

Case Report

ENDOBRONCHIAL RUPTURE OF PULMONARY HYDATID CYST FOLLOWED BY ANAPHYLACTIC SHOCK AND FULL RECOVERY

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Abstract: A 36 years old woman with a past history of multiple hydatid cysts in liver presented with dry cough and low grade fever. She was found to have a large hydatid cyst in the left lower lobe of lung which ruptured endobronchially. At the time of induction of anesthesia, the cyst ruptured and she went into anaphylactic shock and then cardiac arrest. She was successfully resuscitated and later on a partial pneumonectomy was done. She made an uneventful recovery. Although hydatid cyst is a globally prevalent disease and has a low mortality rate but a ruptured cyst has a relatively high mortality and morbidity in terms of dissemination of disease into other parts of body therefore a careful follow-up is required in case of rupture of the cyst.

Key words: Hydatid cyst, E. granulosus, endobronchial rupture, anaphylactic shock.

Introduction

Hydatid cyst disease is an ancient disease and these bladder like cysts were described in animals in Babylonian Talmud, by Hippocrates in 4th century B.C., Aratacus in 1st century A.D. and Galen in 2nd century A.D.^{1,2}, but Francesco Redii was the first to describe the parasitic nature of these cysts and Pierre Simon Pallas in 1766 hypothesized that these cysts are larval stages of the tape worms³. In 1853, Carl von Siebold demonstrated the development of adult tapeworm in dogs fed with cysts from the sheep⁴ and human infection by infected dogs was demonstrated in 1863 by Bernhard Naunyn⁵. The attempts to remove these cysts surgically were also made in 16th century which has been the most effective treatment so far. Cystic echinococcus is an emerging zoonotic infection with worldwide distribution and estimated human burden of disease is 285, 407 (95% CI).⁶, and still on the rise especially in the countries previously thought to be free of disease.⁷ The recorded data may be an underestimation of the actual disease burden as it remains clinically silent for many years.

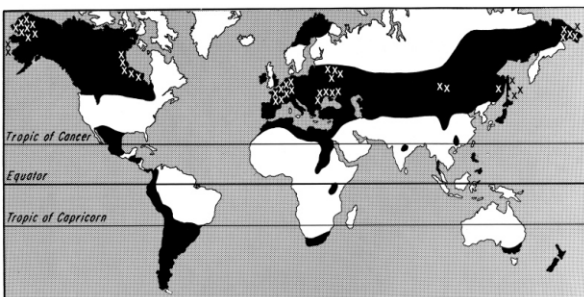


Figure-1: Global distribution of E. granulosus (black) & E. multilocularis (x).

The greatest prevalence of cystic echinococcus is found in temperate zones including south America, the entire Mediterranean littoral, southern and central parts of former Soviet union, central Asia, China, Australia and parts of Africa, mainly the cattle raising countries^{8,9}. (Fig.1)

Case

A 36 years old woman of Afghan origin presented with persistent dry cough and low grade fever for last three months. There was no other complaint. In past she had multiloculated hydatid cysts (fig. 2) in liver for which she was operated in Afghanistan three years ago. She was married, housewife with two healthy children. Her family history was strongly positive for hydatid disease and her mother, brother and sister-in-law had suffered from this disease in past. They had a



Figure-2: CT abdomen showing multiloculated hydatid cyst in the right lobe of liver.



Figure-3: chest radiograph showing a round opacity in left lower lung (hydatid cyst before



Figure-4: CT chest; a large fluid filled cavity in left

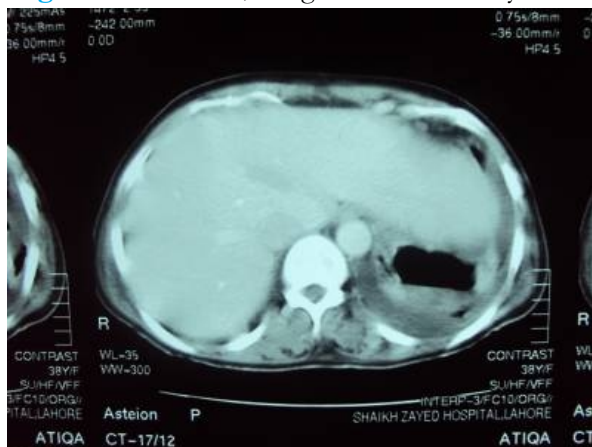


Figure-5: CT abdomen showing ruptured hydatid



Figure-6: Chest radiograph showing a large fluid filled cavity in left lower lung (ruptured hydatid cyst)

