BREAST IMAGING

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Primary Hydatid Cyst of the Breast: A Case Report

Cystic hydatid disease is caused by the parasite *Echinococcus granulosus*. The breast is a rare primary site of hydatid disease that accounts for only 0.27% of all cases. The cyst is usually asymptomatic and should be included under differential diagnosis of a breast lump, especially in endemic areas of this disease. Herein, we described the biopsy-proven case of a 46-year-old woman with primary hydatid disease of left breast.

Keywords: breast, echinococcus granulosus, hemagglutination, mammography, ultrasonography

Introduction

Hydatid disease is caused by the larval form of *Echinococcus granolusus*. Humans are occasional intermediate hosts of this organism. Hydatid disease still represents an important medical and surgical problem in many regions, specially in countries where sheep breeding constitutes an important means of livehood. Although the liver and lung are the most common involved organs, hydatid disease can occur in all viscera and soft tissues.^{1,2} Seventy percent of the hydatid cysts are detected in the liver, 20% in the lung, 2% in the kidneys, 2.5% in the heart and less than 2% in the muscle and brain.^{3,4} Other rare sites are bladder, chest wall, parotid glands, thyroid and breast.² Primary hydatid disease of the breast is rare but can present as a lump in breast. It accounts for 0.27% of all occurances.^{1-3,5}

Fine needle aspiration cytology (FNAC) which might show the diagnostic hooklets or the laminated membrane, can provide a pre-operative diagnosis.^{1,6} Mammography, ultrasonography and serologic tests can also be done, which are not definitive.^{1,4,5} Complete exision of the cyst without spillage can be a curative procedure.^{1,3} Although primary breast involvement is rare, radiologist should be aware of this potentially serious disease in areas endemic for the disease.^{7,8} Hyda-tid disease is endemic in Iran and it represents a major health problem. During the last ten years, five patients with primary hydatid cysts of the breast were reported from Iran in literature. Herein, we have reported on another case with this rare condition.

Case Presentation

A 46-year-old woman, a housewife, who reared cattle and worked in a farm, presented with a large painless lump in her left breast for five years. She had no history of injury, nipple discharge, fever or other symptoms. She had no risk factors for breast cancer.

Physical examination of the left breast revealed a very large firm painless lump measuring $8 \times 10 \times 10$ cm with lobulated margins in the central part of the breast; the mass had no adhesion to the skin. There was no nipple retraction, skin

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thickening or axillary lymphadenopathy. Physical examination was otherwise normal. The clinical diagnosis was consistent with a giant fibroadenoma or a large cyst in the breast. Ultrasonography of the left breast showed a multiloculated cystic lesion measuring $8 \times 10 \times 10$ cm with a thick wall and mobile internal echos (Fig. 1). Mammography revealed a very large, dens lobulated mass without calcification at the site of palpable lump in the left breast (Fig. 2). Our preoperative diagnosis was a complicated cyst in the breast. FNAC from the breast lump was inconclusive and a clear fluid was obtained. Lumpectomy was done and at surgery, complete excision was performed. On sectioning, it was realized that this large lump could be a hydatid cyst since a laminated membrane was seen at the site of the mass. The mass contained a clear yellowish thick fluid and has a dens surounding fibrous tissue. Histologic examination revealed the diagnostic hooklets, laminated membrane and confirmed a primary hydatid cyst of the breast (Figs. 3 and 4).

Post-operative indirect echinococal hemagglutination test was positive (1,900) suggesting hydatid disease (a titer>1,100 is considered positive result).

Post-operative mammography and ultrasonography of the contralateral breast and abdomen, as well as chest radiogram were normal to rule out any dissemination in other organs. She remained free of any recurrance 20 months after surgery.

Discussion

Hydatid cysts are caused by larval tapworm of the genus Echinococcus. E. granulosus is the most common species, but E. multilocularis and E. olgarthus also infect man. The disease is a serious problem in sheep and cattle raising areas, particularly Australia, New-Zealand, the Middle East, Mediteranean countries, Africa and South America.^{2,8}

Hydatid disease of the breast is rare, accounting for only 0.27% of the localization of the cyst.^{1-3,5} The breast can be the only primary site or part of disseminated hydatidosis. In spite of its rareness, primary involvement constitutes a differential diagnosis of the breast lump in endemic areas.¹ It generally affects women 30–50 years of age, although a wide age range (26–74 years) has been reported.⁴ It usually presents as a slow progressive painless lump without any specific signs and symptoms; when secondary infection exists, the lesion is clinically indistinguishable from breast abscess.^{2,4,9}

Clinically, a hydatid cyst in the breast might mimic cysts, fibroadenomas, phyllodes tumors, circumscribed carcinomas, chronic abscesses, and cystic



Fig. 1. Ultrasonogram demonstrates a lobulated multiloculated cystic lesion with thick wall in the breast.

