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Multimodality Approach In Common and Unusual Locations Of Hydatid Cyst- A Caseseries

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Abstract

Introduction

Hydatid disease is caused by the parasite Echinococcusgranulosus. Humans are accidental intermediate hosts and the parasite commonly affects the liver and the lungs. Primary extrahepatic, extra pulmonary hydatid disease is rare. We are presenting a rare case series of hydatid cysts in various locations within the body. With this case series we want to create awareness about occurrence of hydatid cyst at different anatomical locations and their differential diagnosis at such locations.

Aim

In this case series we described the radiological appearance of diseases due to E.granulosus in common and unusual anatomical locations and imaging characterization of hydatid disease to differentiate from other cystic lesion. To create awareness for the same. Comprehensive systemic review of imaging features of hydatid cyst is lacking.

Study Design

Retrospective descriptive study done at American International Institute of Medical Sciences and GBH Hospital,Udaipur

Methods and Material

7 patients who were diagnosed as a case of hydatid disease at the Department of Radiodiagnosis, AIIMS Udaipur over a period of 3 years starting from January 2019 to December 2022 were included in the study.1.5T MRI, Voluson P8 USG, Siemens CT Scan has been used in this study. Patient's clinical and demographic data, location of the lesion, blood investigations, imaging findings, details of medical and surgical intervention done, and pathological examinations (wherever applicable) were reviewed. All patient information and records were kept confidential.

Conclusion

Hydatid cysts can be present in any anatomical location. Presentation at times is misleading.FNAC of Hydatid cyst is contraindicated due to chances of anaphylactic reactions. Diagnosis of hydatid cysts should always be considered as a differential diagnosis of every cystic mass in any anatomical location before diagnostic invasive intervention especially when they occur in areas where the disease is endemic. Treatment options are medical management, PAIR (cystablation), and total surgical excision which is the gold standard therapy followed by postoperative albendazole.

Keywords: Hydatid disease (HD), CT Scan , Ultrasonography(USG), Echinococcusgranulosus, Cystic lesion

Introduction

Hydatid Disease (HD) is an infestation that is caused by the larval stage of Echinococcusgranulosus [1]. Genus Echinococcus has four species that can cause infection in man. These are E. granulosus, E. multilocularis, E. vogeriand E. oligarthus. E. granulosusis the most widespread and cosmopolitan in distribution. The highest incidence is reported mainly in sheep-rearing countries[2] like Australia, New Zealand, China, South America, the middle east, and African countries.[5] Though India is not primarily a sheep-rearing country, a large number of cases have been reported in all regions of India with predominant prevalence in Andhra Pradesh, Saurashtra, Tamil Nadu and some parts of southern Rajasthan. The life cycle of Echinococcus is indirect and involves two hosts, one definitive carnivore host, and the other intermediate herbivore host. Humans act as an accidental intermediate host and ingest viable oncosphere-containing eggs, which are shed in the feces of the definitive host. [3]The oncospheres invade the intestines, enter the vasculature, and develop into hydatid cysts in any organ or tissue, producing symptoms. However, the liver acts as the first filter for hydatid larvae, making it the most commonly affected organ followed by the lung.[13]

The most frequent locations of the disease are the liver (50–77%) and lungs (18–35%), followed by the abdominal cavity and brain. In the abdominal cavity, splenic hydatidosis is primarily seen in 1.5–3.5% of cases of abdominal hydatosis.[8] Similarly in the brain, orbital involvement is seen in<1% of cases affected by this larval tapeworm.[8,9] Intracranial hydatid cyst is a relatively rare entity,accounting for only 1–2% of all intracranial space-occupying lesions. They are most commonly (50–75%) seen in children and young adults.[4] Hydatid cysts at unusual sites have been reported around the world including the spleen, kidney, heart, bones, muscles, and cranium, but soft tissue hydatid disease represents less than 3% of all hydatid diseases.[10]

The diagnosis of Cystic Echinococcus is based on the patient's history, clinical findings, hematological and serum biochemical profiles, and serological testing, which may be negative in 10% to 20% of cases. Efforts to improve diagnostic accuracy have led to the integration of a range of imaging techniques into the diagnostic armamentarium [7,12]. The radical surgical removal of the cystic lesion remains the mainstay of treatment with a high success rate [6,12]. Chemotherapy, with benzimidazole compounds, has also been used with some success to sterilize the cyst, decrease the chance of anaphylaxis, and reduce the complications, and recurrence rate post-operatively.[11] In recent years, a third treatment option was introduced (PAIR, puncture, aspiration, injection, and re-aspiration) and is indicated for patients who cannot undergo surgery. Compared with surgery, PAIR plus chemotherapy is associated with greater clinical and parasitologic efficacy with lower rates of morbidity, mortality, and disease recurrence including shorter hospital stays.[14,15] Here is a pictorial representation of life cycle of cystic echinococcus (Figure A).

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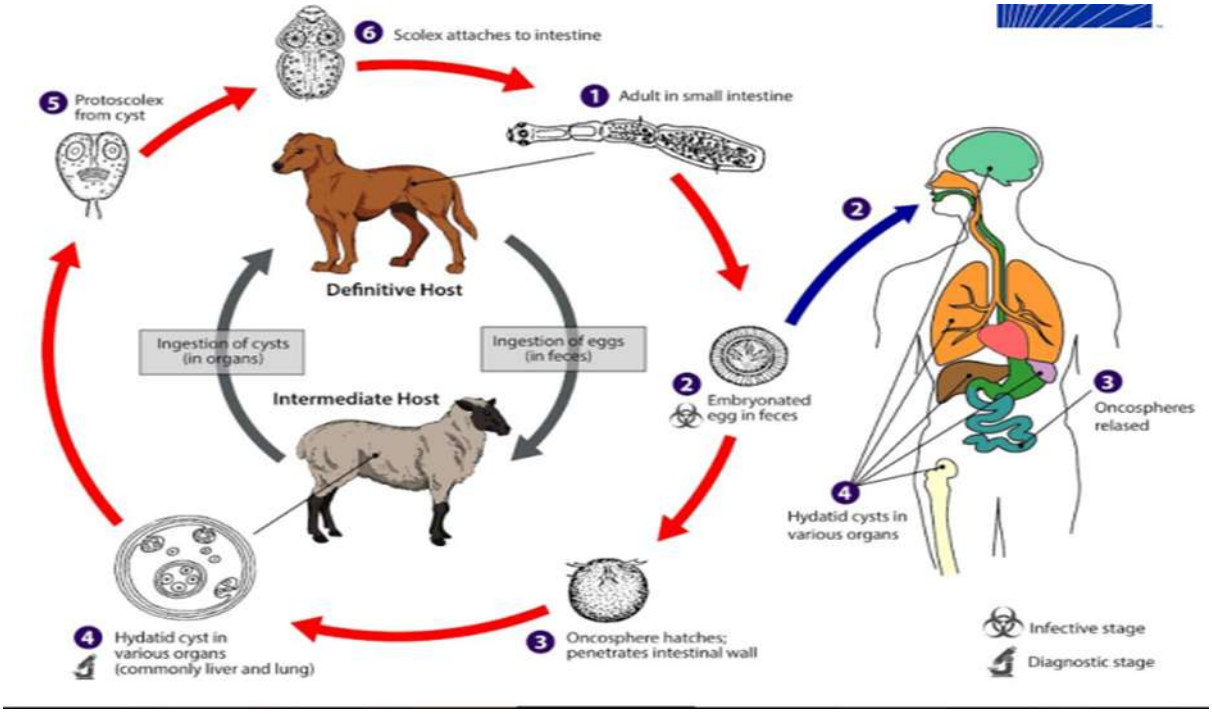
Figure. A: Diagramatic representation of life cycle of E. granulosus



