

## Original Articles

### HYDATID CYSTS

#### A CLINICAL STUDY OF A SHORT SERIES.

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THE subject, hydatid cyst, is familiar to every student of surgery ; it is a subject on which pathology books are expansive; and it is also rather a favourite subject with examiners : but from questioning medical associates in this area, I gather that they are not very commonly diagnosed in Western India, and it seems worth while to record a series of ten cases seen in the last four years, which, together, give a fairly comprehensive clinical picture.

*Incidence.*—In 1943, four cases were operated upon out of a total of 388 surgical operations (excluding minors), making 1 per cent of operation cases. On the average, however, two cases are seen a year, and this appears to be fairly constant, indicating that the condition is not very rare.

*Occurrence in animals.*—An enquiry from the local veterinary surgeon elicited the interesting information that he had on two occasions in the last year found hydatid cysts in the liver when post mortems had been requested because of unexplained death. One case was that of a buffalo which had been beaten heavily. At the post mortem the veterinary surgeon found several hydatid cysts in the liver, the largest being of the size of a mango, and of these at least two were ruptured and had leaked into the peritoneal cavity. Death was apparently due to acute anaphylaxis. In the other case the finding of the hydatid cyst appeared to be incidental.

He advanced the general information that hydatid cysts of the liver were seen only occasionally in slaughter-house inspections, chiefly in sheep, and rarely in goats and cattle. He could not give definite figures. He had no knowledge of the local incidence in dogs of *Taenia echinococcus*.

*Pathology.*—The salient points are familiar, but some are recalled as they help in visualizing the clinical picture.

*Taenia echinococcus* eggs are excreted in the faeces of the dog (the definitive host) in a developing state, the hexacanth or boring embryo. The egg is 30 to 36 $\mu$  in length.

On reaching the stomach of the intermediate host, usually sheep, occasionally man or cattle, the chitinous shell is digested, and the active embryo thus liberated bores into the mucous membrane of the small intestine and reaches a radicle of the portal vein, whence it is carried to the liver, being deposited near the hepatic surface on account of the terminal character of the blood vessels. Some ova apparently fail

to be arrested in the liver, and are stated to be carried to the lungs. Even here the ova may not be trapped, and then the general circulation is invaded, with the resulting development of cysts in any part of the body.

It is interesting that it has been recorded that ova are sometimes carried to the general circulation by the lacteals. This seems to afford an easier explanation of the entirely extrahepatic cases of which this series has four. It avoids the necessity of assuming the passage of a hexacanth five times the diameter of a red blood cell through the liver and lung capillaries, and, on the face of it, it does not seem more difficult for a boring embryo to find a lacteal than a portal radicle in the intestinal mucosa. The relative numbers of portal radicles and lacteals for instance, or some other physical factor, might provide an explanation why the majority find lodgement in the liver.

The remaining six cases were in, or arising from, the liver, and two of these had general dissemination in the peritoneal cavity, due to rupture of a primary liver cyst. One of these at operation was found to have numerous cysts on the surface of every viscus with a peritoneal covering, and also deeply embedded in the layers of the mesentery. In some cases the mesenteric cysts were so deeply embedded that access was difficult because of the mesenteric vessels. This is probably due to a mechanical burrowing during growth of the cyst.

The structure of the adult cyst is important in the consideration of treatment. It has three layers, the inner two of which (endocyst) are parasitic, namely, the germinal layer and the laminated membrane; and the outer layer (or ectocyst), which is adventitious, and due to the reaction of the tissues.

It may be noted that in every case operated on, numerous daughter cysts were found. It is stated that daughter cysts are rare in children, and also when the cyst occurs in a well-protected site, such as the brain or marrow cavity.

No case was observed in the lung, which is said to be the next commonest site after the liver.

#### Clinical features

The clinical features of each case are briefly recorded, in the order in which they were seen.

*Case 1.*—Male, age 35. Farm labourer.

Complaint : swelling in armpit. Examination showed a fluctuant non-tender swelling in the axilla, deep to the pectoralis major, about 5 inches in diameter. At operation a pocket was found to extend up to the first rib deep to the axillary vessels. Operation : marsupialization of the ectocyst after removal of as much as possible of the endocyst, and painting with 1 per cent formalin.

