Rare hepatopulmonary hydatidosis in children from a rural district of West Bengal, India: A case series

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Abstract

Background: Hydatidosis (hydatid disease) is a serious human cestode infection, caused by ingested egg of Echinococcus. It is endemic mostly in sheep or cattle-raising areas of the world including India. This disease usually presents in adulthood and relatively uncommon in pediatric population. Co-involvement of lungs and liver, known as hepatopulmonary hydatidosis (HPH) is found in around 6% cases of pediatric hydatid disease and very few cases have been reported previously from India.

Case summary: In this case series we reported three 6 to 8 years old children from rural background with multiple hydatid cyst involving lungs and liver, who presented with non-specific respiratory or gastrointestinal symptoms mimicking pneumonia or abdominal pathology. Chest X-ray followed by USG and CECT abdomen and thorax showed multiple cystic lesions of different dimensions in right middle and lower lobe of lungs along with right and left lobe of liver. Echinococcus IgG level in serum was high in all the children. They were treated with oral albendazole along with surgical interventions successfully with operative findings of ruptured cyst with hydatid sand and daughter cysts.

Conclusion: Rarity of the pediatric hydatid disease involving both lungs and liver prompted us to report three children from rural background with multiple hydatid cyst in both liver and lung.

Keywords: Children; Hydatid cyst; Liver; Lung.

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Introduction

Hydatidosis (hydatid disease) is serious human larval cestode infection, caused by ingestion of egg of the parasite Echinococcus (Echinococcus granulosus and Echinococcus multilocularis).It is a major public health problem especially in sheep and cattle farming areas throughout the world and prevalent in few states of India like Andhra Pradesh, Tamil Nadu, Jammu and Kashmir and Rajasthan **(Lee et al.,2005)**. Individually lungs and liver are the two common sites, but simultaneous involvement of lung and liver may be seen in 4% to 25% of patients with hydatid disease. Incidence of disseminated hydatid disease is rare in pediatric population and only few cases have been reported from eastern part of India. Here we have presented three cases of hydatid disease with concurrent involvement of lungs and liver in children from rural West Bengal.

Case Report

Case 1: A 6 years old girl from rural background presented with cough, low grade fever, breathing difficulty, nausea, vomiting and right upper abdominal pain for last 1 month .Positive findings were diminished breath sounds in right side of chest and 3cm palpable liver. Chest X-ray and USG thorax showed right sided pneumonia with pleural effusion and cystic cavity with air fluid level **(Fig 1A)** . USG abdomen showed two cysts in liver, one in right lobe (5.6cm ×4.2cm) and another in left lobe (4.2cm ×4.3 cm) . Blood investigations almost normal with haemoglobin 11gm/dl , WBC count 12,300 /cu,mm , with neutrophil 35% , lymphocytes 53% , Eosinophils 12% , platelet 5,20,000 /cumm , ESR 30mm /1st hr . Urea, creatinine and liver function test were normal. Pleural fluid study for cytology, biochemistry, gram stain, AFB stain , CBNAAT and culture sensitivity were within normal limit. CECT of thorax showed large thick wall cavity (8.6 cm ×8.2 cm) at basal region of right lower lobe , containing air fluid level . Multiple collections with enhancing wall was noted in segment 5 of liver in CECT abdomen (**Fig 1B**). Hydatid disease was suspected and Echinococcus IgG level in serum came out positive. MRI abdomen also showed two cysts in liver whereas MRI brain was within normal limit. Repeat USG of liver showed stage CE1>5cm (WHO classification) of cyst. Along with conventional antibiotics treatment, albendazole at 15mg /kg /day in two divided doses orally added for three months and patient sent for operative management of lung and liver cysts .Thoracoscopic right lung cystectomy, tube pneumonostomy and open drainage with omentoplasty of liver hydatid cyst were done under general anaesthesia and epidural analgesia (**Fig 2A**) . Operative finding was ruptured cyst with pale walls in right lower lobe of lung , filled with air and daughter cyst **(Fig 2B)** . In liver, two cysts with hydatid sand and daughter cysts were found without any cyst to biliary communication . Post operative recovery was good.