Methods & Materials

A retrospective descriptive study done at the American International Institute Of Medical Sciences and GBH Hospital in the Department of Radiodiagnosis, AIIMS Udaipur over a period of 3 years starting from January 2019 to December 2022 was included in the study.1.5T MRI, Voluson P8 USG, Siemens CT Scan has been used in this study. Patient's clinical and demographic data, location of the lesion, blood investigations, imaging findings, details of intervention was done, and pathological examinations (wherever applicable) were reviewed. All patient information and records were kept confidential.

Results

Seven patients were diagnosed with hydatid disease at various sites. The age of patients was between the range of 30 - 75 years. Nonspecific abdominal pain and a non-tender palpable abdominal lump were the most predominant symptoms other symptoms varied according to the location of the cyst. Surgical treatments include complete cyst excision (cystopericystectomy) in most patients. Partial nephrectomy, lumpectomy, and splenectomy, were performed whenever cysts invaded these organs. Eosinophilia was observed in 6 out of 7, patients. The diagnosis was confirmed by hydatid serology, ultrasonography, MRI/CECT, and histopathological examination of the specimen. In the histopathological examination of all cases, we have searched structures in conjunction with HC to be able to come up with a definite diagnosis.

Case 1: Spleen hydatid (the third most common location of hydatid disease involvement after the liver and lung)

A 35-year-old female patient presented to our hospital with a complaint of pain in the left hypochondrium for the last 7 days along with multiple episodes of vomiting. She had a history of contact with animals and was non vegetarian. Her blood counts showed eosinophilia. Local findings are suggestive of palpable spleen grade-I. Her ultrasonography (Figure 1a) suggested a well-defined anechoic cystic lesion measuring 12x10cm with homogeneous internal echoes seen at the superior pole of the spleen. The lesion shows no internal daughter cyst. CECT Whole abdomen was done (Figure 1b and c) which suggested a grossly enlarged spleen measuring approx 170mm in size and the presence of subtle peripheral enhancing fluid attenuation, well-defined cystic lesion measuring 116x107x119mm (AP X TR X CC)is seen involving

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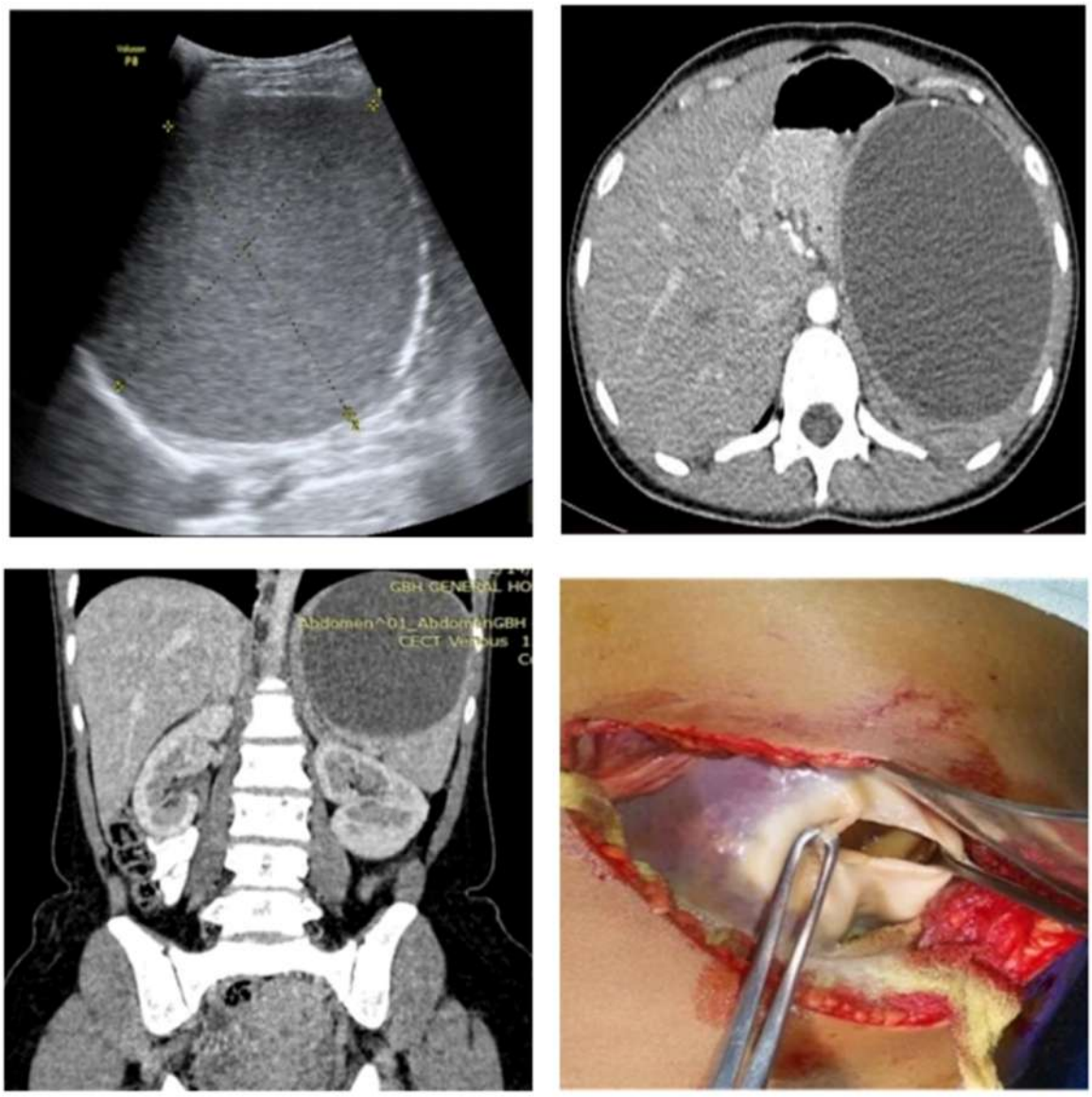
upper pole of the spleen. The lesion is showing multiple peripheral multifocal punctate as well as curvilinear calcific foci. The diagnosis was confirmed by serology tests and IG. Splenectomy was done under GA (Figure 1d) and she was started on a course of Albendazole 400 mg and Praziquantel 600 mg twice daily. On her 2 months follow up she had symptomatic relief and was planned for a 12 months course.

