# DOI: http://dx.doi.org/10.5915/13-2-11931 HUMAN ECHINOCOCCOSIS IN SAUDI ARABIA\*

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## SUMMARY

Human echinococcosis is not an uncommon disease in Saudi Arabia and some other Middle East countries due to the presence of sheep, dogs and humans living in close contact. Although it is supposed to be a benign disease, the onset of complications which are liable to occur makes its morbidity not much inferior to malignant disease. From the data collected from the main hospitals in various districts of Saudi Arabia and from their own experience, the authors tried to evaluate the various epidemiological and nosological aspects of the disease. Its incidence, salient clinical features, complications and methods of diagnosis are presented. The effectiveness and safety of some scolicidal materials and the various surgical techniques used in dealing with the hydatid cyst cavity left behind are discussed.

#### INTRODUCTION

Human echinococcosis is a very ancient human disease being known since the days of Hippocrates over two thousand years ago when he described the "liver filled with water...etc."<sup>10</sup> Al-Rhazes, the Arabic physician made reference to hydatid cyst of the liver in 900 A.D.<sup>1</sup>. The nature of the lesion was unknown and was considered to be growths or tumours until Tyson<sup>25</sup> in 1687 gave the first suggestion of its parasitic nature. However, the exact life cycle of the echinococcus granulosus worm and the method of human infection was only established by about the middle of the nineteenth century. Thanks to the efforts of Dew 1928<sup>23</sup> and other workers in this field, the detailed clinical aspects of the disease had become completely clarified by the beginning of the twentieth century.

Infestation by the disease is not uncommon in Saudi Arabia because of the presence, in close contact, of the carnivorous definitive host, the dog, and the herbivorous intermediate host for the parasite, the sheep. Eggs excreted in the dog's feces, contaminate plants or water, and infection of the intermediate host occurs by ingestion of the contaminated food by sheep, cattle or man. In the small intestine, the embryos liberated from eggs, penetrate the intestine and enter mainly into the portal circulation, very rarely into the lymphatic system. Most of them become trapped in the first filter, the liver, while the remainder pass through the hepatic veins to the lungs which act as the second filter where a good proportion of them are arrested. The minority of the embryos pass through this second filter into the general circulation where they can settle in any other tissue. Some of these embryos are destroyed by the body phagocytes while the remainder grow to form the hydatid cysts.

The extent of hydatid disease has not before been evaluated in Saudi Arabia in detail. The aim of this work is to assess the prevalence of the disease in the Kingdom, its various nosological aspects and the methods used for its diagnosis and treatment.

### MATERIALS

The 188 patients included in ths study are only those who were operated upon in various main hospitals in different districts of Saudi Arabia, i.e., Riyadh, Taif, Jeddah, Khamees Mushate Hospitals during the past five years. We did collect more than 230 cases from different hospitals in the five districts of Saudi Arabia but detailed information was available for only 188 patients. Patients with hydatid disease accounted for 0.3% of all general surgical operations and 5% of the major general surgical operations.

The age of the patients varied between 6 and 80 years the maximum age incidence being between 25 and 45 years (Table 1). The disease was rare below 20 years of age. One hundred twenty-two of the patients were females (62.76%) and 66 of the patients were males (37.24%). The highest incidence was found in the patients of the South Western Region followed by the Central Region, and most of the patients operated upon in districts studied originally came from the south western region. A few cases came from other Middle East countries like Yemen, Egypt, Syria, Iraq and Palestine. Three patients in the series gave a history of previous operations for hydatid disease.

#### Table 1:

Age Distribution of Hydatid Disease

Age	No. of Cases
1-10	8
[]-20	29
21-30	36
31-40	62
41-50	39
51-60	6
61-70	7
71-80	1

The clinical presentation of hepatic hydatid disease varied but the most common was mild right hypochondric or epigastric pain or discomfort (96%) followed by an upper abdominal mass (92%). A few patients complained of dyspepsia, anorexia, occasional vomiting or pain referred to the right shoulder. Fever, guarding and tenderness was encountered only in infected cases. Cough, chest pain and expectoration were the dominant findings in pulmonary hydatids. A few cases were silent, the disease being discovered on routine investigations for unrelated conditions.

Unusual presentations were obstructive jaundice (2 cases from rupture of the cyst into the biliary tree), generalized peritonitis (one case, from intra-peritoncal rupture of the cyst) and retention of urine (one case, from compression of the cyst on the bladder neck).

The most common organ affected was the liver (82.45%) followed by the lungs (29.8%) (Table 2). The right lobe of the liver was far more commonly affected (96% of hepatic hydatids). The sex distribution in lung infection was nearly equal but hepatic affection was more common in females (78 females: 37 males).