*Case 2.*—Male, age 35. Farmer.

A history was given of a swelling in the liver area for an indefinite period, which, one year previously, was followed by general abdominal pain and distension (rupture of liver cyst). Increasing swelling since with dyspnoea. The physical signs were of ascites and multiple nodules in the liver, which were at first considered to be malignant secondaries with general

carcinomatosis of the peritoneal cavity from some bowel primary, or a coarse cirrhosis. There were also several abdominal swellings which were difficult to define because of the ascites. Tapping was unsuccessful and he refused laparotomy. The general health, however, was better than would be the case in late secondaries or advanced liver damage. He returned after three months with more swelling, and many rounded masses were now clearly felt in the abdomen. He agreed to operation, and very numerous cysts of all sizes were found in the liver and throughout the peritoneal cavity. Complete removal of all was not practicable, but all the large ones and as many as possible of the small ones were incised through the ectocyst, and the endocyst removed complete. Post-operatively he did well, and there was relief from symptoms for  $2\frac{1}{2}$  years. He has recently attended again, in good general condition, with an incisional hernia at the lower end of the operation wound and one obvious mesenteric cyst the size of an orange. The liver enlargement is, if anything, less, and he has no ascites. He has consented to a second operation soon.

*Case 3.—Female, age 17. Farm labourer.*

Complaint : increasing swelling of the abdomen for three years. At first it was in the left loin and not painful, but it had gradually extended and was moderately painful. On examination it was found that the tumour extended from the left costal region to the left pelvis, crossed the midline anteriorly, and bulged the left loin. On pelvic examination it was felt in the left fornix. Its surface was regular, it was tensely cystic and dull to percussion.

It was at first taken to be a hydronephrosis, but this was disproved by a uroselectan x-ray examination, which showed a small deformed kidney shadow above the swelling with the ureter following a wide curve along the medial border of the tumour. The diagnosis being uncertain, it was approached at operation by the anterior route to give adequate room. The peritoneal cavity was opened through a left lateral incision, and the peritoneum was found to be reflected just to the left of the opening. A needle was inserted into the cyst through the peritoneum, and at first clear fluid was withdrawn. It was then pushed further in, and thick yellow pus was aspirated. In view of this, the opening in the peritoneum was closed, and the anterior incision closed in layers, and a lumbar incision was made. The usual innumerable cysts of all sizes were extruded, *some of which contained clear and some purulent fluid*. The ecto-sac was marsupialized and the wall swabbed with 1 per cent formalin. This patient returned fourteen months later for a different complaint and there was a soundly healed wound and no sign of recurrence.

*Case 4.—Female, age 35. Farmer's wife.*

Complaint : moderate pain in the epigastrium for one and a half years. Examination showed a curious fluctuating mass, the presenting part being about the size of an orange, in the epigastrium, moving with respiration, and also moving from side to side easily for about three inches to the right and left, rather like a pendulum, indicating a deep attachment. At operation its anatomical relation to the liver was very much like that of a huge gall-bladder (to the left of the normal organ), covered by peritoneum, with a distended 'fundus' and gradually getting narrower as it extended backwards and upwards on the inferior surface of the liver. Total excision was attempted but the 'neck' was so deep that this was given up in favour of incision and marsupialization.

*Case 5.—Male, age 30. Village carpenter.*

Complaint : noticed a hypogastric swelling for one month. No serious disturbance of bowels or micturition. Examination showed a broad tensely fluctuant swelling arising out of the pelvis, superficially resembling a distended bladder. P.R. there was a smaller fluctuant mass, the size of a lemon, felt high up in the anterior rectal wall, presumably the lower end of the swelling. Operation : extraperitoneal

marsupialization. The bladder was not exposed at operation.