Figure 1: (a) Ultrasonographic View. (b) CT axial view. (c) CT coronal view. (d) Intraoperative view.



Case 2: Spine hydatid

A 39-year-old male presented with severe pain in the lower back with weakness, walking difficulty, numbness, and urinary incontinence for the last 2 months. The pain was not relieved by conservative management. CNS and neurological examination states intact cranial nerves, spastic paraplegia with power 2/5, exaggerated lower limb reflexes, the definite sensory level at D10 to D12 dermatome, and autonomic disturbances such as retention of urine. Local Examination suggests paralysis of lower limbs with tenderness in the lower back. Clinically cord compression was stated. On examination, 12cm×5cm swelling, and firm consistency was present in the lower back region. Routine blood investigations (including RFT and LFT) and all

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routine blood tests were fairly normal, except for slightly lower Hb levels (9 gm%) and slight eosinophilia (E-8%). Ultrasound of the abdomen and pelvis revealed no significant abnormality, no mass or space-occupying lesion in any abdominal viscera. X-Ray lumbar spine (AP and Lateral) shows a lytic lesion involving D 10 to D12 vertebral bodies with partial collapse of the vertebra. MRI scan of the spine was performed on a 1.5 Tesla scanner and it showed a large multiloculated fluid containing mass in the adjacent paraspinal musculature extending into epidural space and causing compression of the thecal sac and causing cord compression resulting in T2 hyperintense signals in the spinal cord at D10 to D12 levels (Figure 2a,b,c). Based on such findings, differential diagnoses considered were – cystic neoplasm of bone-like Giant cell tumor, hydatid disease, Pott's spine, vertebral pyogenic infection, multiple myeloma, and enchondroma was made. It also showed the involvement of the spinal canal and adjacent paraspinal soft tissues. Diagnostic partial excision of the posterior paravertebral component of the lesion was done revealing cystic swelling containing daughter cysts (Figure 2d,e) The histopathology report showed features consistent with a hydatid cyst. (Figure 2f) The postoperative period was uneventful. The patient was started on Tab. Albendazole 400mg twice a day for 4 months.

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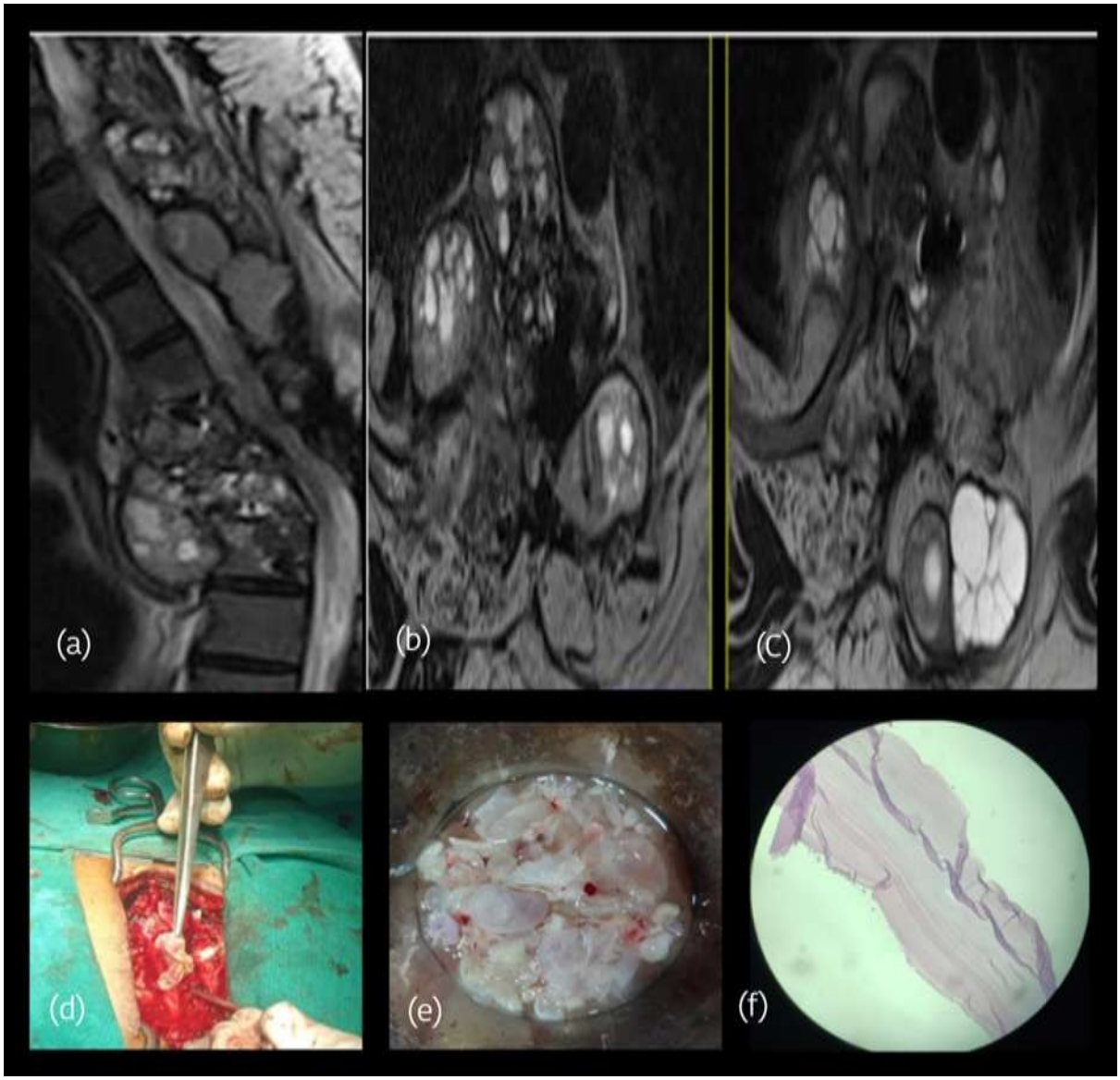
Figure 2: (a, b, c) T2 weighted MRI images. (d) Intraoperative Image. (e) Post-operative Specimen showing multiple thin walled, fragile, translucent cysts, having thin, watery serous content within. (f) Histopathological examination of cyst wall