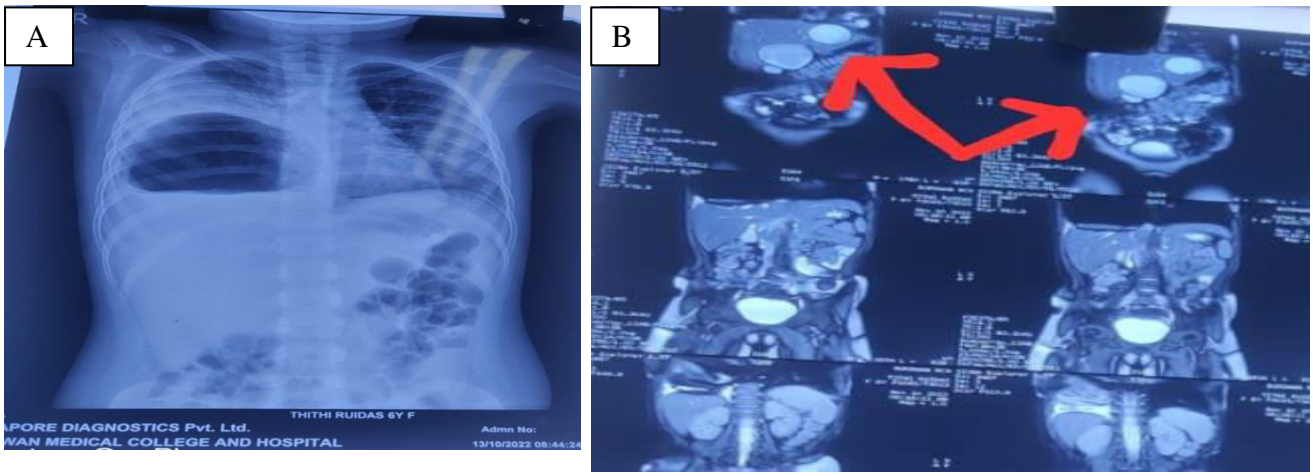

Fig 1 (A) Chest X-ray of case 1 showing cystic lesion in right lung field. (B) : CECT abdomen of case 1 showing multiple cysts in liver

