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Case Report

Concurrent pulmonary and hepatic hydatid cysts managed with single stage surgery

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ABSTRACT

Most of the case reports on hydatid cyst have documented its diagnosis and treatment involving either lungs or liver. This case report is unique as it documents the standard diagnostic and treatment approach followed for curing concurrent multiple hydatid cysts involving liver and right lung simultaneously via single stage surgery. A 52-year-old male presented with symptoms of simple pneumonia along with mild pain in the right upper quadrant. Hydatid cyst was considered as the differential diagnosis after physical examination. Both CT scan and MRI confirmed the presence of multiple cysts both in the liver as well as right lung. A single stage radical removal of cysts from both the organs was performed. The patient was hemodynamically stable, and no complications were reported postoperatively. This case report highlights the importance of considering hydatid cyst as a differential diagnosis in the light of vague presenting symptoms. Also, it emphasizes on the benefits of single stage surgery for removing cysts from both the organs simultaneously.

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Introduction

Hydatid disease is a parasitic infestation caused by *Echinococcus Granulosus* also known as hydatid tapeworm infection. The tapeworm requires 2 hosts to complete its life cycle. One being definitive host like dogs and dingoes, and other being humans as intermediate hosts. After 6 weeks, the hydatid cyst matures to form tapeworms inside definitive host and start shedding their immature eggs in the feces. Humans contract the disease by ingesting food or water contaminated by these immature eggs [1].

The eggs hatch to form hexacanth embryos or oncospheres which get lodged in vital organs like liver, lungs, and brain to form hydatid cysts. [1].

The parasite prevails in a wide range of climatic conditions ranging from the hot climate in Africa and parts of Asia to cold climate of Russia, mostly among people of lower socioeconomic status due to poor hygienic conditions [1,2].

The diagnosis of hydatid cyst is based on the patient's medical history, physical examination, diagnostic modalities like X-ray, ultrasound, CT scan and MRI, along with hematological, serum biochemical profiles, and serologic testing [3]. Efforts to improve diagnostic accuracy have led to integration

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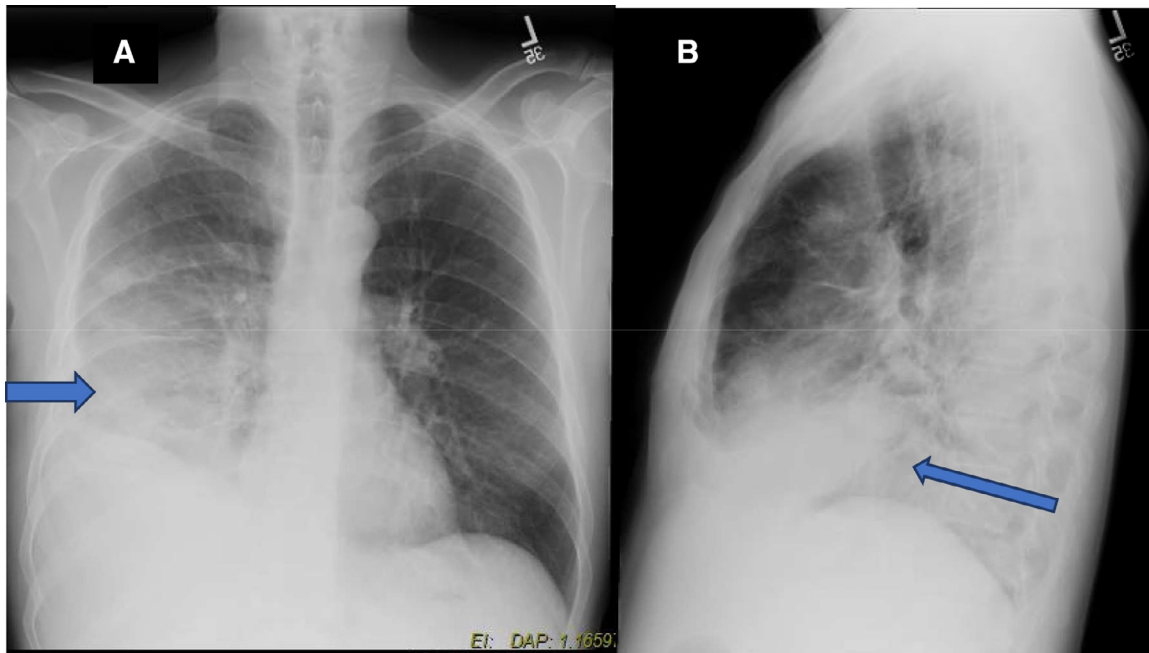


Fig. 1 – Chest X-ray (A). X-ray chest comparing left and right lung (B). Arrow showing airspace with probable cavitation in the right lung.

of a range of imaging techniques into the diagnostic armamentarium. The radical surgical removal of the cystic lesion remains the mainstay of treatment with a high success rate [4,5]. Chemotherapy, with benzimidazole compounds has been used with some success to sterilize the cyst. In recent years, a third treatment option for patients who cannot undergo surgery known as PAIR (Puncture, Aspiration, Injection, and Re-aspiration) has been introduced [6,7].