Case 3: Liver hydatid (most common site)

A 78-year-old male presented with abdominal pain for the last 1 month not associated with vomiting or jaundice. No history of fever or weight loss. Abdominal ultrasound (Figure 3a) suggested a large cystic lesion measuring approx. 11.93cm x 10.24cm with multiple internal septations and a few daughter cysts seen involving segment II of the liver. CECT (Figure 3b,c,d) of the Whole Abdomen was done showing the presence of peripherally enhancing fluid attenuation well defined cystic lesion measuring 58 x 65 x 68mm in size is seen involving segment II of the liver. The lesion is showing internal septae, Few curvilinear calcific foci are noted along the capsule suggestive of hydatid. Postoperative fluid collection measuring 59 x 54mm is seen in the perihepatic region along segment VII and VIII of the liver. Minimally prominent bilobar IHBR is seen. Routine investigations were completed. TLC was 13000 and eosinophils were 11%. Differential diagnoses include liver abscess, hematoma, cavernous hemangioma, lymphoma, and cystic metastasis. He was put on Tab Albendazole 400mg OD. PAIR was done for the cyst in the liver using absolute alcohol. Aspirated fluid was sent to the Microbiology and Pathology departments for investigations. The reports included an Acid-fast bacillus stain which showed protoscolex and it confirmed the diagnosis as hepatic hydatid.

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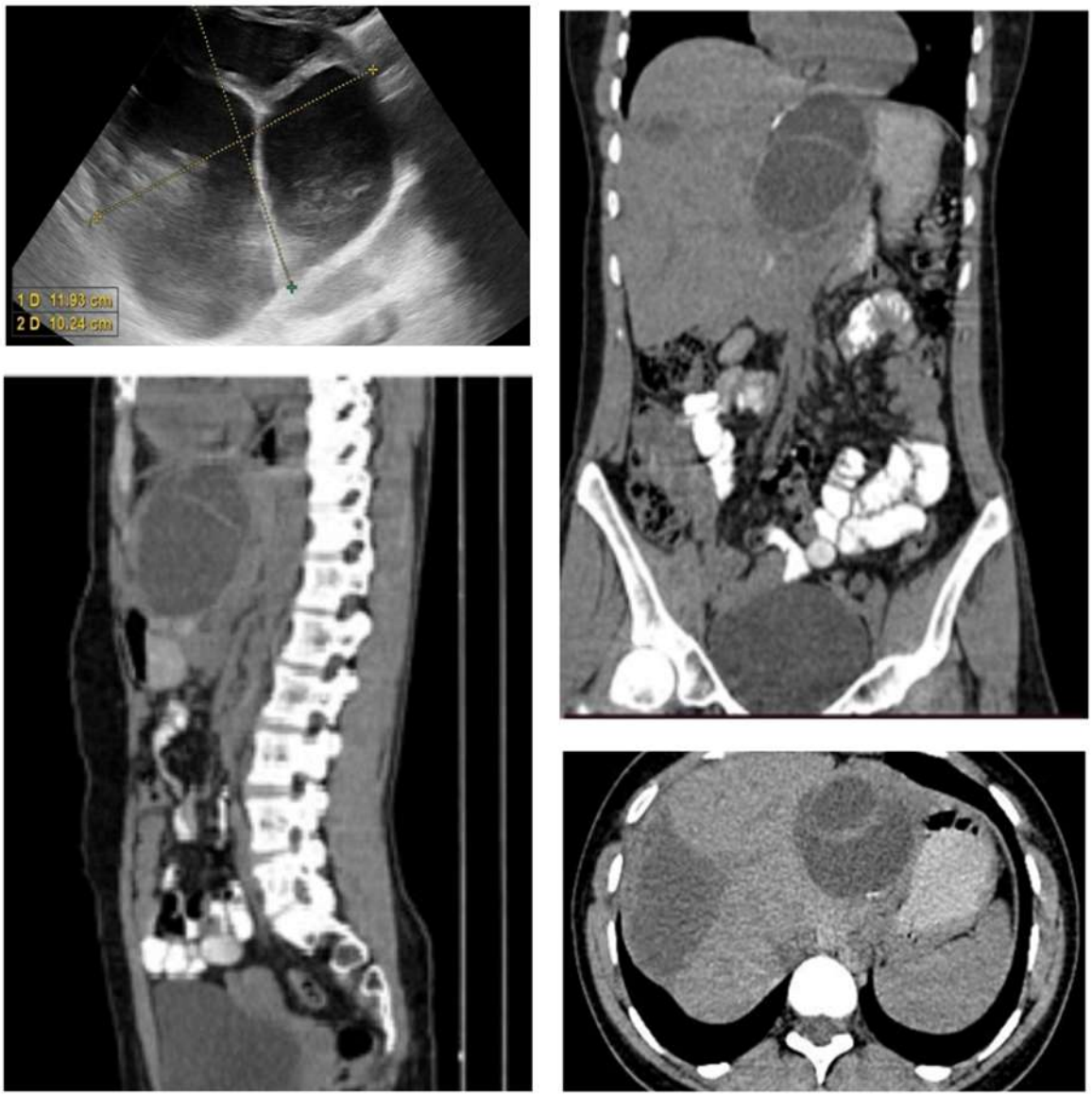
The patient recovered from treatment with Tab. Albendazole 400 mg was given twice a day for 4 months.

Figure 3: (a) Ultrasonographic view of Liver Hydatid Cyst. (b) CT Coronal View. (c) CT Saggital View. (d) CT Axial View



Case 4: Brain hydatid

72-year-old Patient presented with seizures and had complaints of Headache, Vomiting, and gradual progressive right hemi paresis for 3 months, and visual field disturbances in the left eye. NCCT brain was done which revealed well defined low attenuation cystic lesion measuring 38 x 33mm in size involving the left frontoparietal region with adjacent mild edema. No internal soft tissue components/septations/calcification was noted. Differential diagnoses include – Hydatidcyst, Intraparenchymal arachnoid cyst, Neuroglial cyst, Cystic neoplasm like pilocytic astrocytoma, pyogenic or fungal abscess, and porencephalic cyst.