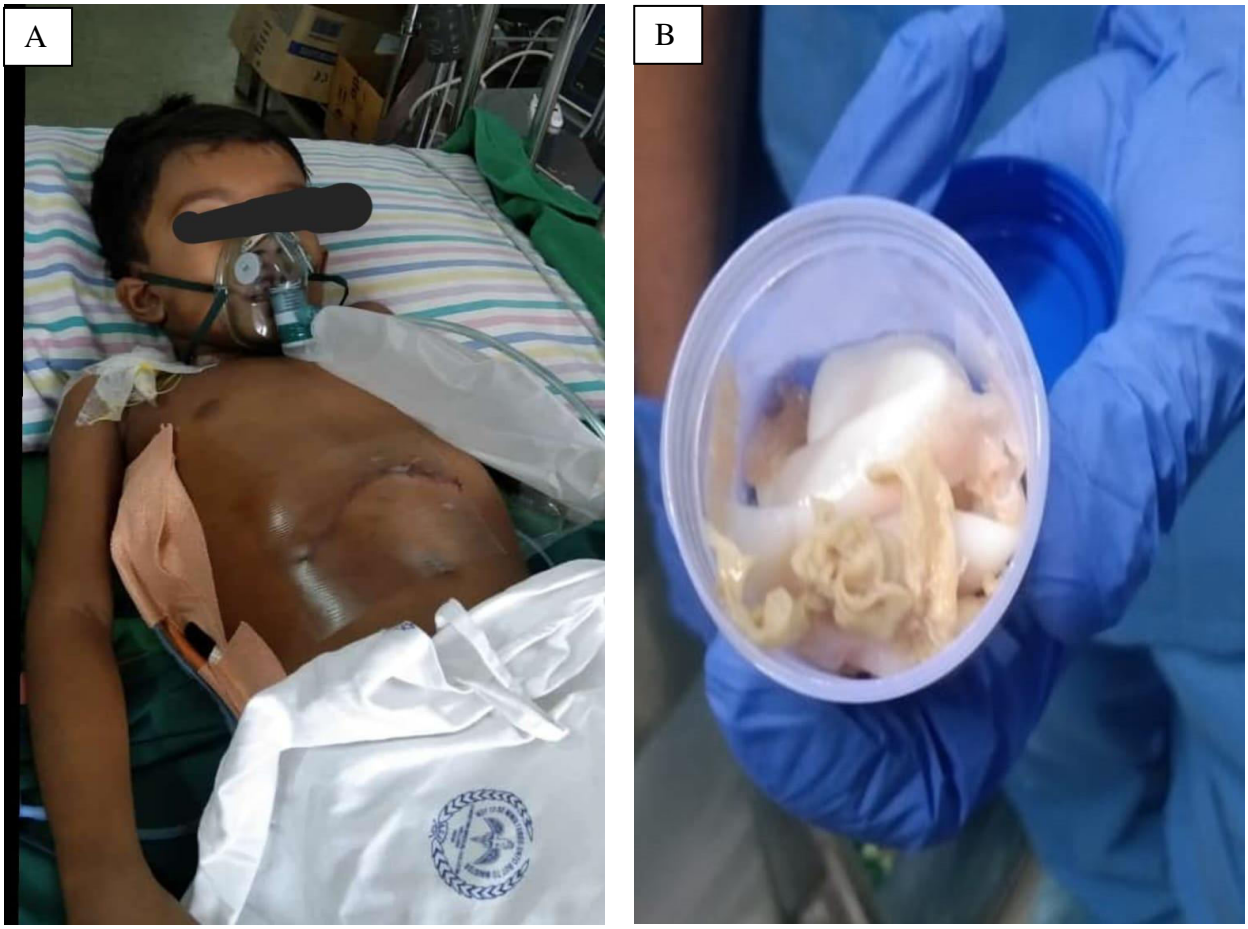

Fig 2 (A) – Post operative picture of case 1; (B) Resected cyst from case 1

Case 2 : A 6year old village boy presented with fever with nausea and vomiting for last 6 days with history of cough and occasional abdominal pain for last 1 month . He had no history of contact with TB . Child was toxic with coarse crepitation on right side of chest and chest X-ray showed right sided pneumonia with consolidation. Blood reports showed haemoglobin 12.5 gm /dl ,WBC count 16,500 /cumm with neutrophil 45%, lymphocytes 50% , eosinophils 5% and ESR 25 mm /1st hr. Fever profile including scrub IgM, typhidot test, CBNAAT sputum, blood and urine culture were negative and LFT was normal. Due to persistence of symptoms and cavitary shadow on chest X-ray (**Fig.3**) after conventional antibiotic treatment, we advised CECT thorax, which showed cavitary lesion with irregular wall(thickness 5mm) and air fluid level in right middle lobe and ground glass opacities in left sided lingular segment. USG abdomen showed one small cyst of 4.1cm ×3.8 cm in right lobe of liver . Oral albendazole was started after getting the positive blood report of echinococcus antibody and child was referred for operative management. Child recovered well after