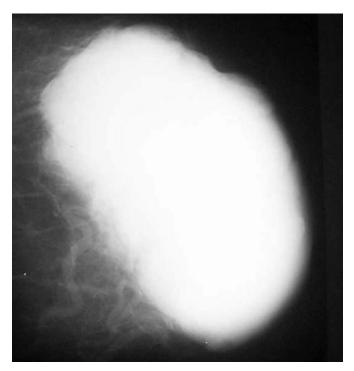


Fig. 2. Mammography demonstrates a large dens lobulated mass without calcification.

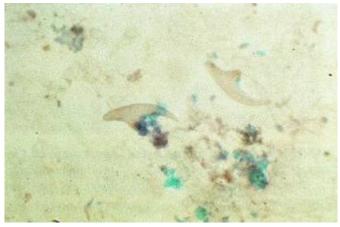


Fig. 3. Papanicolao stain showes hooklets in the backgraund of cystic debris.

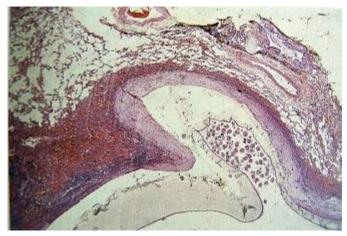


Fig. 4. Histological section shows fibrous capsule and the laminated membrane in the wall of the cystic mass.

mastopathies.^{2,3,10}

In our patient, the clinical findings were nonespecific and our pre-operative diagnosis was a giant fibroadenoma or a large cyst in the breast. The diagnosis of hydatid disease of the breast based on results of FNAC has previously been reported. FNAC may show the laminated membrane or dignostic hooklets.^{1,2,4,6} No anaphylactic or urticarial reactions have been reported as a complication of this procedure, but the risk of contamination should always be kept in mind.¹¹ FNAC usually provides the pre-operative diagnosis, although mammography, ultrasonography and serologic tests can also be done. As in the case reported here, the diagnosis is frequently delayed until the time of FNAC or surgery.^{2,11}

Mammography usually shows a circumscribed, homogenous mass with smooth margin. Differential diagnosis includes cysts, firboademomas, phylloides tumors, and rarely, circumscribed carcinomas.^{1,5} Mammography may also reveals a calcified mass simulating a calcified cyst or a calcified fibroadenoma. Vega and Ortega were the first to report the characteristics ring-shaped structures inside the hydatid cyst of the breast in an accidentaly-preformed overpenetrated view.⁵ They suggested that this finding might be the result of the presence of multiple daughter cysts inside the fluid-filled cysts. In most reported cases in the literature, hydatid cyst of the breast showed the mammographic appearance of fibroadenomas or cyst.^{1,5} In our patients mammography revealed a lobulated dens mass with ring-shaped structures inside the lesion.

The sonographic appearance of a mammary hydatid cyst is variable and depends on the age and complications of the cyst. The sonographic findings are similar to those in other organs. Hydatid cyst at earlier stages, may be unilocular, whereas older cysts are usually multilocular.¹² They may either be made daughter cysts or have a solid appearance made up of multiple septated cysts.¹² Hydatid cyst of the breast usually shows a well-defined cystic mass which may be completely anechoic or may have internal echos. Tanju reported a fluid level in the hydatid cyst of the breast.² Bijay reported three cases of primary breast hydatid disease, which presented as painless lump in the breast with nonspecific mammographic and unltrasonographic features.⁴

Indirect hemagglutination tests may help to confirm the diagnosis. The positive serum reactions might occure even in the absence of associated liver and lung involvement.^{1,3,5}

Mammography, ultrasonography and/or magnetic resonance imaging (MRI) of the contralateral breast should be done pre- and post-operatively to rule out dissemination in other breast.¹³ Abdominal ultrasonography and a plain chest radiography are mandatory to exclude liver and lung involvement.

Surgery is still the most effective therapy for hydatid disease which exists in any location.^{9,14,15} In hydatid cyst of the breast, total cystectomy is the treatment of choice. The principal objectives of the surgical treatment are to remove all parasitic elements, avoidance of spillage of the contents of the cyst, and removal of the cyst with maximum conservation of the breast tissues.^{14,15}

Fine needle aspiration of the cyst fluid and its replacement with a scolicidal agent with combination therapy with albendozole can be an effective alternative treatment to surgery.¹⁶ Treatment with albendazole for six months after the breast surgery is also recommended to decrease the recurrence rate. Antiparasitic medication before and after surgery may be helpfull to reduce the cyst size and for elimination of the parasite.

In conclusion, hydatid disease still represents an important medical and surgical problem in many regions and, despite it rareness, primary breast involvement might constitute an important differential diagnosis of breast lumps in areas endemic for the disease. Due to widespread traveling between different countries, radiologists should be aware of this rare but potentially-serious breast disease.

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