lungs and, less commonly, in the brain. The breast is a rare site of presentation for this disease. An isolated hydatid cyst in the breast is very rare, even in endemic regions, occurring in only 0.27% of all hydatid cases. Although these cases are very rare, they should be considered severe. A hydatid breast cyst may mimic a fibroadenoma, phyllodes tumor, chronic abscess, or even carcinoma. Therefore, a high incidence of misdiagnosis may occur. Preoperative diagnosis can be made using needle aspiration and imaging facilities such as breast ultrasonography, mammograms, and computerized topographic images. Nevertheless, in most cases, the final diagnosis cannot be made until the pathological report is obtained postsurgery. This paper reports the first case of a bilateral hydatid cyst of the breast that was diagnosed preoperatively by radiological facilities. We present the patient history, physical examination, and investigations, discuss the risk factors, and present our treatment plan.

Keywords: breast lump, echinococcus granulosus, hydatid cyst

Introduction and importance

Hydatid cyst disease is a parasitic infestation caused by the tapeworm *Echinococcus granulosus*. In cystic echinococcosis (CE), the human is an aberrant host infected by oral ingestion of excreta from an infected dog^[1]. Sheep, cows, and even fish are the intermediate hosts of *E. granulosus*^[2]. Moreover, humans can become infected via the ingestion of food contaminated with feces of infected dogs.

Many patients are infected in their childhood. Most cases are from rural areas owing to poor sanitation, such as villages, due to poor water drainage and low socioeconomic status^[2]. The usual presentation is the formation of fluid-filled cysts in the liver or lungs and, less commonly, in the brain. Many have been reported

HIGHLIGHTS

- A hydatid cyst of the breast is an extremely rare disease.
- Symptoms may mimic those of breast cancer in that they are hard and painless.
- Diagnosis before surgery is of paramount importance to avoid cyst rupture and anaphylactic shock.
- We report the first case of bilateral hydatid cyst of the breast that was diagnosed preoperatively.

in the spleen, kidneys, muscles, bones, and retroperitoneum^[3]. The breast is a rare site of presentation for this disease.

An isolated hydatid cyst in the breast is very rare, even in endemic regions, occurring in only 0.27% of all hydatid cases^[1]. Parasitic infestations of the breast are usually due to tapeworm larvae, *Taenia solium*^[3].

There is not enough literature on breast infestation. Most reported cases were diagnosed postsurgery after definitive histopathological examinations. The preoperative diagnosis is difficult to reach based on the classical modalities (the history, examination, and investigations) as an isolated breast hydatid cyst may mimic a fibroadenoma, phyllodes tumor, chronic abscess, or even carcinoma^[1]. Radiological findings range from a purely cystic lesion to a completely solid appearance. Therefore, a high incidence of misdiagnosis may occur. Herein, we report the first case of a bilateral hydatid cyst of the breast that was diagnosed preoperatively by radiological facilities.

In compliance with the SCARE (Surgical Case Report) guidelines^[4], we report a case of bilateral hydatid cyst of the breast that was diagnosed before surgery. The patient was successfully treated surgically.

This report presents the first case of a bilateral hydatid cyst of the breast that was diagnosed preoperatively by radiological

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facilities. We believe our study makes a significant contribution to the literature because the breast is a rare presentation site for this disease. This work also adds to the limited reports on this disease condition in the literature. Our reporting of this case would also limit the incidence of misdiagnosis.

Case presentation

A 38-year-old married housewife discovered a mass in her left breast while showering, and the mass grew slowly. Consequently, the patient was brought to a public hospital.

By taking a relevant history, the mass had grown by 3 cm in the past 2 months. There was associated bilateral breast skin dryness. The patient mentioned that there was no weight loss, no feeling of hotness, and no back or bone pain. There was no abdominal pain, shortness of breath, or hemoptysis. The patient was worried that her breast mass might be malignant.

The patient had free medical and surgical records with no previous breast condition complaints. The patient's medical records showed that she was not on any medications, and none of her family members had similar medical conditions or genetic disorders. The patient's psychological profile was also free.

There were no known allergies or history of smoking. There was no family history of breast cancer. The patient mentioned that she had lived in a rural area when she was a child, and there were castles and dogs near her house. Moreover, she had no recent traveling history.

