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Original contribution

Satellite cysts and biliary fistulas in hydatid liver disease. A retrospective study of 17 liver resections

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Summary In Switzerland, the preferred mode of treatment for hydatid liver disease caused by *Echinococcus granulosus* is surgery, giving us the opportunity for a retrospective histopathologic study of 17 consecutive liver resections. We focused on the occurrence of satellite cysts and of biliary fistulas and their effects on bile ducts. Of 17 patients, 6 (35%) had one or more satellite cysts, to be distinguished from internal daughter cysts. Small areas of fibrinoid necrosis within the fibrous pericyst, a surprisingly constant histologic finding, offer a simple explanation for the occurrence of such satellite cysts as well as for the development of biliary fistulas. Large fistulas with gross drainage of cyst contents into bile ducts were present in 5 patients (30%). The accompanying cholangitis was distinctly granulomatous in 2 of them, an observation rarely mentioned in the literature. All 5 patients with large fistulas also had chronic sclerosing cholangitis and dilatation of smaller bile ducts, in all probability the result of chronic cyst fluid leakage through preexisting, clinically silent smaller fistulas. Dilatation of small bile ducts is rightly considered a precursor sign for large fistulas. Awareness of the histopathology of these complications facilitates the interpretation of ultrasound and radiologic imaging, sheds light on their pathogenesis, and may influence the choice of treatment.

1. Introduction

Hydatid cysts represent the larval stage of the dog tapeworm *Echinococcus granulosus*. The disease is endemic in many countries and depends on the opportunity for the parasite to establish a durable life cycle, most often a sheep/dog cycle. Humans are accidentally infected by ingestion of parasite eggs which can develop into hydatid cysts, most frequently located in the liver. In Switzerland it is a rare disease, encountered almost exclusively in immigrants from endemic areas. Although classically described as a single cyst, preoperative imaging often shows more than 1 cyst. Further complications include biliary fistulas, bacterial superinfection, and, rarely, rupture into the peritoneal cavity. Treatment is surgical or by percutaneous drainage combined with intracystically injected scolicidal agents, and chemotherapy. The incidental observation of a distinctly granulomatous cholangitis in a patient with drainage of the cyst through a large biliary fistula prompted us to review our material consisting of 17 liver resections. We focused on biliary complications and satellite cysts, the latter being of special interest when percutaneous drainage is planned.

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2. Patients and methods

Of the 23 patients treated surgically for *Echinococcus* infection between 1991 and 2002 at Kantonsspital Aarau, 4 were infected by *Echinococcus alveolaris* and 19 by *E granulosus*, 17 thereof had hydatid cysts in the liver and were included in this study. All those 17 patients were immigrants from endemic areas. Fourteen were adults, 7 women and 7 men, median age 38 years (range, 19-52 years), and 3 were children, a girl of 8 and 2 boys of 14 and 11. In 14 patients abdominal pain was the principal clinical complaint, 1 patient had painful jaundice, in 1 patient the cyst had been detected incidentally, and in 1 patient we have no clinical information. One patient had recurrent liver disease 3 years after a primary intervention elsewhere. The surgical procedures were pericystectomy (6), segmentectomy (7), right hepatectomy (3), and left hepatectomy (1). Gross findings are based on pathology reports and supplemented by documents of preoperative imaging procedures. An average of 11 tissue blocs per patient (from 4 to 30) were available and all slides reviewed.

3. Results

Classically, the hydatid cyst is described as a single cyst confined by a parasitic capsule, the laminated membrane. Host tissue surrounds this parasitic shell by a layer of reactive fibrous tissue, called pericyst. Eleven (65%) of our patients had such simple cysts measuring between 2 and 18 cm, on average 9 cm.

Six (35%) of our patients, however, had one or more satellite cysts. A distinction has to be made between internal daughter cysts, satellite cysts, and cysts at completely different sites in the liver. In the first case, frequently observed in our material and not counting as multiple cysts, a mother cyst contains a certain number of smaller, thin walled, floating daughter cysts without contact to host tissue (Fig. 1).