Further MRI brain contrast was planned, which revealed a well-defined T1 hypointense / T2 hyperintense cystic lesion measuring 38 x 33mm involving the left frontoparietal region (Figure 4a,b,c). No evidence of restriction on DWI / susceptibility on GRE noted. No internal

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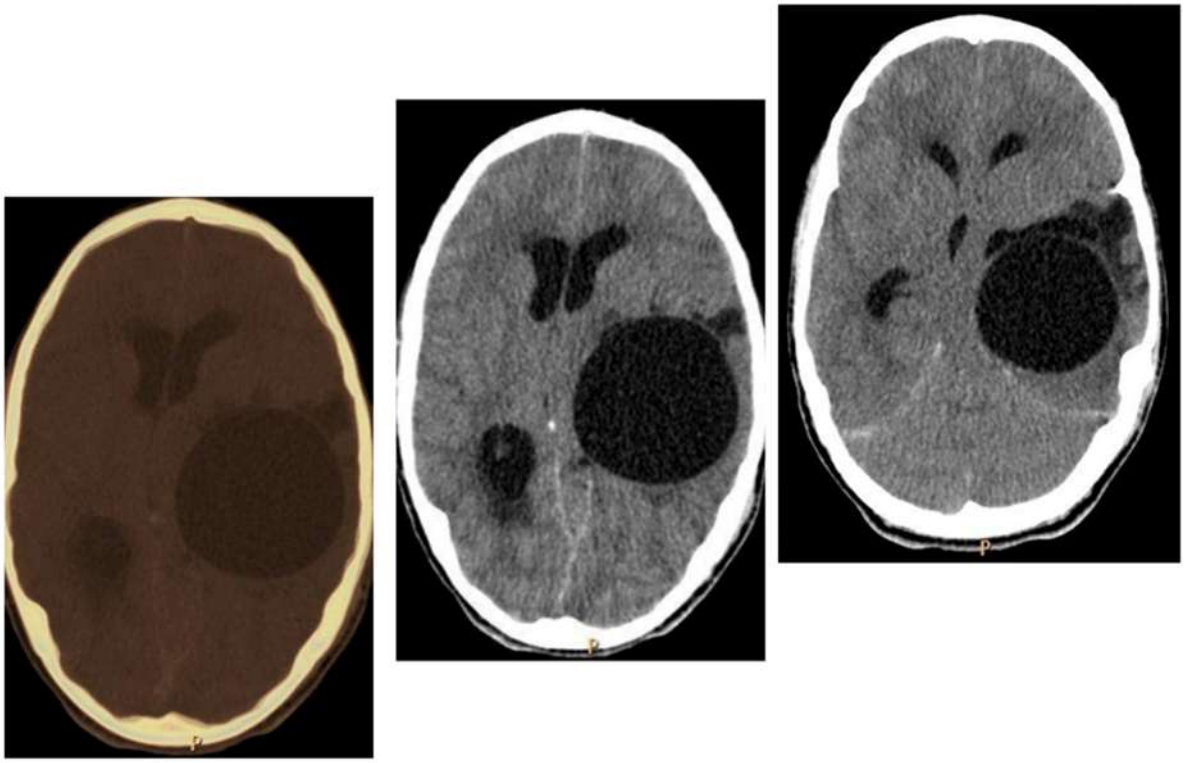
septations/daughter cysts were seen. Mild perilesional T2/Flair hyperintense vasogenic edema was noted. No midline shift was seen. Histo pathological study (H and E) of the cyst wall showed germinal epithelium and scolices attached to it and that confirmed the diagnosis of brain hydatid cyst. Further management was done under a neurosurgeon. The patient underwent craniotomy and cyst excision after routine investigations and anesthesia clearance.

Figure 4: MRI Brain



Case 5: Renal hydatid

A 56 year old patient presented with vague abdominal pain. In physical examination, low blood pressure and pervasive tenderness of the left lower quadrant of the abdomen were found, and the patient did not have any apparent history Urine analysis shows a little mucus, epithelial cell, and a high amount of RBC. Sonography (Figure 5a) of the right kidney showed a normal dimension of 121 mm and parenchyma thickness of 23 mm, without any stone or obstruction. Left kidney showed a dimension of 165 mm and a parenchyma thickness of 18 mm, and one typical cyst with a dimension of 120 × 93 mm was seen. Differential diagnoses includes: Complex left renal cortical cyst – Bosniak grade 3, Cystic renal neoplasm, and Hydatid cyst. A computed tomography scan (Figure 5b,c,d) illustrated a subtle peripherally enhancing cyst measuring 120 × 93 mm involving the mid pole of the left kidney. Few internal daughter cysts were seen. No enhancing soft tissue component was noted. Diagnosis of likely hydatid cyst was made. Differential diagnoses include – cystic renal neoplasm. Partial nephrectomy and cystectomy were scheduled for the patient,(fig 5e) and the specimen was sent to the pathology department. Results showed the definitive diagnosis of hydatid cyst and the protoscoleces were seen in the tissue section. After the surgery, the patient was discharged from the hospital without any symptoms. After diagnosis, the patient received Tab. Albendazole 400 mg twice a day for 4 months. The patient was followed up for 3 months, every 2 weeks by ultrasound, liver test, urine analysis, CBC, and platelet count test to look for any

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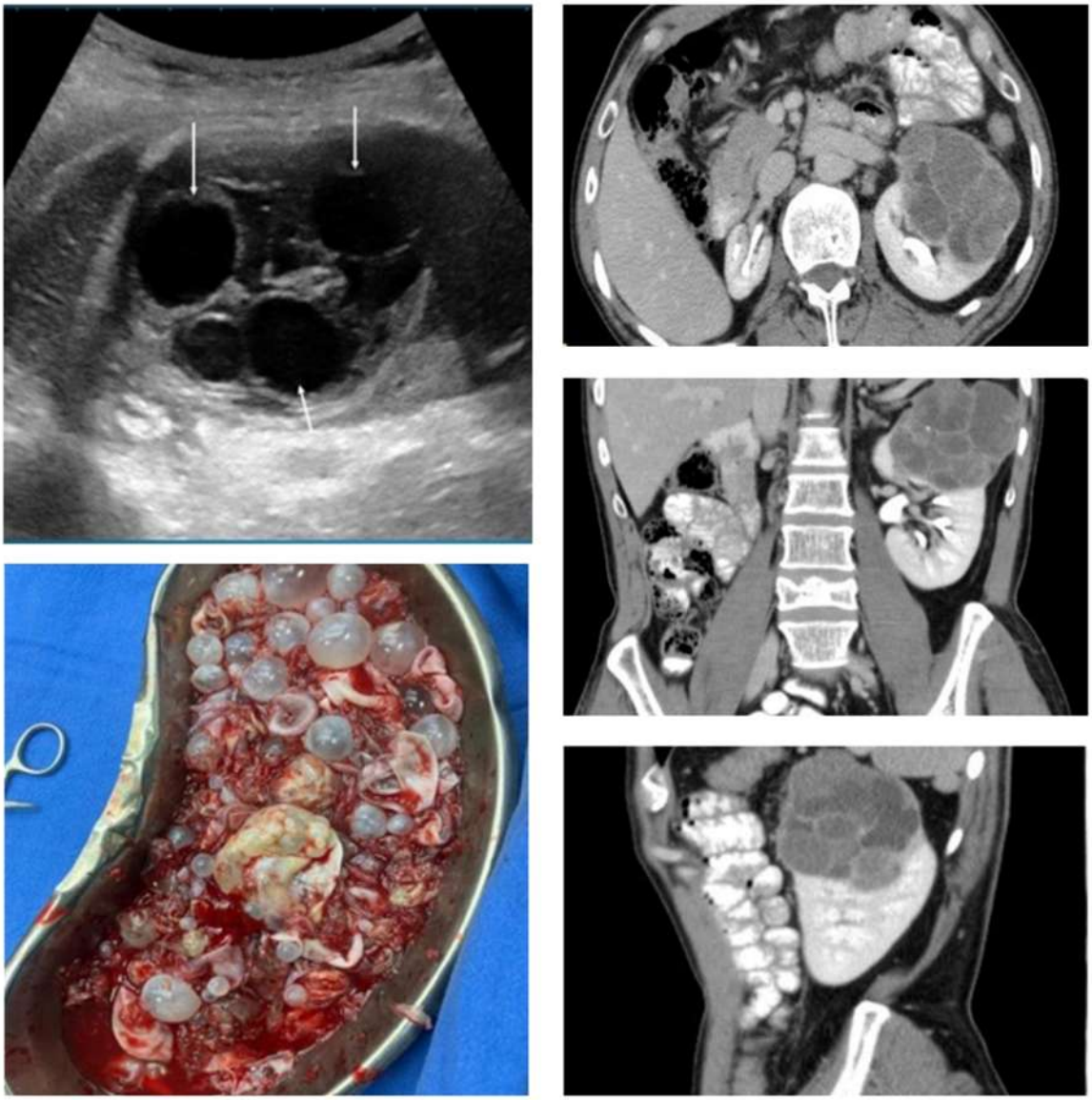
cysts. Afterward, the patient was followed up monthly for 1 year, and no recurrence was observed.