*Case 6.—Male, age 32. Farmer.*

Complaint : small slightly painful swelling in the right epigastrium. On examination it was clearly a swelling projecting from the anterior surface of the right lobe of the liver, about the size of a walnut. Fluctuation could not be detected. General health good. On screening, the liver not raised, and the right diaphragm moved normally. At laparotomy the swelling was found to be cystic, and clear fluid was aspirated. A diagnosis of hydatid cyst was made, which was later confirmed by finding scolices and hooklets. On feeling the upper surface of the liver, separated by at least 4 inches of normal liver, was an irregular, lobulated, and not obviously cystic, swelling 6 inches by 4 inches in diameter. On this account a fairly extensive honeycombing of the liver was likely—the alveolar type of hydatid—and radical treatment was clearly impossible, so the anterior cyst was aspirated and 10 c.c.m. of 1 per cent formalin injected. Post-operatively, aspirations and injections of formalin were continued weekly for about a month. Eight months later the patient complained of pain, and aspiration showed a small amount of pus, so a second operation was performed. This time no cyst was evident on the surface of the liver, but it was localized by a needle and the liver incised down to it, the opening being enlarged by a finger. There was not excessive bleeding, and, after the loculi had been broken down as much as possible with the finger, a wide tube was inserted and left in as long as there was any discharge. The wound healed in about fourteen days, and the pain was improved, although the patient still complains of discomfort; it is probable that further trouble may arise from the deeper parts of the lesion which could not be treated.

*Case 7.—Boy, age 10. Villager. (Outpatient only.)*

Complaint : large painless swelling in right upper abdomen. On examination the right lobe of the liver was enlarged greatly and contained a cystic swelling, in which a thrill could easily be elicited. The father refused admission for operation, and further investigations were not possible.

*Case 8.—This was also seen as an out-patient only, at a village clinic.*

*Male, age 30. Labourer.*

Complaint : abdominal swelling ever since the age of 10, which was vouched for by his father. General health good, and discomfort slight, but the increasing size was causing anxiety. Examination showed several distinct globular abdominal swellings, about the size of a grape fruit, in each of which separately a thrill could be felt easily. The character of the thrill was exactly the same as is felt in a tense ascites on flicking with one finger and appreciating the vibration some distance away with the other hand. No primary cyst could be felt in the liver, and there was no free fluid discovered. The patient was instructed to attend hospital, but has not yet done so.

*Case 9.—Female, age 22. Weaver caste.*

Complaint : swelling on the antero-lateral aspect of the upper third of the right thigh for 2 months. It increased in size, became painful and then burst, about three weeks before admission, extruding what the patient described as egg-shells, together with pus. It was treated with simple dressings and a course of sulphonamides by a private practitioner and it apparently healed and the discharge ceased. Four days before admission the swelling increased and on admission was about 4 inches in diameter. On examination the lump was firm, roughly globular; fluctuation was not detected, and it was attached to the deep tissues (muscles) but not to bone, and there was an adherent scar. Elsewhere the skin was free. Treatment : It was first aspirated and thin yellow pus was obtained. It was therefore incised and a large number of hydatid cysts, mostly the size of a grape, together with about 3 to 4 oz. of free pus was discharged. Some of the individual cysts contained clear fluid, some purulent.

In view of the accessibility it was decided that a total excision of the hydatid cyst in this case should be done. However, this proved less easy than was anticipated, and required a wide dissection of the upper third of the thigh. The ectocyst was loculated and surrounded by dense fibrous reaction up to a full  $\frac{1}{2}$  inch in diameter, and the only way to remove it adequately was found to be removal of the whole of the upper third of the right vastus externus, showing the intermuscular sheaths and the main femoral nerve trunk as it divided into separate branches to the vastus group.

Before this fact was appreciated, some of the femoral nerve branches were picked up in a haemostat. They were, however, not divided except for the one to the vastus lateralis and the resulting quadriceps weakness is improving and is expected to recover fully, in the course of about 6 months. The leg was put into plaster in extension at first to rest the quadriceps muscle, but she is now walking without plaster and having electrical treatment. There is a saucer-shaped cavity which is healing from the bottom by granulation.

*Case 10.—Male, age 45. Farm labourer.*

Complaint: pain followed by swelling of right upper abdomen for six weeks. Examination showed a seriously ill man. He had a tender tense globular swelling of the liver extending 6 inches below the right costal margin, and across the midline. No thrill detected. No ascites. He had diffuse bronchitis.

Blood count: Hb. 75 per cent, R.B.C. 3,770,000, W.B.C. 8,000, polymorphs 76, lymphocytes 24, eosinophils nil.

