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Percutaneous evacuation (PEVAC) of multivesicular echinococcal cysts with or without cystobiliary fistulas which contain non-drainable material: first results of a modified PAIR method

H G Schipper, J S Laméris, O M van Delden, E A Rauws, P A Kager

Background: Surgery is the treatment of choice in echinococcal cysts with cystobiliary fistulas. PAIR (puncture, aspiration, injection, and reaspiration of scolecidals) is contraindicated in these cases.

Aim: To evaluate a modified PAIR method for percutaneous treatment of multivesicular echinococcal cysts with or without cystobiliary fistulas which contain non-drainable material.

Patients: Twelve patients were treated: 10 patients with multivesicular cysts which contained non-drainable material and were complicated by spontaneous intrabiliary rupture, secondary cystobiliary fistulas, cyst infection, or obstructed portal or hepatic veins; and two patients with large univesicular cysts and a ruptured laminated membrane, one obstructing the portal and hepatic veins and one a suspected cystobiliary fistula.

Methods: The methods used, termed PEVAC (percutaneous evacuation of cyst content), involved the following steps: ultrasound guided cyst puncture and aspiration of cyst fluid to release intracystic pressure and thereby to avoid leakage; insertion of a large bore catheter; aspiration and evacuation of daughter cysts and endocyst by injection and reaspiration of isotonic saline; cystography; injection of scolecidals only if no cystobiliary fistula was present; external drainage of cystobiliary fistulas combined with endoprostheses or sphincterotomy; catheter removal after complete cyst collapse and closure of the cystobiliary fistula.

Results: In all 12 patients initial cyst size was 13.1 (6–20) cm (mean [range]). At follow up 17.9 (4–30) months after PEVAC, seven cysts had disappeared and five cysts had decreased to 2.4 (1–4) cm (p=0.002). In eight patients with multivesicular cysts, a cystobiliary fistula, and infection, cyst size was 12.5 (6–20) cm, catheter time 72.3 (28–128) days, and hospital stay 38.1 (20–55) days. At 17.3 (4–28) months of follow up, six cysts had disappeared and in two cysts residual size was 1 and 2.9 cm, respectively (p=0.012). In four patients without a cystobiliary fistula, cyst size was 14.4 (12.7–16) cm, catheter time 8.8 (3–13) days, and hospital stay 11.5 (8–14) days. At 19.3 (9–30) months of follow up, one cyst had disappeared and three cysts were 8.5 (69–94)% smaller (2.2 (1–4) cm) (p=0.068).

Conclusion: PEVAC is a safe and effective method for percutaneous treatment of multivesicular echinococcal cysts with or without cystobiliary fistulas which contain non-drainable material.
tomography (CT) scan, and serology, and confirmed by microscopy of cyst fluid. Ultrasound, CT scan, and serology were used for follow up. All patients were treated with albendazole (median 11 weeks; range 3 days–2 years) before referral to our centre. When albendazole treatment had not been initiated, we started albendazole before the procedure and continued it for six months after the procedure at a dose of 10 mg/kg/day without interruption.

**Inclusion criteria**

Patients with multivesicular echinococcal cysts (Gharbi type 3) with or without a cystobiliary fistula and containing non-drainable debris were included (fig 1). Patients with univesicular cysts (Gharbi types 1 and 2) were included only if there was a cystobiliary communication or compression of the hepatic or portal veins or bile ducts. Exclusion criteria were: age <18 years or >75 years; no informed consent; serious coagulation abnormalities; known allergy to local anaesthetics or albendazole; pregnancy or women who refused contraception for the time of albendazole treatment; and cysts or albendazole; pregnancy or women who refused contraception for the time of albendazole treatment; and cysts or at least 50% reduction in cyst size at follow up imaging, and disappearance of complications such as pain, cystobiliary fistulas, vascular or biliary compression, and infection. Direct treatment results were evaluated at the end of the procedure: early results at six months and late results two years after PEVAC. The secondary end points of the study were recurrence of cyst cavity to >50% of its initial size, vascular or biliary compression, fistulas, pain and infection within two years after PEVAC, and death, withdrawal from the study, or loss to follow up. Three examples of cyst appearance before and after PEVAC are shown in figs 3–5.

**Statistics**

The Mann-Whitney rank sum test was used to compare results between patient groups and the Wilcoxon signed ranks test for paired observations within patient groups (SPSS for Windows).

