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Case Report

Multiple Cystic Echinococcosis in a Jordanian Patient: – Case Report and *In Vitro* Culturing –

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In Jordan, unilocular hydatidosis is endemic and widely distributed in both man (El-Muhtaseb, 1984; Shennak *et al.*, 1985; El-Muhtaseb and Shihabi, 1986) and animals (Ajlouni *et al.*, 1984; Abdel-Hafez *et al.*, 1986; Al-Yaman *et al.*, 1987 and 1988). In this report we describe a case infected with numerous hydatid cysts in multiple locations of the viscera and the attempts made to culture the parasite *in vitro*.

Case Report

A 65 year old male first presented to Karak hospital, south Jordan, in September 1991 with chief complaints of epigastric abdominal pain and occasional vomiting. Preliminary examination revealed a huge epigastric mass mainly to the left of the midline which moved when the patient respired. Multiple masses were also felt in the pelvic cavity. Examination by ultrasound showed a 15×15 cm cyst in the left lobe of the liver and 2 cysts about 5×5 cm in size in both iliac fossae coming out of the pelvic cavity. The patient was advised to have surgery but he declined until a year later when he presented again at the same hospital with the same upper abdominal and pelvic masses now associated with severe pain, repeated vomiting, loss of weight and frequent, urgent and burning micturition. Ultrasound was repeated to reveal the same cysts in the viscera. Tests of the liver function, ECG, chest X-ray and urine analysis were all normal. The patient was operated on the 5th of November 1992. Operative findings were consistent with those of ultrasound and revealed the presence of multiple hydatid cysts. A large cyst (15×15 cm in size) projected from the left lobe of the liver and was causing pressure deformity of the gastric outlet. Two large cysts (5×5 cm in size) projecting out of the pelvis and sinking deep in the pelvic wall were located lateral to the bladder and attached to its wall. Some 20 mesentric cysts and 1 splenic cyst (5×5 cm in size) were visible. An additional 67 small cysts, embedded in the lining of or suspended within the peritoneal cavity, were counted. Several small cysts (1×1 cm in size) were also found suspended from the lateral wall of the pelvis surrounding the pelvic vessels. The liver cyst was removed by pericystectomy with omentoplasty. The two large pelvic cysts were evacuated and closed suction drainage was applied inside the cavity after washing with a 5% solution of Povidone iodine. This effective antiseptic and disinfectant has the advantage of low irritability. Mesenteric cysts were partially excised and the raw area was covered by part of the omentum. The operation lasted 6 hours after which the patient was kept in hospital and put on mebendazole as a follow-up treatment of the remaining cysts. The patient recovered and was discharged from the hospital 12 days post surgery. Fig. 1 shows the patient prior to surgery. The hydatid cysts distributed in the viscera of the patient were externally visible, causing a deformity of the body contours.

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Fig. 1 Protrusions in the body line of a 65 year old patient resulting from a heavy infection with hydatid cysts.

In vitro Culture

An attempt to culture protoscolices (psc's) isolated from daughter cysts recovered from one of the pelvic cysts was made. The viability of the psc's was 80% and the culture was maintained *in vitro* for 60 days using the diphasic culture medium as described by Smyth (1985) and Hijjawi *et al.* (1992). Initially, the psc's evaginated at a rate of 90% (day 0). This was followed by the appearance of the excretory canals (day 6) and the excretory bladder (day 9) which correspond to stages Ps2 and Ps3 respectively according to the nomenclature system of Smyth (1985). Apart from an increase in the length of the cultured parasites to a maximum of 0.5 mm, no further development was observed up to the termination of the culture (Fig. 2). This was despite the normal appearance and observed mobility and activity of the cultured parasites.

Discussion

The large number of hydatid cysts present in many of the visceral organs of the patient were such that it was possible to trace the external contours of the cysts on his body surface (Fig. 1). The repeated vomiting of the patient was attributed to the pressure exerted by the liver cyst on the gastric outlet. The two large pelvic cysts pressing on the bladder neck were probably the cause of frequent, urgent and burning micturition. The endemicity of hydatid disease in the patient's region, his lack of awareness of the means by which it is transmitted to humans combined by poor hygiene must have resulted in his consumption of a large number of eggs in one or separate dosages. However, the huge number of small cysts distributed in the peritoneal and abdominal cavities may have resulted from the rupture of one or more of the cysts present in the viscera.