Within 24 hours of admission he had a temperature of 104.2 and he was taken to the theatre. A needle was inserted and thin pus was found. Examination of this was made at once and one definite hydatid hooklet was found. Therefore laparotomy was done under local anaesthesia. The abscess of the liver was obvious but there was no adhesion whatever of the liver to the anterior peritoneum. As delay in waiting for adhesion was not considered possible, a long strip of gauze was soaked in 1 per cent formalin, and packed several times round the most prominent part of the abscess, shutting it off from the rest of the peritoneal cavity. A stab incision was made and an enormous quantity of pus and cysts was discharged. A large bore tube was inserted and found to go right through to the posterior part of the liver, at least 7 inches. The wound was packed around the tube.

The fever subsided in 48 hours and the abscess drained freely. When the packing around the tube was removed after 7 days, the drainage area was well shut off by adhesion from the general peritoneal cavity and no evidence of general peritoneal infection had been observed. However, the man gradually went downhill, and developed anorexia, pharyngitis and glossitis; in spite of intravenous glucose, and liver injections and nicotinic acid he died 6 weeks after the operation from what was presumed to be liver failure.

#### Differential diagnosis

The series illustrates several of the varieties of hydatid disease, and for purposes of considering diagnosis, the following classification is suggested:—

##### 1. Uncomplicated cysts:

- (a) Hepatic.
- (b) Extra hepatic.

##### 2. Complications:

- (a) Rupture.
- (i) Acute anaphylaxis.

- (ii) Dissemination (usually in the peritoneal cavity).

- (b) Suppuration,

and the diagnosis of each group will be considered briefly.

#### 1. Uncomplicated cysts.

The average clinical picture is of a person under 35, most usually in this series, but not always, a male, generally employed about a farm, in good general health, who complains of a tumour which is slowly growing and has been known to be present for years in most cases, but not giving rise to acute discomfort. On examination there is a tense spherical swelling which can be observed to have grown in the plane of least resistance, which usually means towards the surface of the body (or viscera). If it is superficial, a definite thrill is easily elicited. The skin over it is not attached to it or tender, nor is it attached to the surrounding tissues generally except in the case of origin from a viscera, when it may be sessile or attached by a pedicle.

(a) In the case of uncomplicated cysts in the liver, it has to be distinguished from *secondaries in the liver* (by the good general condition and the absence of any primary on careful search); from *coarse cirrhosis* by the good general condition again, and by the more irregular and hard surface in cirrhosis. The history and the Wassermann test may help.

(b) Uncomplicated extra-hepatic. Usually a cystic well-defined swelling as described above in any part of the body where such do not usually occur would give rise to a suspicion of hydatid cyst. No cases of the cysts in the brain or lung or bone have been encountered which would give rise to difficult diagnostic problems. However, it can be noted that the intense fibrosis around a hydatid in muscle proved misleading, and also that another case was reported to me by a colleague to have been diagnosed first as a cold abscess.

#### 2. Complications.

##### (a) Rupture.

(i) *Anaphylaxis*.—In this series the danger of acute anaphylaxis is illustrated in the veterinary case.

##### (ii) *Dissemination in the peritoneal cavity*.

—In differential diagnosis the possibilities of tuberculous masses in the abdomen and peritoneal carcinomatosis have to be considered. The chief difference lies in the good general condition, and the well-defined separate cystic masses as compared with the generally doughy feeling of a tuberculous peritonitis with or without irregular masses, or the hardness with irregularity of a carcinomatosis. If separate thrills can be felt as in case 8, the diagnosis can, I think, be regarded as certain. Ascites may be present in any of these conditions and obscure the diagnosis.