**PATIENTS AND RESULTS**

**Patients**

Twelve patients with hepatic echinococcosis, mean age 38 years (range 22–61), immigrants from Morocco (five), Turkey (three), Pakistan (one), Syria (one), Afghanistan (one), and Greece (one) were treated for recurrent and severe upper abdominal pain. Ten patients had multivesicular so-called “mother and daughter” cysts (Gharbi type 3) containing non-drainable material. Two patients had univesicular cysts...
Complications

Cystobiliary fistulas and infections were the main complications, which occurred only in patients with multivesicular cysts. Less frequently observed complications were significant obstruction of portal and/or hepatic veins in three patients and perforation of a gastric ulcer into the cyst in one patient. Allergic reactions (transient fever, skin rash, eosinophilia) due to leakage of cyst fluid were observed in three patients following changing or removal of the catheter.

Three patients presented with spontaneous intrabiliary rupture. In five other patients the cystobiliary fistula became radiologically apparent on days 8, 17, 20, 25, and 53, respectively, after starting PEV. All but one cystobiliary fistula were endoscopically treated by introducing an endoprosthesis into the CBD or by sphincterotomy. In one patient a small cystobiliary fistula closed spontaneously (table 1).

A primary cyst infection was diagnosed in two patients and a secondary cyst infection in seven patients. All patients were successfully treated with antibiotics. In one patient with a primary infection, S. milleri and anaerobes were cultured from the cyst cavity. In the other patient, culture was negative due to antibiotic treatment. S. morbillorum, S. epidermidis, and C. freundii were cultured from the cyst cavity in two patients who were readmitted with cyst infection one and five months, respectively, after a PAIR procedure. Remarkably, in both patients bacterial culture of cyst fluid at the end of PAIR was negative.

Contrast injection at endoscopic retrograde cholangiopancreatography (ERCP) caused cholangitis and secondary cyst infection in five patients; three also developed a mild transient pancreatitis. S. milleri, C. freundii, C. albicans, S. sanguis, K. pneumoniae, H. influenzae, anaerobes, and enterococci were cultured from their blood, cyst cavity, or bile.

Cyst viability

In the initially sampled cyst fluid of nine patients, viable protoscolices were diagnosed in addition to fragments of the laminated membrane or hooklets. Remarkably, in both patients with multivesicular cysts which became infected following a PAIR procedure, protoscolices were still viable. In two patients with spontaneously infected cysts, viability of daughter cysts could not be assessed because cyst content was too purulent. In one patient with a multivesicular cyst, no protoscolices or hooklets were diagnosed.

Catheter time

In patients with a cystobiliary fistula, catheters were removed 72.3 (28–128) days after PEVAC, 53.6 (7–120) days after endoscopic treatment of the cystobiliary fistulas (mean (range) (table 1). Hospital stay was 38.1 (20–55) days. In patients without a cystobiliary fistula, catheters were removed after 8.8 (3–13) days and hospital stay was 11.5 (8–14) days. Catheter time and hospital stay were significantly longer in patients with a cystobiliary fistula (p=0.007).

Cyst size

Before treatment, cyst size was 12.5 (6–20) cm in patients with and 14.4 (12.7–16) cm in patients without a cystobiliary fistula (p=0.570). At the end of the procedure, when the catheters were removed, all complaints and complications had disappeared. Cavities had collapsed in all but one patient in whom the daughter cysts were partially evacuated. At follow up, 17.3
(4–28) months after treatment, cyst cavities had disappeared in six of eight patients with a cystobiliary fistula and were reduced to 1 and 2.9 cm, respectively, in the other two (p=0.012). In four patients without a fistula, cyst cavity had disappeared in one and cyst size reduced to 2.2 (1–4) cm in the other three, 19.3 (9–30) months after treatment. This 89 (69–100)% reduction in cyst size was not significant for this small number of patients (p=0.068). When all 12 patients were considered as a group, cyst size was significantly smaller 17.9 (4–30) months after treatment than before: 1.6 (0–7) cm versus 13.1 (6–20) cm (p=0.002). In none of the patients had complaints or complications recurred. No patient died, withdrew from the study, or was lost to follow up.

