Complete Resolution of Bilateral Pulmonary Hydatid Cysts with Prolonged Administration of Albendazole: A Rare Occurrence

Zeba Siddiqui¹, Shahbaz Habib Faridi², Bushra Siddiqui³, Kaushal Deep Singh²

¹Department of Medicine, Era’s Lucknow Medical College, Lucknow, Uttar Pradesh, India.
²Department of Surgery, JN Medical College, Aligarh Muslim University, Aligarh, Uttar Pradesh, India.
³Department of Pathology, JN Medical College, Aligarh Muslim University, Aligarh, Uttar Pradesh, India.

ABSTRACT

Hydatid disease is a zoonotic disease caused by Echinococcus species. Echinococcus granulosus is the most common species which is responsible for hydatid disease in the humans who act as accidental intermediate hosts in the life cycle of the parasite. Pre-operative albendazole therapy followed by surgery is the mainstay of treatment for pulmonary and hepatic hydatid cyst at present. Not much data is available to support the sole use of albendazole for complete cure apart from its usage for those patients who have refused surgery or cannot undergo surgery because of any reason. We hereby report the case of a 12 year old female patient who was diagnosed with bilateral pulmonary hydatid cysts that responded well to Albendazole 15mg/kg /day given for a period of nine months with a drug free interval of one week every month. There was complete clinical and radiological resolution of hydatid cyst after the treatment. There has been no recurrence on follow-up of one year.

Keywords: Pulmonary hydatid disease; Albendazole; Echinococcus granulosus, Medical management.

INTRODUCTION

Hydatid disease is caused by the larvae of the tapeworm Echinococcus. Worldwide six species are recognized but only four are of public health concern.[¹] Human infestations are mainly caused by Echinococcus granulosus which causes cystic form of disease and has worldwide distribution. Liver is the most common site of cyst formation (~65%), followed by lungs in 10-30% cases and other sites such as spleen, kidney, orbit, heart, brain, breast and bones can also be involved in about 10% cases.[¹-³] In children, lungs are the commonest site of cyst formation.[⁴] Pulmonary hydatidosis affects the right lung in ~60% cases, 60% are located in the lower lobes, 30% have multiple pulmonary cysts and 20% develop bilateral cysts.[⁵] Majority of lung cysts are primary cysts but can occur following intrathoracic rupture of a liver cyst however only 20-40% patients with pulmonary hydatid disease also have liver involvement.[¹, ⁶] Hydatid cysts may remain asymptomatic for several years.
Most common symptoms of uncomplicated pulmonary hydatid disease are cough (53-62%), chest pain (49-91%), dyspnea (10-70%) and hemoptysis (12-21%) while larger cysts may cause symptoms by compression of adjacent structures.\[^4\]

There are cases in literature where medical therapy alone has cured small size pulmonary hydatid cysts but our patient had massive, bilateral hydatid cysts, which showed complete resolution with medical therapy alone. This sort of dramatic and effective resolution of pulmonary hydatid cysts with medical therapy alone has not been reported in literature.

**CASE REPORT**

A 12-year-old female presented to the outpatient department with complaints of cough, progressively increasing difficulty in breathing, fatigue, weight loss and loss of appetite for last one month. There was no history of fever, cough with expectoration, hemoptysis or chest pain. Physical examination revealed normal vitals. There was mild pallor but no icterus, clubbing or cyanosis was found. On examination of the chest, wheezes were audible bilaterally and there were no crackles. Cardiovascular system and other systems were unremarkable. Investigations revealed hemoglobin of 9.5 gm/dl, total leucocyte count was 5,500 cells/mm\(^3\), differential count revealed 80% polymorphs and 20% lymphocytes. Chest radiograph showed round homogenous opacities with smooth borders in bilateral parenchymal fields suggestive of bilateral pulmonary hydatid cysts (Figure 1a & b).