##### (b) *Suppuration*.

The differentiation of a suppurating liver hydatid from an amoebic liver abscess can be very difficult or impossible clinically. Both patients are very ill. A history of a pre-existing lump previously not painful suggests a hydatid, but may often not be obtained. A

history of recent dysentery, if present, would suggest tropical abscess. Aspiration in a hydatid will give thin yellow pus, while from a tropical abscess it is a rich reddish brown. Microscopical examination, if fortunate, may, in the case of hydatid cyst, show a hooklet or hooklets or rarely scolices. In amoebic infection the finding of amoebæ is uncommon. If a hooklet is found, then clearly operation and drainage are indicated. If not, and if doubt remains, a course of emetine has an almost magical effect on an amoebic abscess without secondary infection. There remains an intermediate group where the pus is not typical of amoebic abscess and the condition does not clear up quickly with emetine, nor are hooklets or other definite signs of hydatid disease found. Here there is a valuable place for the Casoni test, but this has not usually been available. I treated two cases of this kind by repeated aspirations plus sulphonamides, and they eventually improved and left, but one was reported to have died soon after of recurrence. After the experience of case 10 of a hydatid suppuration in the liver, I suspect that they may also have been infected hydatids, and would now explore and provide open drainage. Even if they proved not to be hydatids, after the failure of an adequate course of emetine and several aspirations, open drainage would be good treatment.

A curious observation was made in case 3 that aspiration gave successively clear and then purulent fluid. This observation was repeated in cases 5 and 9, when immediately it led to the exclamation, "Another hydatid!" In both cases some daughter cysts contained clear and others purulent fluid. The purulent fluid was sterile on culture on one occasion when tested. It would be interesting to know whether this has been observed before, and has, in fact, any diagnostic significance. The value of it, however, is limited by the fact that transperitoneal aspiration of a suspected hydatid cyst from the surface is inadvisable, unless it is followed by laparotomy immediately when proof of hydatid disease is obtained, either by the suggested sign or the finding of hooklets.

#### *Laboratory aids to diagnosis*

These have practically not been available in this series. However, I used some clear hydatid fluid from case 6 for the Casoni test (that from sheep not being available) on the patient himself, 0.1 c.c. being given intradermally, and thinking it to be similar to the tuberculin test, I took readings at 48 and 72 hours with a negative result. This was surprising, and the technique was checked and it was discovered that the reaction was almost an immediate one. 0.3 c.c. was, therefore, injected intradermally, and within ten minutes there was a wheal 35 mm. in diameter, which gradu-

ally passed off but was succeeded by a diffuse redness 3 inches by 3 inches when seen at 24 hours. The control was negative.

The precipitin reaction (Welch and Chapman) was once attempted but was inconclusive, and the complement deviation reaction (Weinberg and Parva) was not attempted. These two tests would need a larger laboratory, but the Casoni test is clearly of clinical value in small hospitals, when hydatid fluid is available.

A recent textbook states that it can be obtained from Allen and Hanbury's, but the Calcutta office does not stock it.

*Eosinophilia* has not been found of diagnostic value. In case 2 when first observed it was 12 per cent, which might be significant, but when taken three months later before operation it was only 2 per cent. In case 5 there was no eosinophilia found pre-operatively. Case 3 showed 2 per cent before operation; a routine blood count taken a month after operation, however, showed 14 per cent. A high count may, therefore, be suggestive, but a normal, or low one, does not rule out hydatid disease.

#### *Treatment*

The ideal procedure might be expected to be complete excision, but even in a case on the limb it proved relatively difficult and unnecessarily mutilating. It should not be attempted if it involves any risk of deep bleeding, or of escape of the hydatid fluid into the peritoneal cavity.

Complete removal of the endosac through the laminated layer is perfectly satisfactory, and was carried out in the multiple abdominal cysts of case 2, the endocyst being removed intact after incision of the ectosac.

In large deeply-placed sacs full of daughter cysts, this also was not practicable, and the procedure generally adopted was to open the ectosac near the surface after carefully packing off and protecting the surrounding tissues with towels wrung out in saline, and on top of that placing gauze soaked in 1 per cent formalin. All daughter cysts were then removed carefully, and the endosac of the main cyst removed as fully as possible, and then painted with 1 per cent formalin swabs. The adventitious sac was then brought to the surface and marsupialized by stitching to the skin, this being the standard treatment in the majority of cases. It would be better still to marsupialize the sac before opening it, but that was not always possible. The cavity was packed and dressed daily, and irrigation with 1 per cent formalin was also carried out several times again. A purulent discharge resulted, which lasted in this series from two to six weeks. The packing was maintained until it was gradually pushed out by the diminishing size of the cavity.

In eight cases there was no serious reaction after operation, and no evidence whatever of anaphylaxis.