pet dog at home. She was investigated in Afghanistan, was found to have a large fluid filled cyst in lower lobe of left lung (**fig-3 & 4**) and was brought to Pakistan for further treatment. She was admitted to the hospital, baseline investigations were done which showed a hemoglobin of 11.2 g/L, white cell count of $14.56 \times 10^9 /L$ and platelet count of $281 \times 10^9 /L$. Her coagulation profile, RFT's, LFT's and serum electrolytes were all normal except for a low serum albumin of 2.7 g/dL. She was also tested for E. granulosus antibodies which came out to be highly positive 1:2038. On second day of her admission she started expectorating as well as nasally regurgitating yellow coloured saltish fluid. Endobronchial leakage of cyst fluid (figs. 5 and 6) was suspected and she was prepared for surgery and put on IV antibiotics and oral antihelminthics. At the time of induction of general anesthesia the cyst partially ruptured into the lung parenchyma and she went into anaphylactic shock, respiratory distress syndrome (fig. 7) and cardiac arrest. CPR was started and she was resuscitated successfully after 5 minutes. Later on after stabilizing the patient, left sided pneumonectomy (fig. 8) was done and she made a

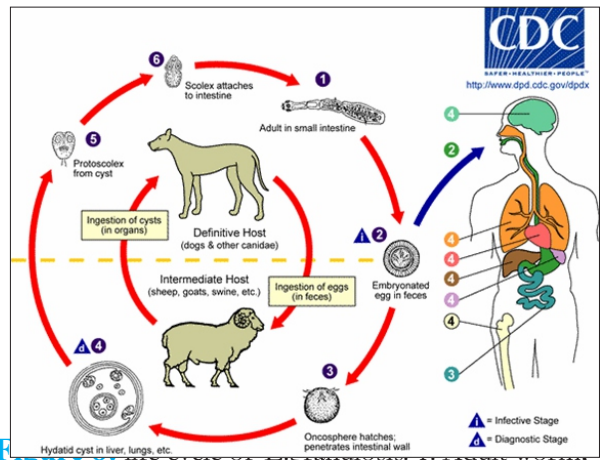


Figure 7: Chest radiograph after development of ARDS.



Figure 8: Chest radiograph after left sided partial pneumonectomy

be followed up on outpatient basis. She attended the outpatient department after a couple of weeks and was found to be in an excellent health without any chest symptoms.



2. Eggs; 3. Oncosphere; 4. Hydatid cyst with daughter cyst; 5. Protoscolex; 6. Scolex.

Discussion

Echinococcus is a parasitic cestode of phylum platyhelminthes commonly known as tapeworms. Three forms occurring in humans are
 E. granulosus (cystic hydatid disease)
 E. multilocularis (alveolar hydatid disease)
 E. vogeli and E. oligarthrus (polycystic hydatid disease)

The adult parasite is a small tapeworm about 3-6mm long that lives in the small intestine of definite hosts like dogs or other canids, where it lays eggs which are passed in their feces, and ingested by intermediate hosts like sheep, goat swine etc. The eggs hatch in their small bowel and the oncosphere migrates into different organs by penetrating the gut wall and through the circulatory system where they develop into cysts which gradually enlarge and produce proctoscolices and daughter cysts which fill the cyst interior. Again when the dogs and other canids feed on the viscera of these animals they become infected and the life cycle goes on. Humans get infected accidentally when they come into contact with the fecal matter of definite hosts and ingest it. Molecular studies using mitochondrial DNA sequence have identified ten distinct genotypes (G1-G10) within E. granulosus with different host affinities; the sheep strain G1 being the most common¹⁰.

In spite of several WHO/FAO sponsored meetings on cystercerosis / hydatidosis the disease is still on the rise in countries previously free of it and also no increase in the public awareness of the global nature of disease has been seen^{8,9}. It is evident from the life cycle of echinococcus that disease is preventable at various stages in both the definite and intermediate host. Important measures to break the chain of transmission include:

- İ Prevention of dog feeding on carcasses of infected sheep
- İ Stray dog population control
- İ Restrict home slaughter of livestock
- İ Prevention of food and water contamination by fecal matter from dogs
- İ Hand washing after dog handling and before eating food
- İ Vaccination of sheep by EG95 has a cure rate of 86% and 99.3% reduction in the number of cysts. Also vaccination of dogs leads to 97-100 % protection against worm growth and egg production¹¹. Studies also show that most effective intervention against the *Echinococcus* is combination of sheep and dog antihelminthic treatment¹².
- İ The role of better diagnostic tools e.g. dog coproantigen detection has also been elaborated.
- İ Recent treatment modalities include:
- İ Albendazole alone for three months or longer followed by rest and repeated cycles if required.

- İ PAIR (in case of inoperable cysts) which consist of percutaneous aspiration, injection of scolicidal agent (e.g. 95% ethanol or 0.5% cetrimide) and reaspiration.
- İ Alveolar hydatid cyst disease should be treated with wide surgical resection in conjunction with oral albendazole.
- İ Other drug options include mebendazole and praziquantel.

The use of better diagnostic tools, increasing public awareness regarding health and hygiene and new anti parasitic vaccines hold a potential to speed up the eradication of hydatid cyst disease¹³.

It is important to realize the gravity of the condition and its potential complications including death. Prompt detection of anaphylactic shock and its management with intensive care facilities can save life as shown by our case.

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