Computerized tomography scanning of thorax further confirmed the diagnosis. On the right side the cyst size was 6×6 cm and on left side cyst size was 8×7 cm (Figure 2). Enzyme linked immunosorbent assay (ELISA) for hydatid antigen was done which was positive. She was planned for surgical removal of the pulmonary cysts and was ordered preoperative Albendazole 15mg/kg in two divided doses. On her visit at 3 weeks, repeat chest radiograph was done which showed a dramatic reduction in the size of bilateral pulmonary cysts (Figure 3a & b). She also reported an improvement in general wellbeing and the cough had subsided. Considering her response to therapy, she was continued on same dose of Albendazole and was asked to come for regular follow-ups. During the follow-up period serial radiographs of the chest, showed dramatic improvement and she also improved symptomatically with regain of appetite. Within three months of therapy, her pulmonary cysts remarkably decreased in size and at 6 months the condition was completely cured which was suggested by the radiograph of chest (Figure 4a, b & c). She was given Albendazole for a period of 9 months and there was no recurrence in the follow-up of 1 year.
DISCUSSION
Lungs are the second most common site of hydatid cysts and most common site in children.\(^{[2, 4]}\) Most pulmonary hydatid cysts are acquired in childhood. They remain asymptomatic over a long span of time and are later diagnosed either incidentally on chest radiography or present with respiratory symptoms.\(^{[7]}\) Uncomplicated cysts appear as well defined masses, round or oval in shape in chest radiographs. They may vary in size from <1 to 20 cm.\(^{[8]}\) Contrast enhanced computerized tomography has vastly improved the diagnostic sensitivity of hydatid cysts. Casoni’s test and blood eosinophil count have little value because of low positive predictive value and false negative results. ELISA to detect IgG antibody against Echinococcus antigen in pulmonary hydatidosis has only 50-75% sensitivity.\(^{[4]}\) Differential diagnosis in a case of cystic echinococcosis include benign cysts, cavitary tuberculosis, mycoses, abscesses and neoplastic etiology.

Treatment of hydatid cyst of lungs is essentially surgical since the patient can be completely cured by removal of intact cyst and thus achieving complete parasite elimination from body. Surgical options for lung cysts include lobectomy, wedge resection, pericystectomy, intact endocystectomy and capitonnage.\(^{[9]}\)

In general, medical treatment has been used to supplement operative treatment by reducing intracystic pressure and to achieve complete cure; to avoid recurrences and to prevent secondary echinococcosis.\(^{[10]}\) Medical therapy is also used for cysts that are inoperable due to its location or medical condition and in patients with cysts involving more than two organs. Drugs that are used are Albendazole in oral dose of 10–15 mg/kg/day for at least 3 month separated by 14-day drug-free intervals. Mebendazole in dose of 40-50 mg/kg/day for 3-6 months orally and Praziquantel at a dose of 40 mg/kg/week are also used.\(^{[1, 2, 4]}\) Mebendazole attacks the parasite’s germinal layer where as Albendazole reaches high concentrations within the cyst and causes it’s death. Albendazole is preferred because of its high bioavailability and better efficacy. Percutaneous aspiration, injection, and re-aspiration (PAIR) has been used in the treatment of Echinococcal cysts in the liver, spleen, kidney and bones, but should not be used for lung cysts.\(^{[2]}\) Small (<7 mm diameter), isolated cysts, surrounded by minimal adventitial reaction respond completely with medical management.\(^{[11]}\) However, complicated, multiple, multi-organ, large, multi-compartment cysts with thick or calcified surrounding adventitial reactions are relatively refractory to medical management and require surgical excision.\(^{[5]}\) With a vast experience of chemotherapy with benzimidazole drugs, now a third of patients can be completely cured and 30-50% show significant regression of cyst size and resolution of symptoms using drugs alone.\(^{[11]}\) There has been extensive literature regarding complete resolution of small hydatid cysts with oral Albendazole alone.\(^{[11, 12]}\) However for cysts as large as in our case, there are no cases in literature that suggest that chemotherapy alone led to the complete resolution of cysts. Our case report provides the gateway for further studies to compare the therapeutic efficacy of surgery with or without Albendazole versus Albendazole alone for complete cure of pulmonary cystic echinococcosis.

CONCLUSION
Although India is a cattle-rearing country, still not much literature is available to compare medical management versus surgical treatment of pulmonary hydatid disease. Our patient had massive, bilateral hydatid cysts of lungs and she recovered after a nine-month period of Albendazole therapy opening new vistas in the field of management of Echinococcal disease. Many patients who cannot undergo surgery because of any reason can be effectively managed by chemotherapy but patient compliance must be ensured and follow up checkups must be planned properly beforehand. Further comparative studies with large number of patients are needed to establish role of isolated chemotherapy versus surgery and adjunctive chemotherapy; and duration of such a therapy to achieve complete cure.

What this study adds

1. **What is known about this subject?**
   Currently Albendazole is used to supplement operative treatment in patients with pulmonary and liver hydatid cyst. Medical therapy alone with albendazole is used for cysts that are inoperable due to its location or medical condition and in patients with disseminated disease.

2. **What new information is offered in this study?**
   Our patient showed a dramatic response to Albendazole therapy within 3 weeks of starting the treatment. There was clinical as well as radiological improvement so patient was continued on medical management. The cysts completely disappeared at 6 months but the patient was given therapy for 9 months. There was no recurrence after 1-year Follow-up.

REFERENCES


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