#### Table 2: Distribution of Hydatid Cysts in Different Organs and Tissues

	No. of Cases
Liver alone	95
Liver, peritoneum and/or	
intraperitoneal organs	11
Liver and lung	9
Lung alone	47
Other tissues*	26
Total	188

\*These cases include peritoneum and mesentry 6 cases; spleen 3 cases; subcutaneous tissues 3 cases; kidney 2 cases; bone 2 cases; breast 2 cases; gall bladder one case; eye (retro-bulbar tissues) one case; ovary one case; pleura one case; submandibular salivary gland one case; brain one case; urinary bladder one case; uterus one case.

The complications met with in the collected cases were infection (5 cases), calcification (3 cases), intraperitoneal rupture (one case), intrabiliary rupture (2 cases), intrathoracic rupture (two cases) and retenton of urine (one case).

Associated gall bladder disease was present in 18 patients with hepatic hydatids.

The result of the Casoni test varied from one hospital to another ranging between 50% and 80% positive in the patients with operative confirmation (the mean was 75% for hepatic hydatids and 60% for pulmonary ones).

Plain X-Ray revealed a soft tissue shadow or elevation of right dome of the diaphragm, hepatomegaly and/or calcification in all cases of liver hydatids.

Liver isotope scanning and ultrasonography done in some cases revealed silent or cystic lesion and gave an idea about the site and number of cysts in these cases.

The number of cysts in an individual case was variable, the most common being a solitary cyst (87.5%). Multiple cysts from more than one to hundreds were found in 12.5%.

In most of the collected cases formalin in strength of 10% for the cyst and 2% for the abdominal packs isolating the surgical field during operation, was used. Due to the occurrence of some fatalities (7 cases) formalin was replaced in the latter part of the series by sterile hypertonic saline (20%). No danger was encountered with this material.

## SURGICAL PROCEDURE

One of the following surgical procedures was used for hepatic hydatid cysts:

1. Excision in toto including the pericyst (pedunculated cysts).

2. Excision of the endocyst and any contained daughter cysts leaving the adherent pericyst (adventitial layer) which is usually deroofed with a continuous haemostatic suture being applied to its edge.

3. Partial hepatectomy or lobectomy including the cyst.

All the above procedures were followed by closed drainage either of the cavity or the operative field or both. Partial filling of the residual cavity (in "2" above) by the greater omentum was done, when accessible, to enhance filling of the dead space left. Leaving the pericyst without deroofing and closing it with interrupted stitches with external closed drainage was tried in some cases but it was abandoned because the cavity took a long time to fill during which time the drain must be left in. This may entail about 6 weeks postoperative stay in the hospital, sometimes more. No complications resulted following excision plus deroofing of the pericyst.

The postoperative complications in the operated patients included mild superficial wound infection (5%), prolonged biliary discharge ceasing spontaneously (10%), burst abdomen (2%). Death in the first 48 hours after operation occurred in about 5% of whole series. From the available details the cause of death in such patients was attributed to anaphylaxis and/or toxic effect of formalin, although no postmortem examinations were done. The hospital stay for the patients varied from 9 to 42 days, the average being about 14 days.

The long hospital stay in some patients was partially due to the lengthy investigation and preparation of the patient for surgery as well as the occurrence of postoperative complications in other cases.

#### DISCUSSION

From the data available in this series it is evident that hydatid disease is still a major health problem in Saudi Arabia in spite of recent advances and modern equipment now available for diagnosis and treatment. The disease is not only common but still carries a significant morbidity and mortality. Because human echinococcosis is mainly a surgical disease, all surgeons must be aware of its various clinical presentations, complications, proper methods of diagnosis and treatment. During its early stages the disease is silent, the reason for it being rarely encountered below the age of twenty years.

Due to the complexity and variability of presentation of human echinococcosis no characteristic clinical pattern can be described for it<sup>12</sup>,<sup>18</sup>,<sup>19</sup>,<sup>26</sup>. However, any case of hepatomegaly, jaundice, abdominal mass, chest trouble or soft tissue shadow on X-Ray in Saudi Arabia one must think of the possibility of hydatid disease. Needling of the liver or any other abdominal mass should be prohibited if there is any chance of hydatid cyst, as this can cause fatal anaphylaxis and dissemination of the disease in the peritoneal cavity. A high index of suspicion of the disease improved the incidence of correct diagnosis. In spite of this, some cases were diagnosed only after exploration.

The predominance of the disease in women in our series may be attributed to the fact that women are more commonly used in the work of sheep rearing in Saudi Arabia.

The significant difference in the incidence of the disease between various regions of Saudi Arabia may be attributed to more than one factor. First, in some regions the majority of the population is urban and rarely come in contact with sheep and dogs. Second, in the east area although sheep rearing is common, the inhabitants are not accustomed to having dogs with their sheep. This may throw light on an important point in the control of the disease, because elimination of the dogs will minimize the incidence of the disease. Also control of the disease will entail screening and treatment of sheep, cattle, goats and camels. Proper collection and disposal of the offal from slaughter houses and preventing its consumption by dogs is essential to stop the life cycle of the parasite. In one study done in Kuwaitº it was found that 10.4% of sheep imported from Saudi Arabia, mainly from the eastern district were infected with hydatid cysts, even so human disease in this region is rare, explained at least in part, by the rarity of dogs in this district.

