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An Unusual Case of Primary Hydatid Cyst of the Breast and Chest Wall

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Abstract

Introduction: Hydatid disease is common in developing countries but primary breast hydatid cyst is extremely rare even in endemic areas, accounting for only 0.27% of all cases. Very few case reports have been published relating to hydatid cyst of the breast, most of which were diagnosed postoperatively without complete radiological workup.

Case history: We present the case of a 58-year-old female from rural West Bengal (eastern India) who presented with three distinct cystic lesions in her right breast region with radiological features supporting the presumptive diagnosis of primary hydatid cyst of breast.

Discussion: Hydatid cysts are caused by the larval form of *Echinococcus granulosus,* and human beings are accidental intermediate hosts of this parasite. The breast may be the primary site or part of a disseminated hydatidosis. Surgical excision is the treatment of choice. Preoperative diagnosis can be made by radiological or serological investigation but neither is definitive.

Conclusion: This entity should always be considered in the differential diagnosis of slow-growing, benign cystic lesions of any organ or soft tissue region including the breast, especially in tropical countries.

Key words: *Cystic breast lump; imaging; hydatid cyst; treatment*

Introduction

Echinococcus granulosus is a parasite (tapeworm/cestode) that causes a hydatid cyst, generating an important healthcare problem. These cestodes have been reported worldwide, but the prevalence is much higher in developing countries, possibly due to low socioeconomic status, poor hygiene, lack of proper drinking water supply and sewerage. Most cases acquire the infection during their childhood but do not usually present clinical signs and symptoms until adulthood. The usual natural history of an untreated hydatid cyst is gradual enlargement in size; however, it may include calcification and death of the cyst. The disease is caused by larva following ingestion of tapeworm eggs. The echinococcal larval cysts are most commonly found in the liver (70%), lung (20%) and in other organs (10%) such as the gallbladder, pancreas, spleen, kidney, brain, thyroid and breast etc. The breast is a very rare primary site of hydatid cyst and accounts for only 0.27% of cases [1]. Very few cases of hydatid cysts of the breast have been reported in the literature. The largest series of 20 cases of hydatid cyst of the breast was reported in Tunisia [2].

Case Presentation

A 58-year-old Bengali woman presented with a 3-year history of a gradually progressive and painless lump in her right breast. She came from a remote, rural area and belonged to a farmer's family with a history of keeping sheep, cattle and chickens in their farmhouse. The family had no pet dogs, but they were in close contact with a large population of Indian pariah dogs that roamed there, like many other villages in the Indian subcontinent. She did not have any history of injury to the breast, fever or nipple discharge and there was no family history of breast cancer. On clinical examination, she was found to be anaemic with poor nutritional status, and she had three distinct cystic swellings in her right breast: one in the outer half (12cm x 7 cm); the second in the axillary tail region (5cm x 4cm)

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and the third in the infraclavicular region (7 cm x 6 cm). A fluctuation test was positive but transillumination tests were negative for all three swellings. All the swellings were free from overlying skin. There was no nipple retraction, skin thickening or axillary lymphadenopathy. Physical examination of the opposite breast was normal. First and second breast swellings mentioned above were freely mobile in all directions, but the infraclavicular swelling was not mobile. Ultrasonography(USG) of the right breast showed three separate cystic lesions with a thick wall and mobile internal echoes, thin internal septations and cartwheel appearance.

Computed tomography (CT) was performed for further evaluation which showed two rounded hypodense masses in the right breast with thin internal septations and a third lesion overlying the anterior chest wall. Peripheral enhancement was seen without internal enhancement. Serum anti-echinococcus granulosus IgG antibody(ELISA) was 22u/mL(positive). The chest x-ray and abdominal USG were normal. From the above findings, our provisional diagnosis was primary hydatid cyst of the breast. Fine needle aspiration biopsy was avoided for the fear of anaphylactic reactions. Albendazole was given for four weeks but achieved no reduction in size. Hence, surgical excision was planned under general anaesthesia.