This case report highlights the concurrence of multiple hydatid cysts in lungs and liver which is very rare and focuses on how the disease can be diagnosed and cured from both the organs in single stage surgical management.

Case presentation

A 52-year-old febrile male patient presented with the history of insidious abdominal pain in the right upper quadrant for 1 year. He also complained of shortness of breath, cough productive of yellowish sputum, and chest pain associated with high fever (reaching up to 39°C) with chills and rigors for 2 months. At the time of arrival in OPD, patient presented with tachycardia and tachypnea and borderline blood pressure.

Based on history of illness and physical examination, chest X-ray, CT scan of lungs and liver along with MRI were suggested. Chest X-ray indicated of an airspace disease in the right lung probably with cavitation. There was mild right sided pleural effusion also (Fig. 1 A and B). The patient belonged to low socioeconomic status living in south region in a village, which also helped in making the provisional diagnosis.

The CT scan revealed cavitating lesion at the right lower lobe associated with lung atelectasis and consolidation with loculated plural effusion as shown in Fig. 2 A and B.

CT scan of liver showed multiple well-defined cystic lesions, the largest was noted in segment # 8/7 measuring about 12 cm. All these cysts demonstrated the water lily sign with serpinginous membrane noted within the cyst, a characteristic of hydatid cysts. Water lily sign is seen when there is detachment of the endocyst membrane to form floating membranes within the pericyst, thereby, mimicking the appearance of a water lily (Figs. 3 and 4).

MRI also demonstrated the similar findings and showed multiple cystic lesions that were high in signal intensity on T2-weighted images with internal folded membranes representing water lily sign. The largest one was noted in segment #6/#7 measuring 8.6 × 5.2 cm.

Then, the patient was evaluated by Inter Departmental team consisting of specialists from Pulmonary Medicine, ICU department, Thoracic surgery and infectious disease team. All routine hematological and urine investigations were carried out to assess the physical fitness of the patient for the surgery after taking informed consent. Antibiotic coverage was given 1 day prior to the surgery. The procedure was performed under General Anesthesia. The surgical resection of hydatid cysts in right lung and liver was carried out simultaneously by using standardized surgical approaches in single stage. Two hydatid cysts were radically removed from the right lung.

After completing the thoracic surgery, exploration of the abdomen showed multiple hydatid cysts in the right lobe of the liver. Using the cavitron ultrasonic surgical aspiration (CUSA) and multiple vascular staplers; right hepatectomy was done. The right hepatic vein was secured using vascular stapler. Segment 2 small hydatid cyst was resected separately using cautery and cavitron ultrasonic surgical aspiration (CUSA). After finishing resection, hemostasis was secured, and the drain was fixed on the right side of the liver. The patient was

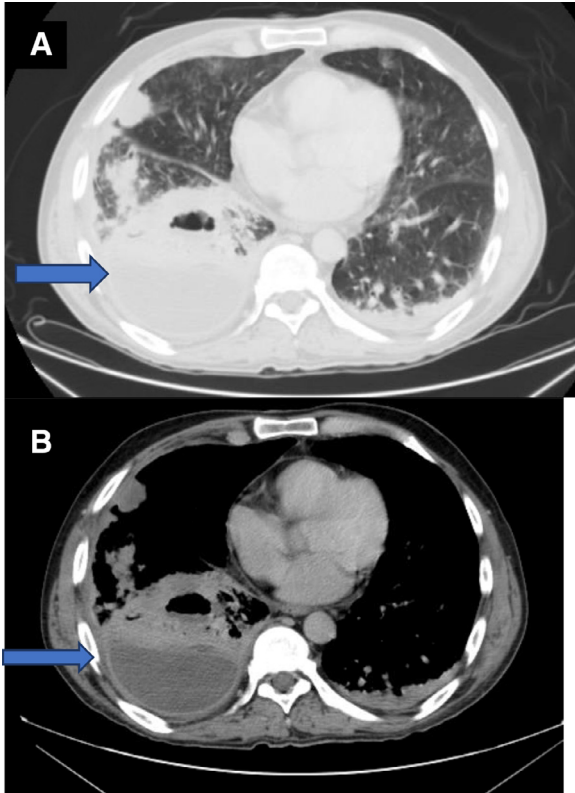


Fig. 2 – CT scan of lungs.