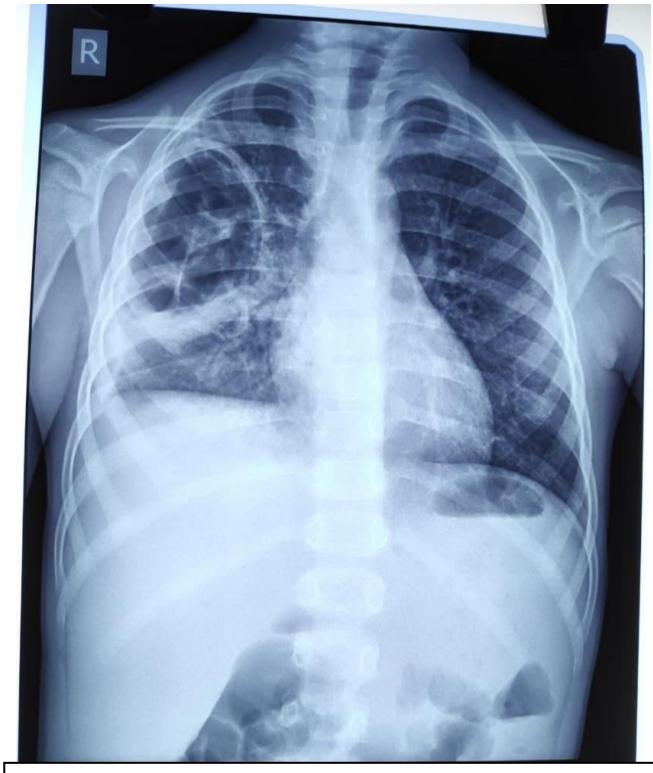

Fig 3.Cavitary lesion in chest x-ray of 2nd case

surgery and albendazole continued for 3 months.

Case 3: A 8 year old rural girl presented with right sided chest pain with cough and dyspnea for last 7 days with diminished breath sound over right thorax. Chest X-ray showed right sided huge rounded opacity(**Fig.4)** and USG abdomen revealed cystic SOL in liver. CECT of thorax and abdomen showed 9.0×8.1×7.7 cm well defined thin walled cystic SOL in middle and lower lobe of right lung (Stage CE1) and 2.6×1.4×1.9 cm cystic SOL in segment 8 of liver with daughter cysts (Stage CE3b). Blood reports showed haemoglobin 13.2 gm /dl , WBC 17,600/cumm , with neutrophil 58%, lymphocytes 38%, eosinophils 2%, CRP value 89 mg /l . Blood culture showed no growth. Other investigations including LFT were within normal limit except elevated Echinococcus IgG level in blood. Management was done according to the protocol. Oral albendazole added and the child was referred to higher center for surgical management. Recovery was good with surgery and three months albendazole therapy.

