Simultaneous Percutaneous Alcohol Ablation of Pulmonary and Hepatic Hydatid Cysts: “PAIR” in a Pair

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Abstract

We present a 48-year-old lady with left chest pain and dyspnea of 6 months duration. Computed tomography scan revealed a left-sided cystic lesion in the lung parenchyma abutting the posterior pleura and chest wall along with a similar cystic lesion in the segment IV of the liver. A diagnosis of lung and liver hydatid cysts was made based on the imaging and positive serology. Percutaneous alcohol ablation of the lung and hepatic hydatid cysts was done using catheter drainage (PAIR-D: percutaneous aspiration, instillation of scolicidal agents, reaspiration of contents, and drainage) in the lung cyst and needle-PAIR in the liver lesion. The procedure was well tolerated without any intra- as well as postprocedure complication. The patient is well during 24 months follow-up without any recurrence or morbidity.

Keywords
► pulmonary
► hydatid
► percutaneous
► alcohol ablation
► PAIR

Introduction

Hydatid disease is caused by the larval forms of the cestode, Echinococcus granulosus where man is the intermediate host.¹ Hydatid disease is a zoonotic disease that is known to occur worldwide.² The highest prevalence of human hydatid disease in India has been reported from Andhra Pradesh, Saurashtra, and Tamil Nadu.³ Most commonly affected organs are the liver (~75%) and lungs (~15%).⁴ It is a major health problem with significant morbidity if left untreated. Lungs act as a second filter for the hydatid cysts in the human body and thus the second most common involved site.⁴ Various treatment options exist, ranging from pharmacological management to surgical resection. For pulmonary hydatid cysts, surgery is considered as first-line treatment with lung-preserving surgeries such as enucleation, pericystectomy, cystectomy or cystostomy with or without capitoneage being the options.⁵,⁶ Due to significant morbidity associated with surgery, there is an upsurge in minimally invasive surgical techniques such as thoracoscopic removal of the cysts located subpleurally.⁷ These techniques have been manipulated as an adjunct to the medical treatment.⁷,⁸

There are a few reports in the indexed literature stating the feasibility of thoracoscopic instillation of scolicidal agents especially hypertonic saline.⁷ Also, there are also stray anecdotal reports of percutaneous injection of scolicidal agents into the pulmonary hydatid cyst.

However, PAIR technique has been extensively used and is now considered as a well-established treatment option in appropriate cases of hepatic hydatid cysts. Encouraged by the results of PAIR in hepatic cysts and coupled with the fact that the pulmonary cyst in our case was abutting the chest wall and had a very easy access, we attempted PAIR-D (percutaneous aspiration, instillation of scolicidal agents, reaspiration of contents, and drainage) in the pulmonary cyst and a needle-PAIR in hepatic cyst in the same sitting. A written informed consent from the patient and clearance from the institutional review board was obtained before the procedure.

Case Report

A 48-year-old lady presented with left chest pain and dyspnea of 6 months duration. Abnormal physical findings were confined to the chest, which revealed dampened breath
sounds over the left lower chest. Routine blood investigations were unremarkable. Chest X-ray revealed a well-defined oval radioopaque lesion in the lower zone of left lung (Fig. 1). CT scan revealed a cystic lesion of size $6.5 \times 5.8 \times 6.2$ cm with moderately thick walls in the anterior and lateral basal segments of left lower lobe (Fig. 2A–C). The lesion was seen abutting the pleura and lateral chest wall with minimal perilesional lung atelectasis. No septae, membranes or solid component were seen inside the cyst. No adjacent pleural reaction/fluid was noted. No other focal lung parenchymal lesion was noted. CT scan of the abdomen revealed a similar cystic lesion of size $5.3 \times 4.9 \times 3.8$ cm in the segment IV of the liver (Fig. 3A). Sonography of the chest revealed an anechoic cyst with no intracystic contents. Indirect hemagglutination test for hydatid was positive. Contact history with livestock or pet was negative. Based on the above findings a diagnosis of lung and liver hydatid cysts was made. The patient was explained about the disease and treatment options. Subsequently, percutaneous alcohol ablation of the lung and hepatic hydatid cyst was planned using catheter drainage (PAIR-D) for the lung and needle-PAIR for the liver lesion because of its smaller size. Approval from the institutional review board was taken before the procedure.

**Technical Details**

Patient received oral albendazole, 10 mg/kg for 2 weeks before and after the procedure. Coagulation profile was normal. The patient fasted overnight. To minimize the possibility of allergic reaction, intravenous promethazine (25 mg) and steroid (hydrocortisone 200 mg) were adminis-
also obtained at 1 year and 2 year intervals. Throughout the follow-up, patient remained asymptomatic. CT scan at 1 year revealed a small collapsed cavity with membranes in the left lower lobe and at 2 years revealed an only small area of subsegmental atelectasis (► Fig. 4C–F). No fluid content was noted at any of follow-up scan. The liver cyst had also collapsed with the suggestion of clumped membranes and absence of fluid (►Fig. 3C) which at 2-year follow-up was reduced to a hypodense lesion of significant small size without any appreciable membranes (►Fig. 3D). Both the liver as well as the lung cysts showed more than 70% volume loss suggestive of a good response to ablative treatment.