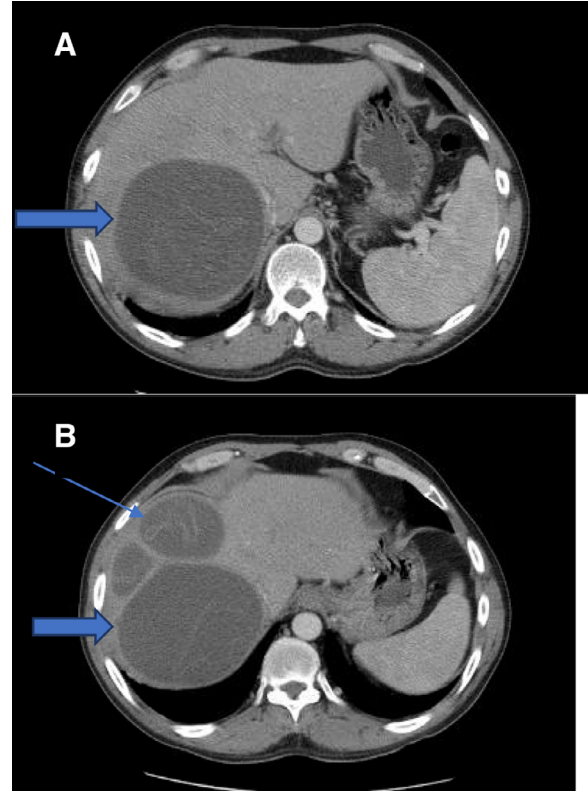


Fig. 3 – CT scan Liver.

stable during the procedure and he was sent to the ICU in a good condition.

The operation time was around 12 hours. Due to profuse intraoperative bleeding, 15 units of packed RBC along with 6 units of FFP were transfused to maintain hemodynamic stability.

After surgery, the patient developed mild right lung base empyema with its possible communication to the abdomen through defect in the right dome of the diaphragm. Hence, thoracentesis followed by pigtail drainage was carried out.

The ID recommended Tab. Albendazole for 6 weeks followed by Ciprofloxacin for the next 14 days.

The pathologic sample of the 2 hydatid cysts removed from the liver showed cystic cavity containing laminated membrane as shown in Fig. 5.

Histopathologic examination showed 3 layers of hydatid cyst:

1. The outer layer known as pericyst composed of modified host cell that form dense fibrous protective zone (yellow arrow).

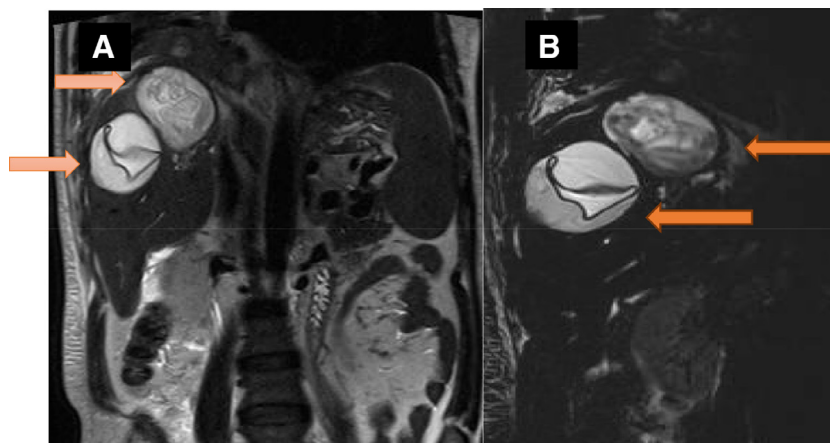


Fig. 4 – MRI.



Fig. 5 – Pathologic appearance of 2 hydatid cysts after cystotomy.

2. The middle layer is laminated membrane, which is acellular (green arrow).
3. The inner layer is germinal layer, where the scolices or daughter cysts and the laminated membrane are produced (blue arrow) as shown in Fig. 6.