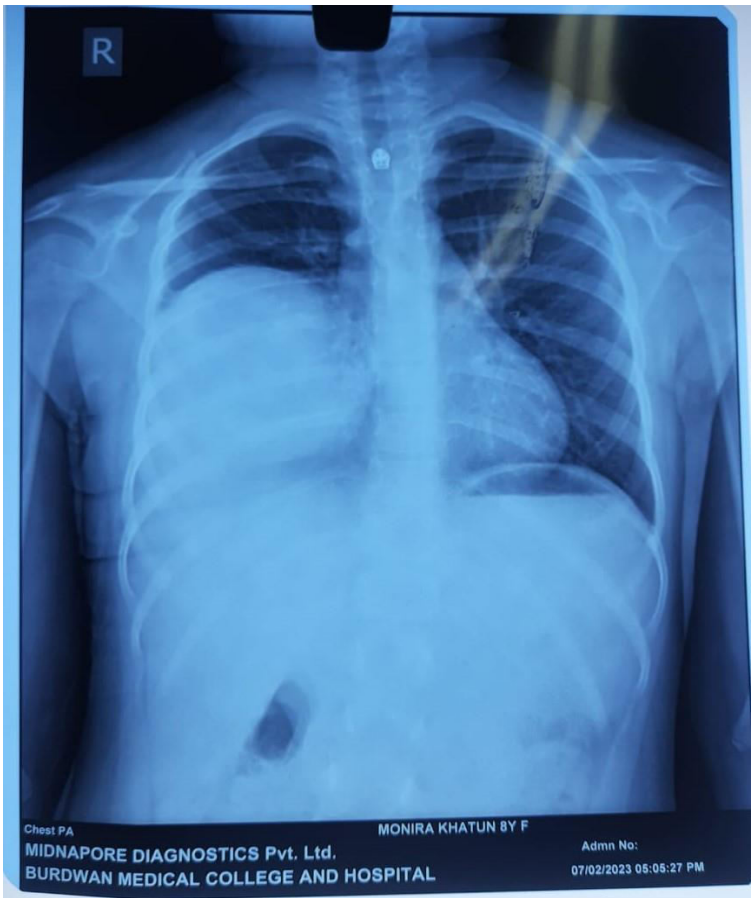

Fig .4. Chest x-ray of 3rd case showing rounded opacity in right lung field

Discussion

Hydatid disease is particularly endemic in sheep or cattle-raising rural areas of India where abundant stray dogs are found. Dog is the definitive host for Echinococcus and sheep or cattle are intermediate host. Human becomes accidental intermediate host by ingesting ova through close contact with infected dog. The egg hatches in the small bowel and releases an oncosphere that penetrates the intestinal wall and migrates through the circulatory system into various organs, especially the liver and lungs. In these organs, the oncosphere develops into a cyst that enlarges gradually, producing protoscolices and daughter cysts that fill the cyst interior. So, it commonly affects the liver (50-70%), followed by lungs (15-30%) and

rarely the other organs like muscle, bone, spleen, kidney, and brain **(Attef et al.,2009)**.

Hydatid disease is rare in children where most commonly involved organ is lungs. Simultaneous involvement of two organs are found in 5-13% of pediatric cases with co-involvement of lungs and liver in only 6% cases known as hepatopulmonary hydatidosis (HPH). Hydatid cyst can be solitary or multiple and the prevalence of multiple pulmonary cysts and bilateral cysts is 30% and 4% **(Aghajanzadeh et al.,2008 ; Mehta et al.,2016)**.

In our cases, the children had multiple hydatid cysts involving both lungs and liver. A recent study from West Bengal, India in 2022 depicted 16.6%(3 out of 18) incidence of hepatopulmonary hydatidosis among children **(Mandal et al.,2022).** Another study from India showed 11.1%(2 out of 18) incidence of both lung and liver involvement in children **(Gupta et al.,2014).** A 15 year long study from Iran showed 10.9% incidence of HPH in 5 to 80 years age range **(Shahriarirad et al.,2020**).Often hydatid cysts are detected as incidental finding as they remain asymptomatic for years. Cough, dyspnoea and haemoptysis are the typical features of lungs hydatid cyst while hepatomegaly and abdominal pain for hydatid cyst affecting the liver (**Shehatha et al.,2008 ;Toleti et al.,2012).**

Our cases presented with a cough, dyspnoea and abdominal symptoms. Diagnosis of hydatid cyst can be established with a combination of history, physical examination, radiological evaluation, and serological tests like ELISA **(Ramos et al.,2001).**

Treatment options for hydatid cysts are surgery, PAIR or medical therapy with albendazole and it depends on the staging of the disease **(Ezer et al.,2006 ; Dincer et al.,2006).** Surgical resection is the cornerstone for the treatment of hydatid cyst of lung and liver where cysts cause compression due to their large size. In case of liver cysts, stage CE 1 and CE 3a < 5 cm treated with albendazole alone whereas CE 1 and CE 3a >5 cm, CE2, CE 3b needs combined medical and surgical treatment. The duration of medical treatment ranges from 3 to 6 month, and it can be prolonged if needed **(Arif et al.,2011).** In our case series we followed combined treatment protocol of surgical resection along with albendazole therapy for three months with good result.

Conclusion

Hydatid disease in rare in children with simultaneous involvement of liver and lungs being rarer. It may presents with non-specific respiratory or gastrointestinal symptoms like cough, respiratory distress, fever or pain abdomen and incidental cystic lesions are found in chest x-ray or USG abdomen. So we should have a high index of suspicion whenever dealing with such cystic lesions in imaging and investigate further to diagnose this rare entity.

Ethics approval and consent : Informed consent was taken from parent or legal guardian of the children to publish the case series.

Abbreviations :

USG : Ultrasonography

CECT : Contrast Enhanced Computed Tomography

SOL: Space Occupying Lesion

AFB: Acid Fast Bacillus

CBNAAT : Cartridge based nucleic acid amplification test

CRP: C- reactive protein

LFT : Liver Function Test

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