On exploration, two cysts were excised from right breast and a third cyst was found adhered to the anterior chest wall below the pectoralis minor muscle which was also excised by splitting the muscle. Daughter cysts and serous fluid were identified upon incision of one of the masses. Histopathological examination confirmed a hydatid cyst. The patient was discharged on the seventh postoperative day without any complications. Albendazole treatment was continued for the next three months. In the last 12 months, USG of the breast, thorax and abdomen has been performed twice and showed no signs of recurrence.

Discussion

Echinococcus granulosus is the most common species of the tapeworm genus Echinococcus causing hydatid cyst, but Echinococcus multilocularis and Echinococcus. Olgarthus can also infect human beings. In India and other developing countries, livestock-rearing is the key livelihood for small and marginal farmers from the rain-fed or unirrigated agriculture causing close contact of humans with animals. Dogs, wolves and jackals are the definitive hosts and pigs, cattle and sheep are usually intermediate hosts of echinococcus and eggs of the parasite released in the stool of the primary hosts. Human beings (accidental intermediate hosts) may ingest eggs by intimate handling of infected dogs or drinking contaminated water (faecal-oral transmission). The breast or other organs can be the only primary site when the larvae bypasses the hepatic and the pulmonary filters or part of disseminated hydatidosis. Although rare, primary involvement constitutes a differential diagnosis of the breast lump, especially in endemic areas [3]. It usually presents as a slowly progressive, painless breast lump without axillary lymphadenopathy, and affects women between 30 to 50 years of age, although a wide age range (26–74 years) has been reported [4]. Clinically, this entity might simulate fibrocystic breast diseases, chronic abscesses, phyllodes tumours or even carcinoma, and when secondary infection occurs, a breast hydatid cyst cannot be distinguished from a breast abscess, either clinically or by mammography [5]. Radiological screening modalities for diagnosis of breast hydatid cyst include ultrasound, mammography, CT and magnetic resonance imaging (MRI) [6].

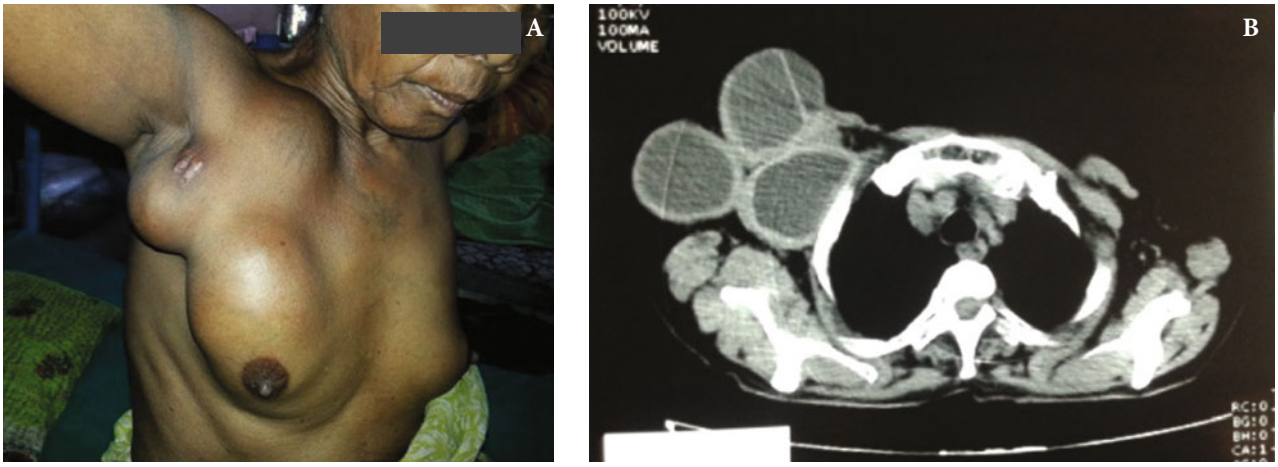

Figure 1. (a) Three distinct swellings in right breast ; axillary tail and infraclavicular region (b) CT scan showing peripheral enhancement of swellings and internal septations.