#### *Follow-up*

Cases 3 and 4 are known to have a healed scar and no recurrence. Cases 1 and 5 have not reported again, and are probably well. Case 2 returned in good general condition after 2½ years, with peritoneal recurrence, prepared for another operation. Case 6 is still under observation and may have further trouble. Case 9 is progressing well and a cure is anticipated. Case 10, the only death in the series, had extensive liver damage from which he did not recover.

#### *Summary*

A clinical series of hydatid cysts is presented, indicating the incidence, presentation, differential diagnosis and response to surgical treatment. Six cases were hepatic, with or without peritoneal dissemination, two extra-peritoneal abdominal and two in the limbs. The general picture is a benign one in the absence of complications, particularly rupture into the peritoneal cavity and suppuration. Rupture may lead to sudden death from acute anaphylaxis, or, at a later date, such a widespread dissemination in the peritoneal cavity that extirpation of the disease is impossible. Also, the alveolar type in the liver cannot be dealt with radically.

It is pointed out that it may be impossible to distinguish a suppurating liver hydatid from an amoebic liver abscess, and it is suggested that in those cases where a course of emetine and aspiration does not have an unmistakable and rapid effect, open drainage should be carried out, and that the frequency of such infected liver hydatids may be found to be greater than previously suspected.

A special sign is mentioned, namely, that on aspiration of a suspected cystic swelling the finding of first clear and then purulent fluid indicates hydatid daughter cysts, of which only some are infected while others remain uninfected. This aspiration is not to be carried out transperitoneally, unless immediately followed by laparotomy if positive.

The absence of anaphylactic phenomena is pointed out when adequate care is taken and treatment is by incision and open drainage with marsupialization of the adventitious sac.

cases occurred, the remaining 13 among other Indian troops.

The epidemiological aspect formed an interesting study, but is outside the scope of this note which deals with certain clinical features and treatment.

#### *Type of case*

Cases were divided on clinical grounds into four groups of varying severity, the group 4 being subdivided into (a) non-fatal and (b) fatal cases.

*Group 1.*—Thirty-nine cases. These were cases of short duration and mild course. The average number of days of fever was 11.6, of which only 8.5 days on the average were spent in hospital. Some cases had only 5 or 6 days' fever in all. The majority presented the clinical picture of mild enteric for a few days, and then suddenly became afebrile and symptom-free (figure 1); but a few had fever as the only sign, and the diagnosis was confirmed by a positive blood culture, Widal reaction, or in one case by the appearance of a typical rash.

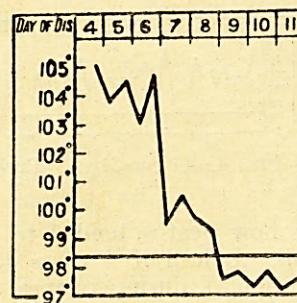


Fig. 1.—Group 1. Typhoid in an immunized subject. Severe onset with toxæmia and delirium. Crisis with disappearance of symptoms on 7th day. Blood cultures taken on 6th and 7th days grew *B. typhosus*.

The very much modified course of the cases in this group is almost certainly due to previous partial immunity. Its importance lies in the ease with such cases may be missed.

*Group 2.*—Forty cases. These also show the influence of previous immunization, usually with TAB. The average number of days of fever was

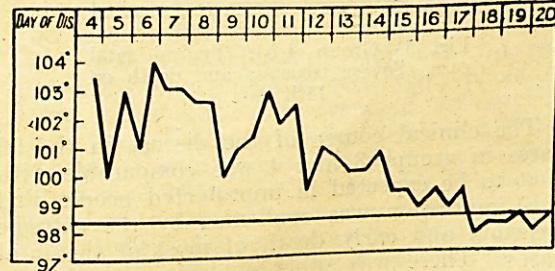


Fig. 2.—Group 2. Modified course of typhoid in a partially immunized subject. Blood culture taken on 6th day grew *B. typhosus*.

## AN OUTBREAK OF ENTERIC FEVER

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DURING the period March to June 1944, 175 cases of enteric fever were admitted to a military hospital in a training area for Gurkha troops. The great majority occurred during one six-week period. Among the Gurkhas 162