Mammary Echinococcosis: two Cases and Literature Review

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Introduction
Hydatid cyst (Echinococcosis) is caused by the organisms Echinococcus granulosus and is seen endemically particularly in sheep raising countries. This parasitic infection is common in Middle East and also in other parts of the world including India, Africa, South America, New Zealand, Australia, Turkey and Southern Europe.1 The disease is manifested by the presence of one or more hydatid cysts usually located in the liver (55-70%) or lung (18-35%) and in both organs simultaneously in 5-13% of the case. Other relatively common sites include muscle (4.7% cases), spleen (2.1% cases) and brain (1.4%). Bone, thyroid, breast and pancreas infections are rarely encountered.1-4 In 532 cases reported by Bickers one case each was located in the orbit, bladder, heart, chest wall, sub-cutaneous tissue, tibia, parotid, and thyroid.4 Hydatid cyst in the breast usually presents clinically as a painless slowly growing mass without regional lymph node involvement.2,5 A retrospective study in the period, 1969-1982 from Salah Aziez Institute, Tunis has reported only 20 cases in the breast.6 The breast is a rare primary site of the lesion and accounts for less than 0.27% of all cases.7-9

Case Report
Case 1
A 30 year old female presented with a history of painless mass in the upper outer quadrant of the breast. It was a slow growing lump and had grown to this size in six months. On examination it was a freely mobile soft mass measuring roughly 6.2 x 4.9 cm in size. No regional lymph nodes were palpable. The specimen received for histopathology showed a cystic piece of tissue, pearly white in colour and soft in consistency with attached breast tissue flap. Microscopic examination showed a cyst wall composed of lamellated basophilic chitinous material. No scolices were identified, adjacent breast tissue showed extensive secretory change. It was diagnosed as a case of hydatid cyst of breast.

Case 2
The other case was that of a 32 year old female who presented with a small cystic swelling in the breast. It was a painless asymptomatic mass without any axillary lymph node involvement. Clinically there was no suspicion of malignancy. We received a gray white fibrofatty piece of tissue measuring 4 x 3.5 x 2 cms in size. On sectioning a cystic cavity was identified measuring 1 x 1 cm in size. Microscopy showed benign breast tissue along with cyst wall composed of amorphous chitinous material. Several brood capsules containing scolices were also identified. It was also diagnosed as Hydatid cyst. Fine Needle Aspiration Cytology was not performed in both cases.

Discussion
Echinococcosis or hydatid disease, is possibly one of the more difficult parasitic diseases to understand because of the peculiar cystic lesions that form when the larval stages of the parasite invade the viscera. Humans serve as accidental hosts. The normal life cycle of E. granulosa involves dogs and foxes as the definitive hosts, within the intestines of which the adult tapeworm reside. Sheep, cattle, or swine serve as the intermediate hosts and develop cystic larval disease.10,11 Hydatid cyst can occur in any organ of the body although breast is an uncommon site but a high suspicion of this disease is justified in endemic regions in cases of asymptomatic painless breast lumps without lymph node involvement. Several cases have been reported in the literature, many from developing countries where the disease is endemic. For example cases of mammary echinococcosis have been reported from Turkey3,8 and India10, a case has also been reported from Pakistan.9 However, the cases are not confined to developing countries and multiple case reports from developed countries have also been published.2,6 The disease can present as a solid or cystic mass in the breast. The solidity is conferred by the fibrous capsule filled with grayish material containing membrane debris. Often a cyst and lump are present together. Primary mammography forms an essential part of pre-treatment evaluation since it enables circumscription of the lesion and study of the regional lymph
nodes, which are normal in all cases. Women 20-50 years of age are said to be affected as in our cases both women were in the third decade and of childbearing age. Although rare, several cases are reported to have been diagnosed cytologically by the detection of hooklets in the pleural fluid or sputum. Diagnosing hydatidosis through cytologic examination of aspirated fluid is hazardous, as it can cause anaphylactic reaction due to fluid leak.3 To date 54 cases of hydatid cyst of the breast have been reported and only one was diagnosed through aspiration cytology.5 Therefore, although hydatid cyst in breast is a rare lesion, there is enough evidence in literature to justify the need to keep this disease in mind when dealing with mass lesions of breast in the setting of developing countries where the disease is endemic. Surgery remains the main treatment in hydatidosis with well founded criteriae and approach in the most frequent locations of the disease: liver11 and lung.12 Surgical planning and techniques are dependent on the number of cysts, the anatomical relations and anatomical changes produced by the parasite growth. Surgical approach and technique depends on correct diagnosis and if it has been made before or during operation. The use of antiparasitic medication or solutions during operation varies according to hospitals and medical tradition - hypertonic saline, ether, formic aldehyde, hydrogen peroxide, cetrimide, rivanol and alcohol are commonly used agents.13 Chemotherapeutic agents without surgery have demonstrated reduction in the cysts size and occasional elimination of the parasite but not demonstrable benefit has been described so far in large homogeneous series and no scientific conclusions can be drawn.14,15 Their mechanism of action is known to be through a blockade of the glucose intake and glycogen deprivation of the parasite with growth retardation and even sterilization of the content. Antiparasitic agents can be used prior to surgery as a safeguard measure and after surgery to prevent further implants and secondary hydatid seeding but not as a sole therapeutic purpose.

References