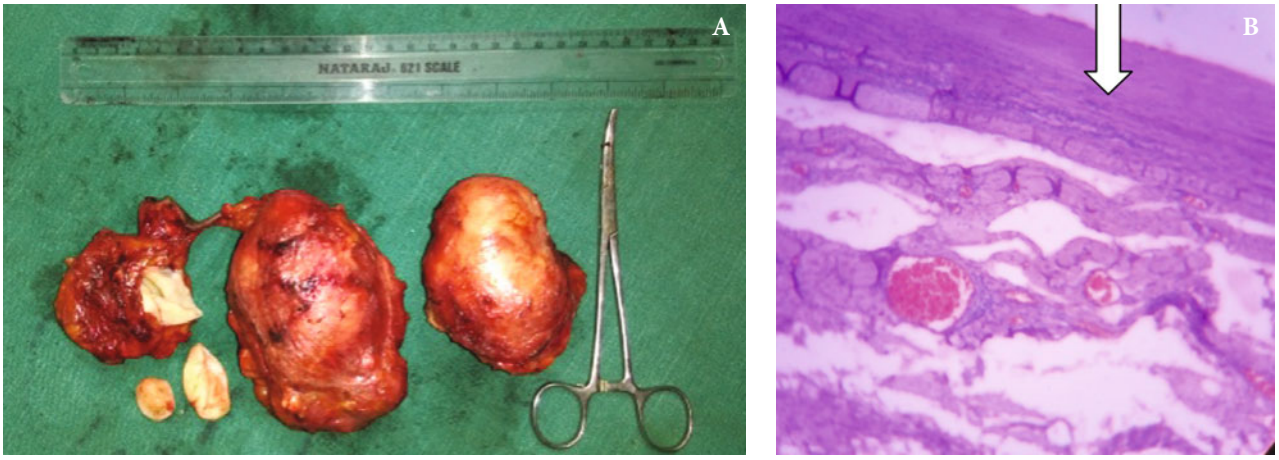

Figure 2. (a) Three separate cysts after surgical excision (b) Eosinophilic laminated membrane on histopathology(white arrow).

The ultrasonographic appearance of the cyst is variable and depends on the age of the cyst. Early cysts may be unilocular, whereas older cysts are multilocular with a heterogeneous appearance due to the presence of multiple small daughter cysts. Gharbi et al. have described five types of ultrasound findings for hydatid cysts [7]. Mammography may show a circumscribed mass; characteristic ring-shaped structures inside the mass in an over-penetrated view strongly suggest a breast hydatid cyst [8]. Computed tomography and magnetic resonance imaging are also important diagnostic tools.

Serological tests like enzyme-linked immunosorbent assay (ELISA), indirect haemagglutination (IHA), and immunoblot techniques can confirm the diagnosis of hydatid cysts after clinical and radiological suspicion of the disease [9].The diagnosis of hydatid cyst can be made by aspiration biopsy by identification of scoleces, hooklets or fragments of laminated membrane [10]. However, the aspiration procedure can cause secondary cyst development in other areas and anaphylactic reaction. Serologic tests may be used to confirm diagnosis and also in follow-up. Surgical excision is the treatment of choice for breast hydatid cysts, the principals of surgery being complete excision of cyst, avoidance of spillage of contents and maximum conservation of the affected organ. Recurrent disease has been reported in 10% of patients undergoing hydatid cyst surgery. Perioperative chemotherapy using albendazole has been shown to decrease the incidence of recurrent disease [11]. The definitive diagnosis of the lesion is made by gross and microscopic examination of the lump following excision.

Conclusion

Although rare, hydatid cyst should always be considered in the differential diagnosis of slow-growing, painless cystic breast lumps, especially in endemic areas. Complete excision of cyst is the treatment of choice. Preoperative diagnosis can be made by exercising a high index of suspicion during clinical examination and with the use of modern radiological and serological tests.

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Ethical Approval-Informed Consent

The authors declare that the study has been approved by their institutional review board. Also the patient gave her written informed consent prior to her inclusion in the study.

Conflict of Interest

The authors have no conflict of interest or competing financial interests with regards to this manuscript.

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