Discussion

The liver is the most common site for hydatid disease with right lobe being affected most frequently. Most commonly patients may remain asymptomatic but few experience a dull pain in the right upper abdomen. There may be a presentation with various complications such as rupture into the peritoneal cavity and causing anaphylaxis, rupture into the biliary system, transdiaphragmatic spread into the lung, peritoneal seedling, portal vein involvement, and its thrombosis. Hematogenous spread can also occur into the lungs, spleen, kidneys, bone, and brain. A large proportion of pulmonary hydatid cysts may remain asymptomatic for a long time until they grow large enough to cause signs and symptoms, such as nonproductive cough, chest pain, and dyspnea. Complications include rupture, secondary infection, pneumothorax, and suppuration may sometimes precede the diagnosis. Various treatment modalities are available for treating pulmonary hydatid disease including medical and surgical options. Till now the only successful treatment for treating uncomplicated cysts included surgery. Medical treatment with benzimidazoles (mebendazole and albendazole) causes disappearances of cysts in 30%, partial response in 30%, and no response in remaining 40% especially if their size is > 4 cm and with thick walls. It is indicated in young patients and patients with high risk for surgery. We used prophylactic albendazole because it has been proven to
have a protective effect against spillage\textsuperscript{10} and also has an additive positive effect when used along with percutaneous treatment rather than any of these treatments alone.\textsuperscript{11} Due to significant morbidity associated with surgical procedures such as enucleation, pericystectomy, cystectomy or cystostomy with or without capitonnage various minimally invasive procedures have evolved for the treatment of pulmonary hydatid disease. Minimally invasive procedures included video-assisted thoracoscopic surgery,\textsuperscript{12} video thoracoscopy and cystectomy,\textsuperscript{9} medical thoracoscopy with instillation of hypertonic saline\textsuperscript{7} and even percutaneous hypertonic saline irrigation,\textsuperscript{13} etc. Traditionally PAIR/PAIR-D have been used in the treatment of hepatic hydatid disease with very good results. Various scolicidal agents such as hypertonic saline, 95% alcohol, and betadine have been tried with equally good results. However, there are only anecdotal reports in literature where percutaneous treatment has been performed using hypertonic saline as scolicidal agent for the treatment of pulmonary hydatid cyst.\textsuperscript{13} According to the literature published by Rai S, P et al, it was found that the cases which responded to albendazole in the initial course itself showed regression in size of the cyst and responded well during further cycles leading to complete response while in others there was an incomplete response to the medical therapy. Further, during their study, they found that the results with hypertonic saline as scolicidal agent in PAIR were not encouraging.\textsuperscript{13} Another report was published by Mawhorter et al in which they had treated pulmonary hydatid cyst with percutaneous aspiration and injection of hypertonic saline. However, in their report, they mentioned about aborting the injection of hypertonic saline, after an initial amount of 150 mL, since the patient developed a vigorous cough during injection followed by bronchospasm and fever which got resolved after a week of conservative management. However, later the patient developed pneumonia and hemoptyisis which were uncontrollable and ultimately pneumonectomy was done. The resected specimen did not reveal any residual hydatid.\textsuperscript{14}

\begin{figure}[h]
\centering
\includegraphics[width=\textwidth]{fig3.png}
\caption{CECT axial image (A) shows a well-defined hypodense cystic attenuation lesion in segment IV of the liver (arrow). NCCT axial image (B) at 1-week post needle-PAIR displaying regression of size of the cyst with internally detached membranes seen as hyperdense structures. CECT axial image (C) at 1.5 years reveals regression of size of the cyst with detached internal membranes and no significant fluid components. NCCT axial images (D) at 2 years reveals significant regression in size of cyst seen as a subtle hypodense lesion. CECT, contrast-enhanced computed tomography; NCCT, non-contrast computed tomography.}
\end{figure}
Percutaneous ablation of pulmonary hydatid cyst using 95% alcohol as scolicidal agent with PAIR-D technique is a feasible option especially in patients with peripherally located cysts and in patients with co-morbidities when surgery is not an option. Our case report also highlights the fact that PAIR is an efficacious treatment option for hydatid cysts occurring at the multiple sites and thereby preventing multiple surgeries for the same disease.

Conflict of Interest
The material is not presented elsewhere.

Note
This study was performed at the Department of Radiology Gobind Ballabh Pant Institute of Post Graduate Medical Education and Research, New Delhi, India.

Fig. 4 Chest X-ray (A) and axial lung window (B) at 1 week after the procedure reveal small collapsed cyst in left lower lobe (arrow) with detached internal membranes and drainage catheter in-situ. Follow-up chest X-ray (C) and CT axial image (D) at 1.5 years reveal regression in size of the cavity with hyperdense detached internal membranes. Follow-up chest X-ray (E) and CT axial image (F) at 2 years reveal a small patchy area of fibrotic opacity in left lower zone with diaphragmatic tenting. No residual cavity is visible in these images. Note absence of fluid contents within the cyst on follow-up images suggesting a good response. CT, computed tomography.
References