On physical examination, there was a left breast cystic mass 3 cm from the nipple at the 2 o'clock position, which was non-fixed, mobile, with regular margins and a smooth surface. In addition, there was a 1 cm measuring mass in the right breast at 12 o'clock. No palpable bilateral lymph nodes were observed.

Ultrasonographic examination showed a 3.3 × 2.9 cm cystic mass with a mixed solid and cystic component. It occupied the upper outer quadrant of the left breast. Examining the left breast showed a 1.1 × 1.3 cm mass at the 12 o'clock position. A focal cystic lesion was observed in the liver. This was correlated with the appearance of hydatid cysts in the liver.

A breast mammogram showed a 3.2 × 3 × 3 cm cystic mass. It was at a 7 cm distance from the nipple, 0.3 cm from the skin, and 6.5 cm from the pectoral muscle. In addition, there was a 12 o'clock right breast mass which measured 1.1 × 0.7 × 1 cm.

Differential diagnoses include phyllodes tumor, breast abscess, breast cancer, and hydatid cyst.

A brain computed tomography (CT) was ordered. However, it revealed free brain tissue without any visible pathology during the examination.

A chest, abdomen, and pelvis CT scan with intravenous (i.v.) contrast showed that the thoracic region was free. However, the triphasic liver CT scan revealed multiple variable-size hypodense lesions, and after contrast administration, they showed wall enhancement (halo sign).

Provisional differential diagnosis was discussed with the patient. The risks and benefits of the surgical procedure were also discussed with the patient, and the prognosis was clarified. The patient showed no signs of concern and was very cooperative. The decision was made after consenting the patient to proceed with a surgical treatment approach for both breasts after a course of Albendazole. Surgery was done under general anesthesia and performed by a senior registered breast oncoplastic surgeon. The

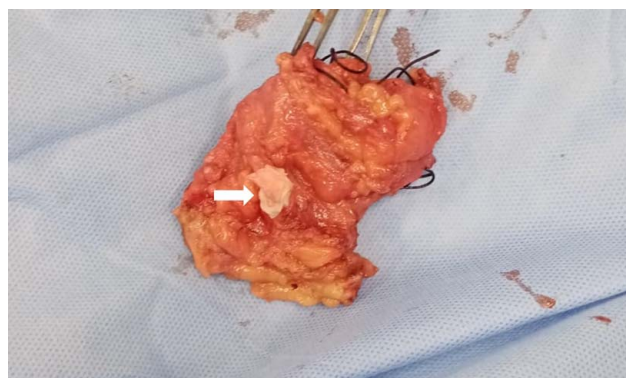


Figure 1. The excised cyst with germinal layer (white arrow).

skin was incised, and two cysts were delivered with no rupture (Fig. 1), Stablon (hypertonic saline) was infused into the wound, and plastic closure was performed with a subcutaneous flap.

Histopathological testing revealed the appearance of mixed cystic and solid components in both samples. Microscopic testing confirmed the diagnosis of hydatid disease based on the acellular laminated wall of the cyst.

The postoperative period was uneventful, and no complications were noted. The patient was followed up for 3 months and was completely satisfied with the procedure and the results. However, the General Surgery Department was consulted for the liver lesion, and a clinic appointment was arranged to follow up with the patient.

Review of the literature

Echinococcal disease is a zoonotic cystic disease that affects humans. It is mainly caused by the infestation of *E. granulosus*. Hydatid cysts are epidemiologically considered endemic in many areas of the world. This disease is usually found in the liver and lungs but may develop elsewhere. A hydatid breast cyst is a rare clinical entity. This is why there are not many studies in the literature regarding this disease. Most published studies are limited to case reports. As a result, some of the published studies and reports have been reviewed in this article.

Age

A literature review of online published studies reporting cystic breast echinococcosis performed by Mutafchiyski *et al.*^[5] concluded that the mean age of 121 reviewed cases was 40.5 years (range: 18–70 years). In contrast, a case series published in Iran revealed that the mean age of the patients was 32 (range: 25–52) years^[6].