The heavy infection in this patient reinforces findings in recent studies of the hyperendemicity of cystic echinococcosis in Karak Governorate. In a retrospective study of surgically proven hydatid cases, some 676 cases were recorded from Jordan from 1985 to 1993 (Kamhawi, in press). Karak Governorate showed the highest rate of human infection at an annual incidence of 8.2 cases per a 100,000 of the governorate's population. Moreover, the highest rate of infection in herbivores throughout Jordan was observed from Karak Governorate

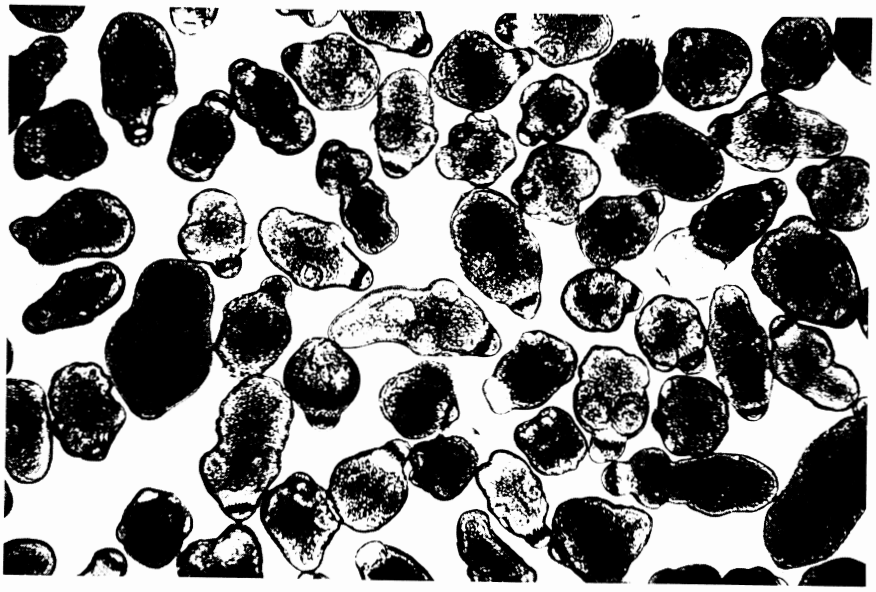

Fig. 2 Sixty day old *Echinococcus granulosus* metacestode stages grown *in vitro*. The cultures were initiated from human hydatid daughter cysts recovered from a Jordanian patient.

(Kamhawi and Hijjawi, under preparation). The infection rate in dogs sacrificed from Karak Governorate was 10% and the worm load was high with 1 dog harbouring some 25,000 adult *Echinococcus granulosus* worms (personal observation).

The cultured protoscolices obtained from the pelvis of the patient did not grow beyond the excretory bladder stage even after 60 days of culturing. This differs from the behaviour of previously described cultures of human origin from Saudi Arabia and Kenya which segmented at 28 and 45 days post culturing respectively (Smyth *et al.*, 1980; Macpherson and Smyth, 1985). It is worth noting that in all the attempts made to *in vitro* culture the three isolates mentioned above, none reached beyond the segmentation stage (S5). In contrast, sheep psc's from Jordan developed *in vitro* up to the 3–5 proglottid stage (S12) (Hijjawi *et al.*, 1992). Moreover, the development of donkey psc's from Jordan was different from those of human and sheep (Hijjawi *et al.*, 1992). Donkey psc's were delayed in reaching the banding stage (Ps4) and were sluggish and abnormal in their appearance. The significance of the differences observed in the behaviour of the three human isolates originating from different countries and in that of psc's from different hosts from Jordan may be a reflection of the complexity of *E. granulosus* strains and the effect of different host microenvironments. It is clear that more *in vitro* cultures of human origin need to be followed up. This is being carried out presently in our laboratory.

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