Figure 5: (a) Ultrasonographic View. (b) CT Axial view. (c) CT coronal view. (d) CT saggital View. (e) Postoperative image of specimen



Case 6: Lung and Liver hydatid

(lungs are the second most frequent site of hematogenous spread in adults and probably the most common site in children)

A 46year old patient presented with pain in the right hypochondrium for the last 15 days along with chest pain, hemoptysis, breathlessness, expectoration, cough, and fever for the last 1 week. He also complained of a salty taste in his mouth. Peripheral blood smear showed leukocytosis, eosinophilia, and raised erythrocyte sedimentation rate being nonspecific parameters observed in patients with this infection. The X-ray (Figure 6a) revealed well defined radiopaque lesion in the right middle lobe of the lung. CECT Whole abdomen (Figure 6b,c,d) was done which revealed a well-defined fluid attenuation cystic lesion measuring 98 x 99 x 104mm (AP x TR x CC) seen involving segments IV-A and IV-B of the liver. Minimally prominent left lobe IHBR noted. No internal septations or daughter cysts are seen, suggestive of liver hydatid cyst. HRCT chest was

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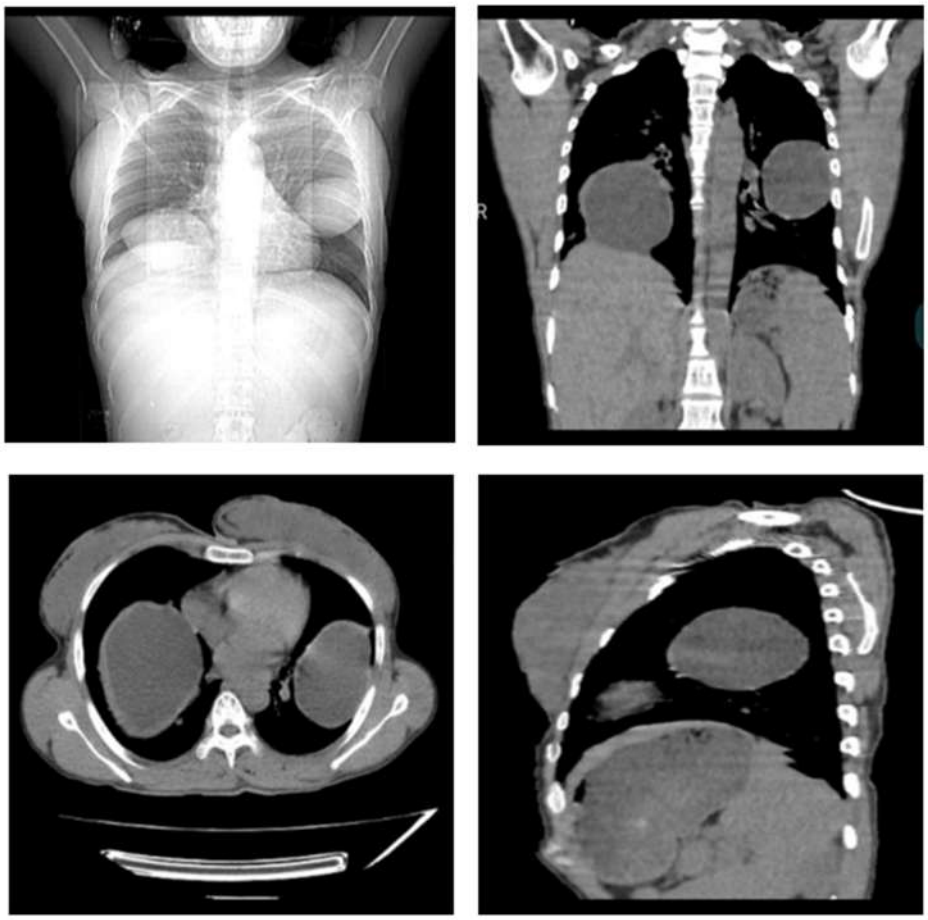
done and a note is made of two similar lesions measuring 59 x 67 mm in the superior lobe in lingular segment measuring 86 x 35 x 76 mm (AP x CC x TR) seen in the right middle lobe of the lung, suggestive of lung hydatid cyst (Figure 7a,b,c). Aspirated fluid was sent to the Microbiology and pathology departments for investigations. The reports included an Acid-fast bacillus stain which showed protoscolex and it confirmed the diagnosis as hepatic hydatid. Differential diagnoses include lung abscess and neoplastic etiology - Tab. Albendazole 400 mg was given twice a day for 4 months. Enucleation (Ugon method) was done as a part of surgery for lung hydatid and PAIR was done for the cyst in the liver using absolute alcohol.