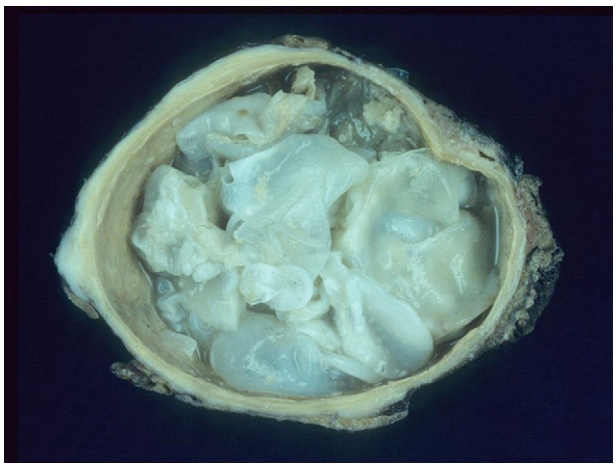

Fig. 1 Hydatid cyst filled with internal daughter cysts.

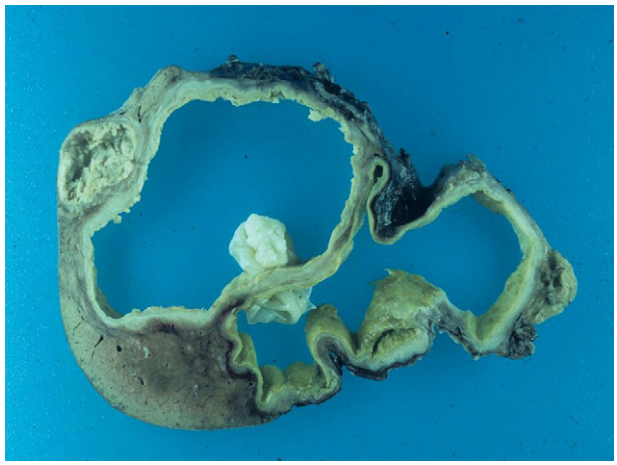

Fig. 2 Mother cyst and satellite cyst, both with their own fibrous pericyst. Small calcified cyst on the left corresponds probably to a second degenerated satellite cyst.

Satellite cysts on the other hand form close to, but outside, the mother cyst, come in contact with host tissue, and are surrounded by their own fibrous pericyst (Fig. 2). If they are numerous they form what is called a complex cyst, not to be confused with *Echinococcus multilocularis* with its much more numerous and much smaller, grapelike, sterile daughter cysts. According to this definition, 3 of the 6 patients with multiple cysts had a mother cyst accompanied by one satellite cyst with respective diameters of 11/6, 10/3, and 7/4 cm. One patient had 3 cysts measuring 15, 10, and 5 cm, and the 2 remaining patients (one of them with recurrent disease) had complex cysts composed of more than 3 cysts.

Cysts at two different sites in the liver, a much rarer event, must be the result of primary infection by more than 1 oncosphere, and by the same mechanism additional cysts in other organs can be explained. In only 3 of our 17 patients

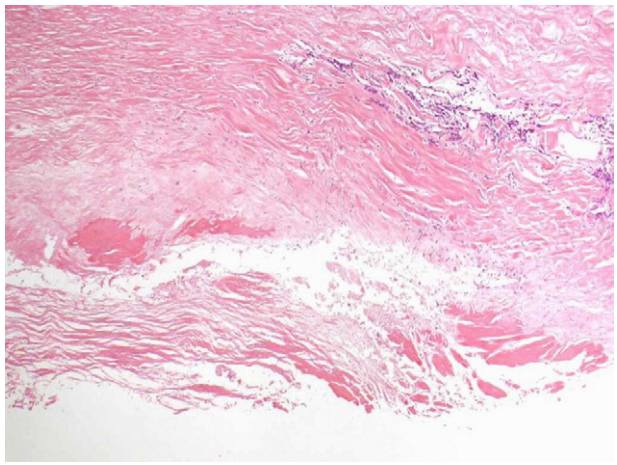

Fig. 3 Areas of fibrinoid necrosis within the inner layers of the pericyst (below), a constant histologic finding.