**Serology**

In comparison with cyst size, the response of total IgG echinococcal antibody titres was slow and started to occur...
Most infections were secondary to a prior intervention. Two patients were readmitted with a cyst infection at one and five months, respectively, after a PAIR procedure. Despite the infection both cysts were still viable. Contrast injection at ERCP also appeared to be a risk for cyst infection. All five patients who underwent ERCP developed cholangitis and cyst infection. In two spontaneously infected cysts, viability and possible infection of daughter cysts could not be diagnosed accurately. PEVAC was performed to support antibiotic treatment.

Our policy was to remove the catheter only when the cyst cavity had collapsed and catheter output was <10 ml/day. We reasoned that closure of the cystobiliary fistula and collapse of asterisks indicate spontaneous or †secondarily infected cysts, or ‡cyst cavity infection after PAIR. Most infections were secondary to a prior intervention. Two patients were readmitted with a cyst infection at one and five months, respectively, after a PAIR procedure. Despite the infection both cysts were still viable. Contrast injection at ERCP also appeared to be a risk for cyst infection. All five patients who underwent ERCP developed cholangitis and cyst infection. In two spontaneously infected cysts, viability and possible infection of daughter cysts could not be diagnosed accurately. PEVAC was performed to support antibiotic treatment.

Three patients presented with a spontaneous intrabiliary rupture and an overt cystobiliary fistula. Of note, in five other patients the cystobiliary fistula became apparent only 1–7 weeks after evacuation of cyst content. Cyst fluid was initially clear but became bile stained in the course of the procedure, in two patients even after about 100 ml were drained. Cystography at the beginning of the procedure could not reliably reveal a cystobiliary communication. The daughter cysts prevented the contrast from reaching the outer limits of the mother cyst. Therefore, care has to be taken with early injection of scolicidal agents into the cavity of multivesicular cysts.

The cystobiliary fistulas were probably firstly masked by the expanded endocyst and subsequently became unmasked by evacuation of cyst content and cyst collapse. However, we cannot exclude the fact that the negative pressure we used to evacuate the laminated membrane contributed to the development of cystobiliary fistulas. In all five patients with secondary fistulae, revealed by contrast injection, initially clear cyst fluid became bile stained in the course of the procedure. In Saremi's method, where the laminated membrane is also evacuated with the use of negative pressure, 34.5% of patients had bile stained drainage fluid and in 15.6% a cystobiliary fistula was demonstrated. In PAIR, where no assisted negative pressure is applied and the laminated membrane is not evacuated, lower fistula rates of 1.7–6.2% are observed.

In the end, in our patients, all eight cystobiliary fistulae closed 53.6 (7–120) days after endoscopic treatment, which is advocated in these cases.

The allergic reactions observed in three patients were probably due to leakage of cyst fluid after changing and removing the catheter. This illustrates the risk of long term use of large bore catheters, the need to closely monitor the patient at regular intervals, and the need for albendazole treatment after PEVAC to prevent widespread abdominal hydatidosis.

Table 1 Decrease in cyst size at follow up after percutaneous evacuation (PEVAC), catheter time (days), and hospital stay, related to the occurrence of cystobiliary (CB) fistulas plus cyst infection in patients with multivesicular echinococcal cysts in the liver. In patient No 8, cyst content was partially evacuated. Patient Nos 9 and 10 had univesicular cysts.
the cyst cavity would be closely related. Therefore, catheter
time and hospital stay were long. In the case of a cystobiliary
fistula, catheter time was 72.3 (28–128) days and hospital stay
38.1 (20–55) days; without a fistula, these times were 8.8
(3–13) days and 11.5 (8–14) days, respectively (p=0.007 and
0.007, respectively). This is comparable with a mean hospital
stay reported in surgically treated patients with cystobiliary
fistulas of 19.8–34.6 days.1–11 Hospital stay in our patients with
cystobiliary fistulas might have been shorter if we had
accepted some residual cyst size by removing the catheter ear-
cier. Another option is to discharge the patient and monitor
whether PEV AC reduces the relapse rate will need to be dem-
created access to a less invasive treatment for more patients.
Hospital stay. PEVAC will not replace surgery but may simply
hospital stay was long but comparable with that of
surgery. PEVAC may be improved to reduce morbidity and
hospital stay. PEVAC will not replace surgery but may simply
create access to a less invasive treatment for more patients.
Whether PEVAC reduces the relapse rate will need to be dem-
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