Discussion

Hydatid disease is a parasitic disease secondary to the development of the larval form of *E. granulosus*. The concurrent presence of multiple cysts in both liver and lungs is a rare phenomenon (prevalence being 5%-25% of total hydatid disease cases) [8]. This is 1 reason indicating the importance of this case presentation.

The age of presentation is usually around 40 years [9,10]. In our case it was a 52-year male. There are 2 schools of thought regarding the gender predilection of this disease. Tekin et al reported a female predominance [11] whereas, Palanivelu et al found male predilection of the disease (5:1) in their study [10].

It is well known that the patients present with varied vague symptoms depending upon the (a) the organ involved; (b) size and site of the cyst; (c) interactions between expanding cysts and adjacent organs; and (d) complications caused by rupture of the cyst [12–14]. Pain, cough, low-grade fever, and the sensation of abdominal fullness are common presenting features.

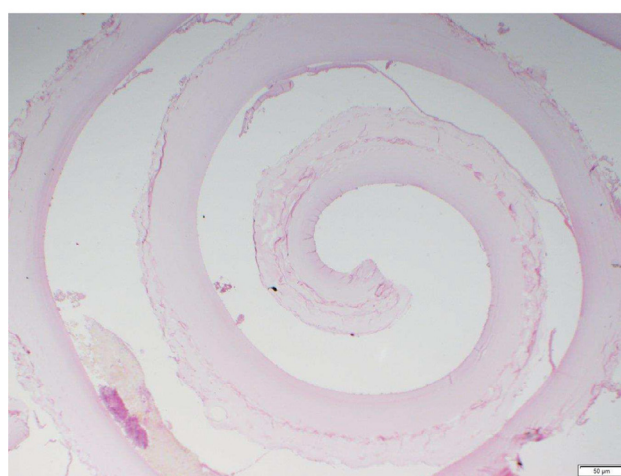
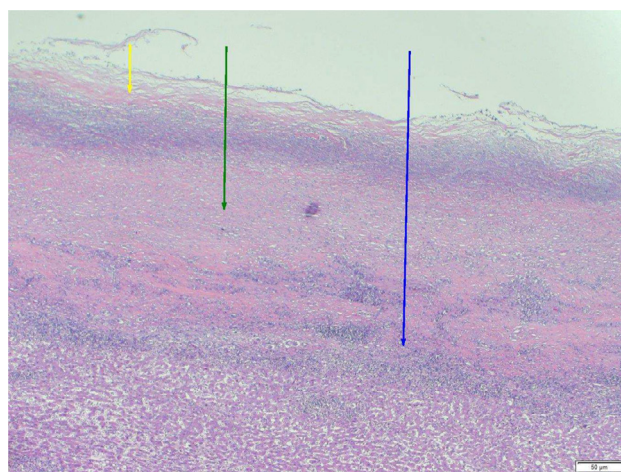


Fig. 6 – Histopathologic examination of hydatid cyst. (Color version available online.)

As the cyst grows, the symptoms become more specific depending on the specific structures involved. Secondary complications include infection or rupture of the cyst [15,16]. In our case also, the patient presented primarily with pulmonary manifestations as simple pneumonia (cough with high-grade fever) along with some abdominal discomfort in the right upper quadrant. Had we treated the patient for simple pneumonia and associated symptoms, the disease would have spread to other organs, along with aggravating the disease to a serious condition. The mortality rate due to this disease is 0.29%-0.6%. Here, we want to emphasize upon the need to consider hydatid cyst for differential diagnosis. This is the second reason why we wanted to document this case report.

Typical diagnostic techniques include ultrasound, CT scan, and MRI [17,18]. Ultrasound is the first diagnostic technique and no further imaging techniques are required if the typical water lily appearance is present [16]. The treatment of choice is radical removal of the cyst. However, preoperative medical treatment should be considered in order to sterilize the cyst, and to decrease the tension in the cyst so that there are fewer chances of spillage and resultant anaphylaxis. Postoperative medical treatment reduces the recurrence rates considerably [15]. We also followed this standard treatment protocol. The

third reason for documenting this case report is single stage surgical management protocol which we followed for treatment. It does not prevent the patient from undue stress but also reduces the cost of the procedure and, the incidence of postoperative complications and recurrence rate.

Conclusions

The concurrent occurrence of multiple hydatid cysts in liver and lung is a rare medical condition. But precise diagnosis at an earlier stage and single stage surgical management can not only prevent future complications but ensure speedy recovery as well.

Declaration of Competing Interest

I declare that i have no significant competing financial, professional, or personal interests that might have influence the performance or presentation of the work described in this manuscript.

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