The relative frequency of pulmonary hydatid disease in Saudi Arabia (29.8% of the series) when compared with some reports from abroad<sup>14</sup>,<sup>23</sup>,<sup>24</sup> makes one think of the possibility of air-borne infection due to the winds and dusty atmosphere which are relatively common in Saudi Arabia. The lungs in such cases act as the first filter instead of the liver.

The diagnostic accuracy of the Casoni test is considered to be about 90% by some authors<sup>2,14,27</sup>. In the present series its diagnostic accuracy varied between 65% and 80% with mean of 75%. This difference may be caused by the hydatid antigen losing its potency on storage. The false negatives were about 25%. The incidence of Casoni false positive was difficult to assess in the whole series, but in a group of cases it was about 4% which is lower than 15% reported in other series<sup>2</sup>.

The use of more recent sensitive sero-diagnostic tests11,15,21 like complement fixation (C.F.T.). indirect haemagglutination (IHA), Latex test(LT) and the "arc 5" immunoelectrophoresis test (IEP), for investigation of suspected cases will improve the diagnostic accuracy up to 95%. Also the use of these tests, especially IEP which does not give false positive results<sup>21</sup>, for screening of the population in the endemic areas will give a correct idea about the incidence of the disease and may lead to early detection of affected individuals. CFT had a special value in assessing the results of surgical treatment of hydatid cysts, because the titres decline rapidly after excision. Persisting high titres after operation may mean a residual cyst or the development of secondary echinococcosis<sup>11</sup>,<sup>21</sup>.

The use of nontraumatic investigations like isotope scanning and ultrasonography in suspected cases of hydatid disease will definitely improve the diagnostic accuracy and will give a good idea about the number, size and location of the cysts. The use of endoscopic retrograde cholangiography<sup>5</sup> seems to be of little value in the diagnosis or treatment of hydatid disease.

The operative mortality in the present series (5%) is comparable to the figures reported by other workers<sup>2,20</sup>. The replacement of formalin which proved to be toxic, by hypertonic saline (20%) which proved to be effective and safe in some of our cases and in other series,  $7, 2^2, 2^3, 2^4$  may further lower the operative and postoperative mortality.

Proper surgical excision of the parasitic elements without contaminating the surrounding host's tissues is more important in preventing secondary echinococcosis as well as anaphylaxis than the false sense of security in depending on the scolicidal material for more than one reason. First, any substance injected in the cyst will be diluted to such an extent that it becomes an ineffective scolicidal material, for it is difficult to assess the exact volume of

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the cyst with any degree of accuracy. Second, in large cysts with many daughter cysts inside (multivesicular cysts) which are common, the injected material will not be distributed evenly so as to affect the various components of the cyst. Third, it is sometimes difficult to aspirate an appreciable amount of hydatid fluid in multivesicular cysts because of the presence of multiple septa which interfere with free aspiration. Also the time necessary for contact between the scolicidal material and the cyst's contents is unknown and accordingly varies from one surgeon to another. Eslami, Saidi and other workers7, 16, 20, 23 who have wide experience in hydatid disease came to the same conclusion after using various scolicidal materials. This may be considered as a point strongly in favour of the use of non-toxic but still effective solutions such as 20% hypertonic saline which is as effective as formalin but are non-toxic.

Filling the residual cavity after evacuation of the parasitic tissues with any of these safe scolicidal material and leaving this material for five to ten minutes will be very effective in destroying any residual scolices and accordingly lower the incidence of secondary echinococcosis.

One of the important problems to be faced when dealing with hepatic hydatids is the management of the residual cavity. Many surgical techniques have been

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evolved but none is perfect or applicable to all situations and each case should be handled on its own merits. Obliteration of the cyst cavity by successive layers of sutures starting from the bottom and suturing the adventitial walls together "capitonage" has been suggested<sup>8</sup>,<sup>16</sup>,<sup>17</sup>,<sup>20</sup> but it is unnecessary and difficult to apply in most cases. Omentoplasty<sup>18</sup>,<sup>23</sup>, although lowering the period of hospitalization and minimizing the incidence of postoperative infection in the residual cavity and adhesions of the viscera to its walls, is difficult to establish in every case.

Partial hepatectomy or lobectomy including the cyst, excision of the cyst in toto, excision leaving the adherent adventitia behind with or without its closure or partially deroofing were all used and proved to be safe and effective. Complete closure of the pericyst necessitates external tube drainage for a long time which may unnecessarily increase the hospital stay.

Despite all the measures taken by different authors to minimize the incidence of secondary echinococcosis, the incidence of recurrence in wellcontrolled cases amounts to  $11\%^{2}$ ,<sup>13</sup>.

The use of mebendazole before surgery to attenuate the scolices has been suggested by some authors, but experience with this drug is still in its early stages. Some have also advised its use as a curative method for selected cases<sup>3</sup>,<sup>4</sup>.

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