Figure 6: (a) Chest X-ray. (b) CT coronal view. (c) CT Axial View. (d) CT saggital view



Case 7: Mediastinum hydatid

A 55-year-old male patient presented with dyspnea, dry cough, and chest pain on and off type for the last 4 years. Rapid breathing pattern noted on general examination. Blood investigations showed leukocytosis, eosinophilia, and raised erythrocyte sedimentation rate. Differential diagnoses include thymoma, teratoma, mediastinum abscess, and pericardial cysts. HRCT (Figure 8a,b) was done for this patient which showed a cystic subtle enhancing lesion in anterior mediastinum . Lesions shows multiple internal daughter cysts. No peripheral calcification is seen. Posteriorly lesion is indenting on left ventricle of heart. Differential diagnosis include - cystic germ cell tumour, cystic thymoma.

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Figure 7: (a) CT Coronal View. (b) CT axial View. (c) CT Saggital View

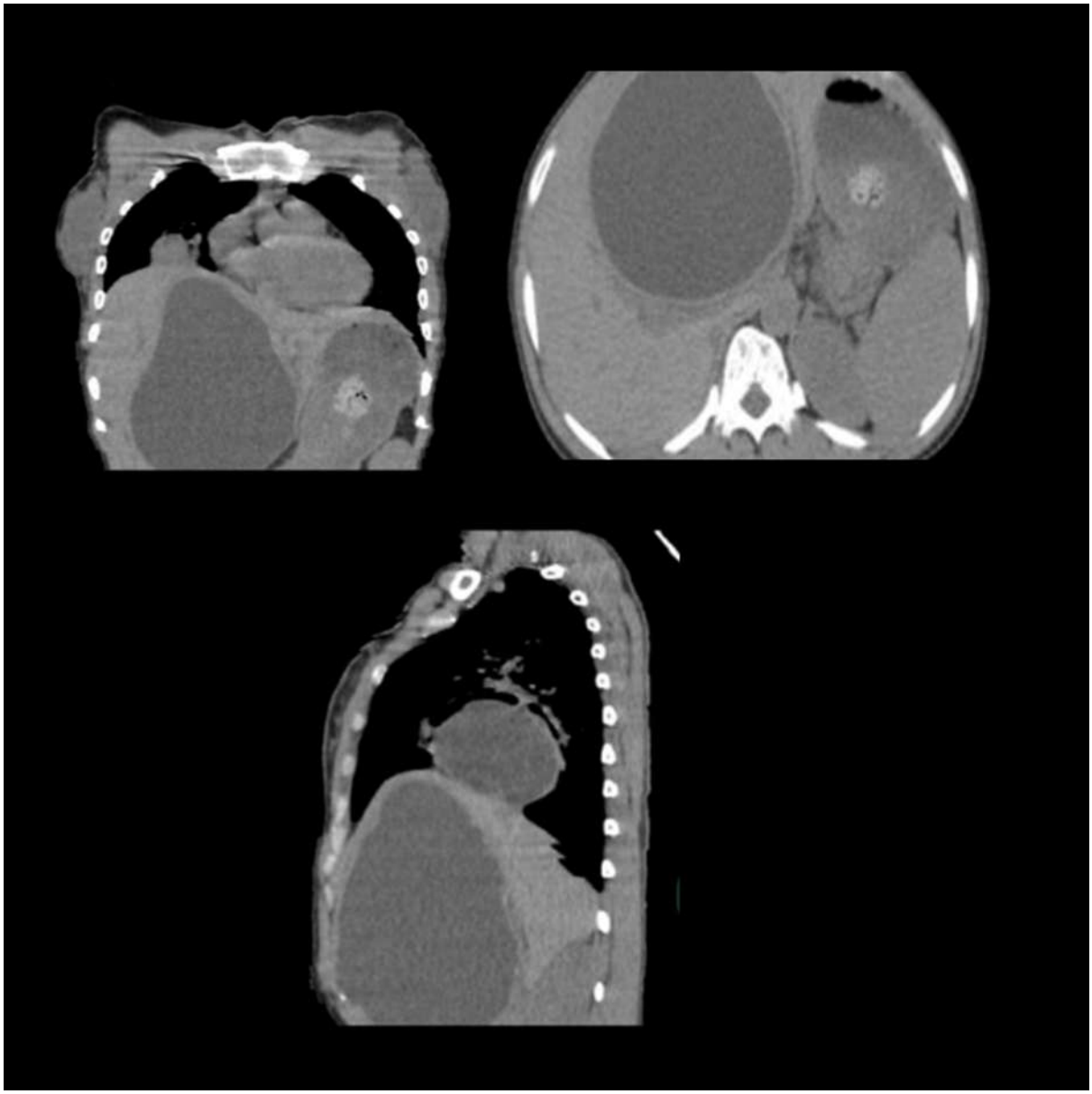

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Figure 8: CT images showing cystic lesion in mediastinum.

