

Case Report

Hydatid cysts at unusual sites: Reports of two cases in the neck and breast

Sajjad Ahmad, Shahela Jalil, Yahya Saleem, Bilqees A Suleman, Nasir Chughtai
Department of Histopathology, Sheikh Zayed Hospital, Lahore.

Abstract

Hydatid cysts in the neck and breast are rare and are incidental finding in neck and breast specimens. It is a zoonotic disease which is due to infectivity with larval stage of *Echinococcus granulosus*. The disease is chronic and cysts can be lodged in different organs. It has cosmopolitan distribution and impact health and economical challenges for many countries throughout the world. Therefore, accurate information on the distribution of the disease is first step for the control and prevention. We present two unusual cases of hydatid cyst found in the posterior cervical region and in the breast.

Introduction

Echinococcus granulosus is a cestode parasite (tapeworm) that causes hydatid infection. These cestodes have a worldwide distribution but the prevalence is higher in developing countries. Infection rate is as low as 1 per 1000 in North America and as high as 10% in the third world. This disease is endemic in cattle and sheep rearing regions of the world. The close association of people with sheep and dogs and the unavailability of clean potable water supplies in Pakistan make it a region endemic to the disease.¹ Majority of the cases of hydatid disease come from rural areas or people

who have settled in urban centers after spending life in villages. Most of the people acquire the disease during their childhood but do not present with the clinical signs and symptoms until late adulthood. The natural progression of an untreated cyst may include calcification and death of the cyst, however more frequently the cyst gradually enlarges.² The disease is caused by larva following ingestion of the tapeworm eggs. The definitive hosts are dogs while sheep are unusual intermediate hosts and humans are accidental intermediate hosts. The disease is manifested by the presence of one or more hydatid cysts usually located in the liver or lung. The cysts (2 to 30 cm) are constituted by an external acellular cuticle and an inner cellular "germinal" layer (10-25 μ) that produces the brood capsules. The larvae (scolices) develop from the germinal layer.³ Involvement of sites other than the liver and lung is rare but no site is immune. Bones may be involved in 0.5%-4% of the cases. Generally vascularized bones like vertebrae, long bones and epiphysis are infected. Renal involvement is seen in 2-3% of the cases. Hydatid disease of the brain constitutes 2% of all intra-cranial space-occupying lesions in endemic regions. Cardiac hydatid disease occurs in 0.02%-2.0% of cases. Other sites that may be involved include soft-tissues, ovaries, pancreas, scrotum, inguinal canal and chest wall but neck and breast are unusual sites of distribution.¹

Hydatid disease in such uncommon sites may cause a significant problem in diagnosis.

Case-1:

A 46-year-old man presented in the surgical outdoor with a slightly tender swelling in posterior cervical region of 5 months duration. The size was increasing gradually. The ultrasound report was of suspected complicated/ hydatid cyst. However his IgG serology for hydatid disease was negative. It was followed by ultrasound guided FNAC which revealed fibrofatty stromal fragments with many scattered scolices, some contained hooklets. Many free hooklets and spherules of calcified rounded bodies were also present. This was reported as a case of hydatid cyst, which was confirmed on clot histology. Patient was put on Albendazole. He did not return for a follow-up visit.

Case-2:

A 21-year-old girl presented with recurrent cystic swelling of left breast with a history of cystectomy three times in the past with non specific histological reports. She was also medically treated for hydatid disease in liver during childhood. On examination she had 3 lumps in her left breast (two lumps in upper inner quadrant, one supraalveolar 12x8cm, other infraalveolar 6x6cm. One lump 6x6cm was in outer lower quadrant), with a surgical scar. The bone imaging revealed evidence of skeletal involvement (thoracic vertebrae). Previous FNAC of two lumps were reported benign inflammatory lesions of breast and revealed macrophages, benign ductal epithelial and myoepithelial cells with few lymphocytes. No hooklets and scolices were identified on FNAC. However fluid aspirated during surgery sent for cytology revealed few scattered hooklets and was reported as suspicion of hydatid disease. This was followed by excision of the multiple cysts with daughter cysts along with pseudocapsule. Histological examination revealed cyst wall composed of eosinophilic chitinous material focally lined by germinal epithelium. Few brood capsules, scolices with hooklets were identified and histology confirmed the diagnosis of hydatid disease.

Discussion

Hydatid cyst at unusual sites have been reported around the world. Hydatid disease in the neck is quite rare, even in areas where the disease is endemic and is still common in the countries including the Mediterranean countries, the Middle East, South America, New Zealand, Australia, and South Asia. The liver is the most frequently involved organ (75%), followed by the lung (15%) and the remainder of the body (10%). The diagnosis of Echinococcus granulosis in an atypical location can be difficult. Histopathological evaluation of the excised specimen and fine needle aspiration cytology

usually leads to the diagnosis. Since puncture of the cyst may lead to an anaphylactic reaction due to spillage of hydatid fluid, the use of fine needle aspiration is controversial at present. Katilmis H⁴ and Beji M⁵ et al reported a case of a 33-year-old female patient presenting with a hydatid cyst in the lateral cervical region. The diagnosis of the hydatid cyst was made mainly with the help of imaging methods and review of the patient's history. In our case report 1 the suspected diagnosis of hydatid cyst was also made by ultrasound.

Aijaz Ahmed Memon⁶ reported a rare case of hydatid cyst (Echinococcus Granulosus) in the submandibular gland of a young man. It was during the operative procedure that the hydatid cyst was found in the submandibular gland and just like our case report 1 this was also confirmed on histology.

Pampori AR⁷ reported a case of a 15-year-old female with slowly progressing non tender swelling in neck. At surgery a typical hydatid cyst was found and diagnosis was confirmed. IgG serology was also reported positive. Eroglu A⁸ presented an unusual case of a hydatid cyst found in the nape of a 66-year-old Turkish woman. There was no pulmonary or hepatic involvement.

Ouedraogo EG⁹ reported 20 retrospective cases observed at Salah Azaiez Institute of Tunis from 1969 to 1982, in women of 30 to 50 years of age living in areas where hydatid disease is endemic.

Hydatid cyst of breast is a benign lesion due to larval stage of Echinococcus Granulosus. As they evolve slowly, clinical features are easily confused with the diagnosis of benign growths. Das DK¹⁰ et al reported the case of 27 years old female presenting with a painless lump of the left breast. Fine needle aspiration cytology was inconclusive. Lumpectomy was done. Histopathological examination confirmed it to be a case of hydatid disease involving the left breast. . Surgical treatment is curative in all cases. Uncu H¹¹ reported the case of a hydatid cyst in the breast, which is a rare primary site for the cyst and a similar case was reported from Pakistan by Kazi.¹² In our case 2, the FNAC reported few scattered hooklets suspicious of hydatid disease. This was finally diagnosed on histology.

Although the diagnosis is confirmed by cytology or histology, imaging modalities such as ultrasonography, CT and MRI are more sensitive than serologic tests and in these cases, the involvement of other organs should be investigated. The cyst should be excised as a whole without being ruptured to prevent any complications.

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