Clinical presentation

In Mutafchiyski *et al.*'s^[5] literature search study, a gradually growing painless mass was the cornerstone of the clinical presentation. Similarly, Tavakoli *et al.*'s^[6] case series concluded that a breast lump was the most common chief complaint of the reported cases. On the other hand, the abscess was the initial presentation in two cases in Mutafchiyski *et al.*'s^[5] review. In addition, regarding physical examination, Mutafchiyski *et al.*'s^[5] study suggest that the size of the cysts was 5.5 cm (range:

1.7–12 cm), while it was 3.5 cm (range: 2–8 cm) in the Tavakoli *et al.*'s report with no associated skin changes or nipple deformities^[6]. In both studies, the majority of cases were unilateral^[5,6].

Diagnostic modalities

In a case series carried out in Iran, mammography and ultrasound were the mainstays of diagnosis modalities. They detected an oval, well-circumscribed mass with smooth margins and homogeneous density, with a benign lesion appearance. Moreover, a cystic mass with multiple thick internal septa and echogenic materials was enormously detected ultrasonographically^[6].

On the other hand, Mutafchiyski *et al.* state that breast ultrasound was the most frequently used diagnostic modality. In contrast, CT and magnetic resonance tomography were performed in a few cases. More importantly, mammography was used in 35% of the cases^[5].

Biopsy

Significantly, in the Iranian case series, a fine-needle aspiration biopsy of the breast was performed in one of the reported cases. In contrast, a core needle biopsy was conducted on two patients. In the pathological analysis, microscopic protoscolices hooklets confirmed the diagnosis of hydatid cysts^[6]. Likewise, in a literature review of 121 cases, fine-needle aspiration with the cytological examination was positive in 59% of the cases, demonstrating scattered hooklets, scolices, and laminated membranes^[5].

Management

In Tavakoli *et al.*'s report, the breast masses were managed by complete surgical excision, and the breast hydatid cyst was confirmed by histological examination in all patients, including the three patients who had undergone excisional biopsies. Postoperatively, all patients were treated with Albendazole (10 mg/kg/day) for 3 months to decrease the recurrence rate of the hydatid disease^[6].

Correspondingly, surgical excision of the mass was performed in almost all of the studied cases reviewed by Mutafchiyski *et al.*; however, one was managed conservatively as the patient refused the operation and was followed up for 2 years without cyst progression. As stated in this review, there was no information regarding the treatment with Albendazole; most cases were not treated, and some authors reported a 4-week postoperative course, whereas few reported a preoperative course^[5].

Clinical discussion

Hydatid cyst, or CE, is caused by infection with *E. granulosus* sensu lato, an endoparasitic tapeworm^[7], resulting in a zoonotic disease that is chronic and complex^[8], and possibly fatal if left untreated^[9]. The main risk factors are raising livestock and coming into contact with dogs^[10].

The lifecycle of *E. granulosus* involves two mammalian hosts: an intermediate and a definitive host^[11]. Carnivores are considered to be definitive hosts, whereas their herbivorous prey serves as intermediate hosts. Humans may act as intermediate hosts if certain circumstances are present; otherwise, they are not involved in the transmission directly^[9].

If inadvertently ingested by humans through contaminated food, eggs hatch into oncospheres and enter the portal circulation and lymphatic vessels via the gastrointestinal tract. They then develop into larvae (metacestodes or hydatid cysts) within the viscera. The liver is predominantly involved, followed by the lungs, whereas the kidneys, heart, brain, bones, and spleen are less frequently involved^[8–10]. Even in endemic regions such as western China, South America, Northern and Eastern Africa, Central Asia, the Mediterranean, and the Middle East, hydatid cysts of the breast are extremely rare^[10,12]. Our patient, who spent her childhood in a rural area, might have come into contact with cattle and canines and gotten infected back then, keeping in mind the absence of any recent travel.

Our patient presented as a typical case of CE of the breast: a female between 30 and 50 years of age complaining of a palpable, painless mass in her left breast that had slowly increased in size over the past 2 months^[13]. However, physical examination revealed a second, smaller mass contralaterally. Both masses were well-defined, mobile, nontender, and had firm consistency. No regional lymph node involvement was observed. To our best knowledge, bilateral hydatid cysts in the breast have not yet been reported in the literature.

