Case report:

Subcutaneous hydatid cyst in forearm – a diagnostic dilemma with neurofibroma Datta S³, Banerjee A², Banerjee U³

Abstract

Hydatid disease caused by larval stage of Echinococcus granulosus usually affects lungs and liver of human and animals. Affection of subcutaneous tissue is rare; though not rarest but often clinically mistaken as neurofibroma or lipoma. Preoperative diagnosis is mandatory to avoid surgical contamination or future accidents. Two middle aged males presented with forearm nodules and one female presented with an arm nodule; all of which on needle aspiration cytology and wet smear examination provisionally diagnosed as hydatid cyst, later confirmed radiologically and surgically. Primary hydatid disease constitutes potentially serious differential diagnoses in subcutaneous swelling.

Keywords: Hydatid cyst; Subcutaneous tissue; Fine needle aspiration cytology.

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Introduction

Hydatid disease is a parasitic disease of human and other mammals caused by larval form of Echinococcus genus¹. Infection can present in three forms - unilocular form caused by Echinococcus multilocular granulosus; form caused by Echinococcus multilocularis; polycystic form caused by Echinococcus Vogeli¹. Human get infected following accidental ingestion of eggs from environmental sources. Humans are accidental intermediate hosts of such organism. The cyst often manifested as slowly growing mass commonly involving liver (75%), lungs (15%) and can involve any part of body except hair, teeth and nails. Isolated involvement of soft tissues is relatively rare^{1,2}. Here, in the three cases, the patients presented with solitary hydatid cyst in subcutaneous tissue of forearm and arm without affecting any other organ.

Report of cases: Two males aged forty years and thirty eight years and one female aged forty three years in separate occasions came in the pathology department with subcutaneous swelling in right forearms and left arm which were mildly painful; clinically diagnosed as neurofibroma or lipoma.

All of the swellings were soft, partially mobile with smooth margins, non-tender. Fine needle aspiration showed similar features in all the three cases. Aspiration yielded hazy pale whitish fluid which on wet smear showed scolices and centrifuged smears fixed with 95% ethyl alcohol and Papanicolau stained smears showed hyalinised membrane like layers with polychromatic oval, spherical structures with eosinophilic granules identified as scolices of Echinococcus species.

Ultrasonography of the affected region revealed a thick walled cystic mass with linear septations; likely to be a hydatid cyst. The indirect hemagglutination test (IHA) showed positivity (1/1024) in two of the cases as it was not done in the younger male. Other routine haematological tests were unremarkable. Liver and lungs were without any significant findings radiologically. The masses got surgically removed under local anesthesia with wide excision of surrounding fibrous tissue avoiding contamination.

Macroscopically, the masses had an appearance of cystic structure filled with hazy fluid and measured 2 cm, 2.2cm and 1.8 cm in diameter respectively (Figure 1).

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Figure 1: Gross appearance of excised soft tissue mass.

Microscopically, the masses showed cyst wall having thick outer laminated, acellular layer with inner thin germinal epithelium (Figure 2).

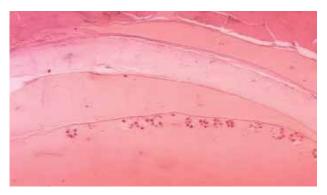


Figure 2: Microscopy of hydatid cyst (low power view) showing laminated membrane of cyst wall.

The cysts contained numerous broad capsules and scolices. It was diagnosed as subcutaneous hydatid cyst of forearm and arm. No localised or systemic recurrences were observed in one and half years follow up of the male patients and one year in case of the female patient.

Discussion

Subcutaneous hydatid cyst can be primary or secondary^{1,2,3}. The ingested parasite ova enters portal system penetrating intestinal wall and get entrapped in hepatic sinusoids; some reach lung and systemic circulation passing through liver; thus causing hydatid disease in other organs accounting for solitary cysts in uncommon sites ^{2,3}. The present patients are primary cases since they had no previous history of hydatid cyst affection or any surgical intervention. The clinical course is non-specific which depends on site, size and pressure imposed by

cyst and can resemble abscess or carcinoma if there is superimposed infection.

Radiology plays a vital role in diagnosing hydatid cyst with respect to cyst wall, daughter cyst, calcification, inner germinal membrane separate from outer wall which are specific findings and it also render knowledge of its relationship to adjacent organs^{2,3,4}. Serology with fine needle aspiration cytology (FNAC) also play eminent role in diagnosing the lesion^{3,4}. However, serology can be negative in extrahepatic and extrapulmonary lesions; with false negative and false positive results. FNAC is of immense help as it shows presence of scolices and hyalinised laminated membrane microscopically which aids in diagnosis of hydatid cyst^{3,4}.

Prousalidis et al reported 9% of extrapulmonary and extrahepatic hydatid cyst cases in a large series of patients⁵. Chevalier et al reported 2% of subcutaneous hydatid cyst cases in his study with presence of cyst in other organs also. Reports of primary subcutaneous hydatid cysts are very rare⁶. Dirican et al failed to find a single case of palmar subcutaneous hydatid cyst in reviewof literature but reported two subcutaneous hydatid cysts which were treated surgically and the cysts were primary as the patients did not have any history of surgical intervention and were devoid of any other affected organ⁷.

Diagnosing a hydatid cyst preoperatively is vital as during surgery exposure of tissue to cyst contents may lead to anaphylactic reaction and local recurrence^{1,7}. The indirect hemagglutination test is positive in more than 80% of hepatic cases. A positive hemagglutination test is significant but negative result does not indicate absence of disease^{2,6}. Presence of daughter cysts and absence of capsular calcification, detachment of membranes, and irregular edges suggest a viable cyst. The best treatment option is total surgical excision without opening the cyst. If the cyst cannot be excised without opening, the cyst contents have to be removed, laminated membrane should be totally excised and cyst irrigated with protoscolicidal solutions^{8,9}.

Other options include percutaneous treatment under ultrasound guidance with needle aspiration of irrigation of scolicidal solutions as well as medical treatment with albendazole¹⁰. Akhan et al reported percutaneous treatment by either a catheterisation technique with hypertonic saline and alcohol or a modified catheterisation technique according to type of cyst and an average of 96.1% of volume reduction obtained in six cysts of four patients. Medical

treatment helped in resolution of cavity infections and cellulitis which appeared as complications¹¹. In conclusion, the provisional diagnosis of hydatid cyst is immensely important to avoid surgical contamination during intraoperative handling of tissue and prevent future occurrence of accidents and

these cases are an addition to literature.

Conflict of interest: None

Ethical approval: was taken from Ethics Committee of Burdawn Medical College about publication of this case report.

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