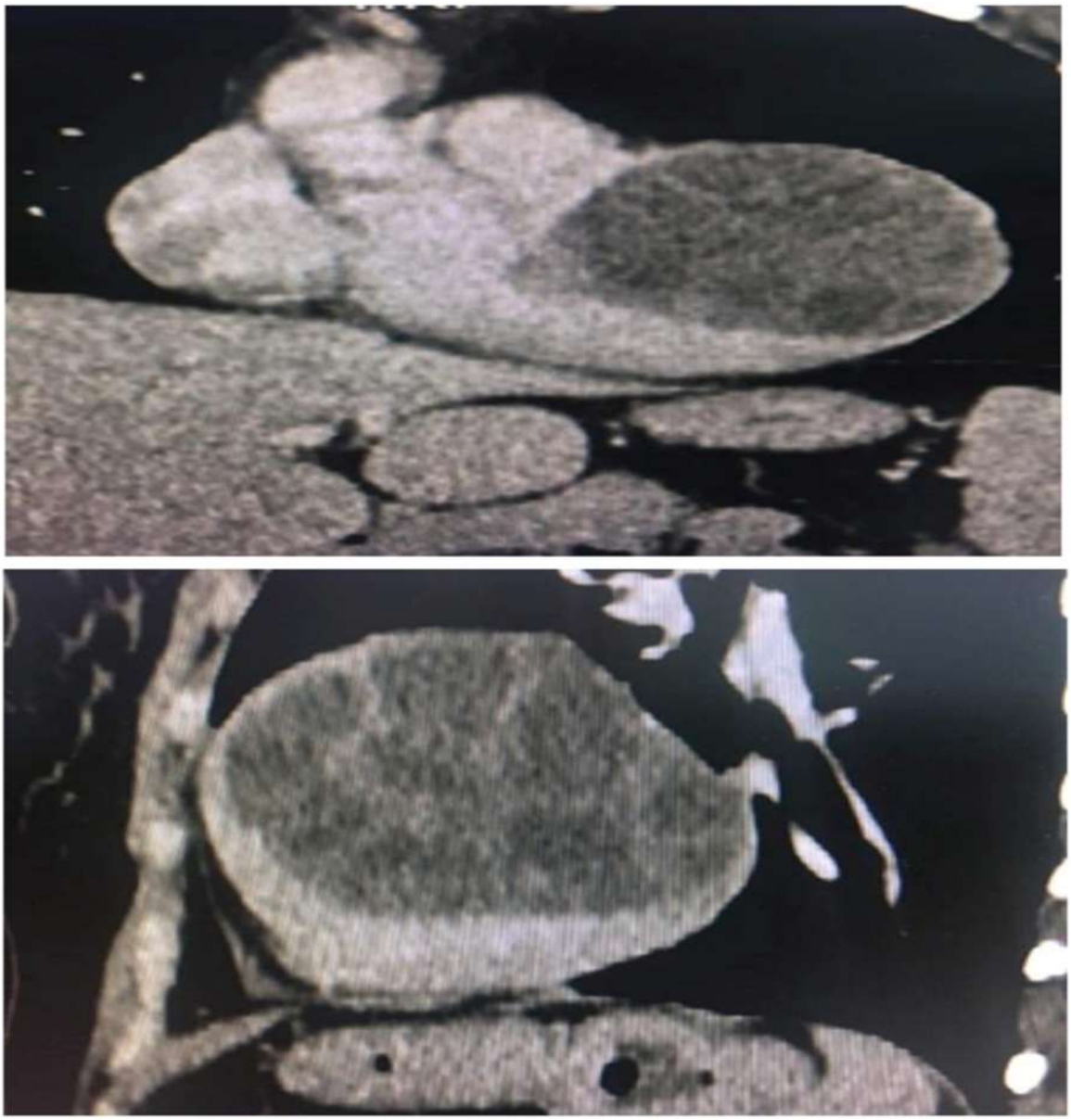

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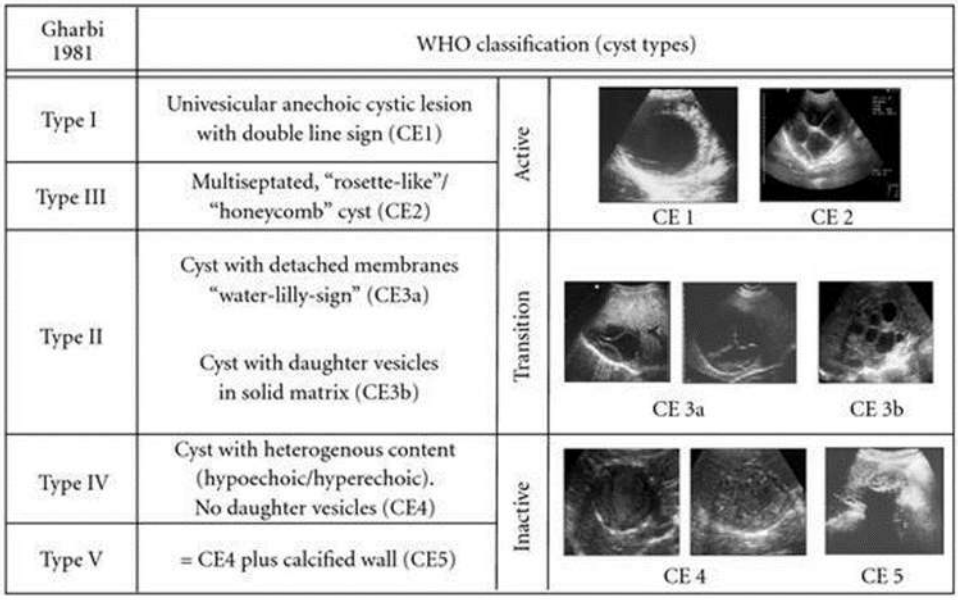
Figure 9: Gharbi classification of Hydatid Cyst



Discussion

From a clinical point of view, manifestations of hydatidosis in humans are variable, most patients seem to tolerate the infection for extended periods without any symptomatology, or they may suddenly show dramatic and acute symptoms [15]. All four patients made an excellent recovery following the surgical excision of the entire cystic masses. Adjunctive Albendazole chemotherapy 10 mg/kg/day was prescribed for 4–6 weeks before surgery to sterilize the cyst and for 2 months postoperatively to reduce the recurrence rate. Localization of hydatid disease at unusual sites usually poses a serious diagnostic challenge. Hydatiddisease in extra-hepatic locations usually follows a silent clinical course unless it grows and produces pressure symptoms or develops complications which may include local pressure, rupture, secondary infection, and allergic reaction. Clinical signs and symptoms are nonspecific and never pathognomic of hydatid cyst.

Routine laboratory tests can only reveal eosinophilia. Diagnosis is established by imaging, serological tests, and histopathological examination.USG is a cost-effective imaging technique but when available, a CT scan is superior owing to its higher sensitivity. Serologic tests are also very useful and usually involve a screening test such as ELISA and Indirect hemagglutination assay. The sensitivity of serology is high (80-100%) for liver cysts, but low for lung (50-56%) and other organs (25-56%). Gold standard treatment is the total excision of the cyst, avoiding its rupture and spillage to prevent any spread of hydatid with subsequent secondary echinococcosis; the anaphylactic shock is also a major risk connected.

Despite major advances in our understanding and treatment of hydatidosis over the past two decades, control of this zoonotic disease remains a challenging endeavor. Fortunately, because of the high risk of infection, the high morbidity rate, and the unpredictable outcome of hydatid infection, there is an increasing realization in international health agencies that hydatidosis is an important disease that causes life-threatening morbidity.

Through this study, we have shared our experience of diagnosis and successful management of hydatid cysts. All patients with hydatid cyst surgery should receive albendazole (10 mg/kg/day) for 6 months to prevent the recurrence of the disease. The risk of recurrence is as high as 11% if anti-helminths are not prescribed post-surgery. The postoperative follow-up consists of clinical examination, liver function tests, and chest X-ray once a month for the first 3 months which is then continued every 3 months till the end of the first postoperative year. Surgical removal of the cyst should be taken very meticulously to avoid its spillage on the surgical field as it may lead to a life-threatening anaphylactic reaction as well as

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incomplete extraction of the cyst leading to a risk of recurrence. Prophylactic measures like community education initiatives and proper hand hygiene after contact with animals like dogs are essential preventive measures. There is a table description (below) of Gharbi classification of hydatid cyst by WHO. (Figure 9).

Conclusion

From our study, we tried to establish that hydatid cysts can be found in all parts of the body and this should always be considered in the differential diagnosis of cystic lesions or unidentified tumor formations, especially in patients from endemic areas.Imaging features highly suggested hydatid cyst and detection of the complications. Postoperative longterm follow-up is also essential to detect any late complications such as local recurrence of the disease and development of hydatid cysts at the usual primary sites.

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