A hydatid cyst mass mimics fibroadenoma, phyllodes tumor, and carcinoma. If infected, it would mimic a breast abscess as well^[12–14]. These were on the list of our differential diagnoses, even though the patient did not have risk factors for breast cancer.

Following detailed history taking and physical examination, imaging modalities were used to assess the lump. Ultrasonography has been deemed the gold standard for diagnosing, staging, and follow-up for this specific disease. It has a sensitivity of 88–98% and a specificity of 95–100%. In addition, ultrasound can provide more details regarding the cyst's location, size, condition, and whether any changes have occurred. Moreover, ultrasonography is noninvasive, painless, quick, easy, and affordable. Overall, it is more convenient for patients and medical personnel^[8,12]. Ultrasound revealed a cystic mass in each breast with mixed solid and cystic components. The mass in the left breast measured 3.3 × 2.9 cm, while the mass in the right breast measured 1.1 × 1.3 cm. Moreover, because primary hydatid disease of the breast is sporadic^[14], the liver was also examined, and a cystic lesion was revealed.

Due to patient-specific reasons or cyst location, if ultrasound cannot be performed, other imaging modalities can be used to detect hydatid cysts, including MRI and CT scans, with MRI being superior to CT scans^[12]. A well-circumscribed cystic mass with capsular enhancement on MRI suggests a hydatid cyst, provided that the physical examination was negative for breast abscess^[14]. At our institute, CT is more readily available than MRI. It revealed multiple hypodense lesions in the liver that were variable in size, and wall enhancement was evident upon i.v. contrast administration. In addition to the breast lesions, the patient's body was otherwise clear.

On mammography, the hydatid cyst is seen as a well-circumscribed mass, and if present, the daughter cysts would appear as ring-shaped structures or internal bands within the cyst. If calcifications are seen, a calcified fibroadenoma is added to the list of differential diagnoses. Unfortunately, mammography cannot confirm the diagnosis of hydatid cysts without daughter cysts^[14].

Serological testing for CE antibodies has been considered a way of diagnosing CE. However, it has not proven reliable owing

to many factors, including high false-negative rates, expensive materials and reagents, and their invasive nature. Moreover, the sample takes a long time to process and is easily contaminated outside laboratory settings^[8]. However, it may be used as a confirmatory test^[14]. Still, this is limited to early cyst stages when the hydatid fluid is confined within the endocyst and in the final stage when the contents are solid and the wall is calcified. Therefore, serology has a limited and minor role^[12].

Treatment options for hydatid cysts include surgery, puncture aspiration injection and re-aspiration (PAIR), a ‘watch and wait’ approach, and chemotherapy. Choosing the best option depends on the location and size of the cyst, its stage, and whether complications are present or not^[6]. The most appropriate treatment plan for the breast hydatid cyst in our patient was the administration of the chemotherapeutic agent Albendazole, followed by surgical excision.

The cysts were examined under a microscope. Protoscolices were observed, with a cellular germinal layer and a thick, laminated acellular layer, all of which are characteristic features of CE^[15].

Conclusion

Hydatid cysts are caused by infection with *E. granulosus* sensu lato via the ingestion of contaminated food or water. It typically presents as a cystic mass in the viscera, with the liver being the most common site, whereas the breast is one of the rarest. Ultrasound is the gold standard for diagnosing hydatid cysts, yet the histopathology results make a definitive diagnosis. The treatment begins with a course of Albendazole, followed by surgical excision.

Ethical approval

This study was approved by the Ethical Committee of Al-Bashir Hospital, Amman, Jordan, which complies with the Declaration of Helsinki guidelines.

Consent

Written informed consent was obtained from the patient for the publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

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Author contribution

A.A.S.: writing the paper and critical revision of the work; M.A.-J.: collecting data; M.A.-K.: writing the introduction section; M.A.-A.: writing the case presentation; H.A.-A.: writing the clinical

discussion; S.A.-D.: critical revision of the work; M.A.-S.: diagnosed and treated the case and the final work revision.

Conflicts of interest disclosure

All authors have no conflicts of interest to declare.

Guarantor

The author accepts full responsibility for this work, has access to the data, and controls the decision to publish.

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