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RESEARCH ARTICLE

INTRAPLEURAL RUPTURED HYDATID CYST OF LIVER PRESENTED AS EOSINOPHILIC PLEURAL EFFUSION: CASE REPORT

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Abstract

Eosinophilic pleural effusions (EPE) are rare with an estimated incidence of 7.2% of all pleural effusions. EPE due to echinococcosis is exceptionally described in the literature. Pleural echinococcosis is almost always secondary to a ruptured primary lung cyst. Intrathoracic rupture of hepatic hydatid cyst (HC) is a rare clinical entity; the rupture into the pleural cavity is less common than in the bronchial tree. We report a case of a 72 year old patient, presented with dyspnea, the chest X-ray showed a right-sided pleural effusion. The initial diagnostic process included two thoracentesis which yield eosinophilic pleural effusions. A transthoracic ultrasound showed cystic formations in the pleura, in addition to an abdominal collection with classic ultrasound images of "water-lily" sign and "serpent sign". CT features suggested ruptured hepatic hydatid cyst in the pleura. Surgical intervention was indicated with postoperative chemotherapy.

Introduction:-

Eosinophilic pleural effusion (EPE) is defined as pleural fluid with 10% or more eosinophils [1]. It is uncommon with an incidence of 7.2% of all pleural effusions [1]. It can be associated a with a wide range of underlying causes, including in order of frequency: malignancy (34.8%), infections (19.2%), unknown (14.1%), posttraumatic (8.9%), and divers (23%) [2]. Parasitic causes are very rare, particularly pleural echinococcosis [3-4]; it is exceptionally described in the literature. The primary pleural hydatidosis is very rarely reported, the majority of reported pleural hydatidosis cases are secondary to lung involvement. Intrathoracic rupture of hepatic hydatid cyst is a rare clinical entity. The rupture into the pleural cavity is less common than in the bronchial tree [5]. We reported a case of hepatic hydatic cyst ruptured into the pleura and revealed by in EPE.

Observation:-

A 72-years-old man with type 2 diabetes, presented with dyspnea, and a dry cough dating back to a month before. Physical examination revealed dullness on percussion and absent breathing sounds and vocal vibrations on right-sided lung field.. The chest X-ray revealed a total opacification of the right hemithorax (Figure1). Full blood count

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showed a leukocytosis of 10920/mm3, lymphocytes of 2566 elts/mm3 and eosinophils of 2675 elts/ mm3. The thoracentesis revealed straw-colored fluid of which the analysis was of exudative nature, with an eosinophil predominance (49%), lymphocytes (22%) in the leukocyte differential count. Total protein value of the pleural fluid was 45 grams. A second thoracocentesis confirmed the eosinophilic character of the pleural effusion with an absolute leukocytes count of 100 / mm3, with 63% of eosinophils, 27% of polynuclear neutrophils, and 10 % of lymphocytes.

Direct examination of pleural fluid for acid-fast bacilli was negative, as well as TB nucleic acid sequence amplification.Two pleural biopsies were performed, that were inconclusive.

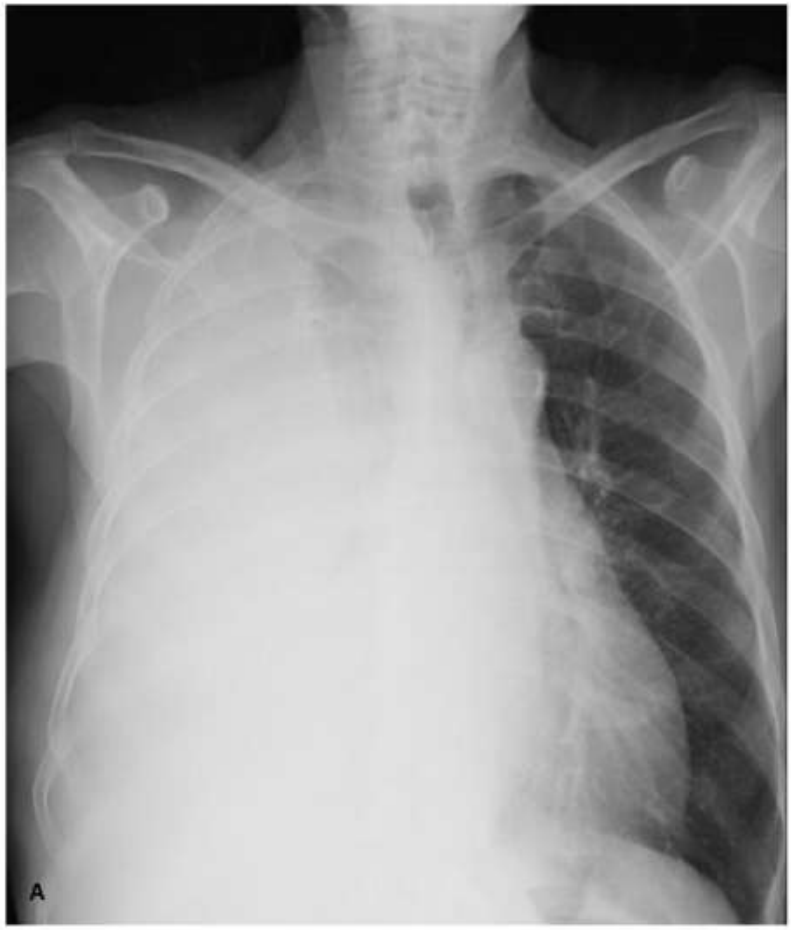

Figure 1:- Patient's chest X-ray: a complete opacification of hemithorax on the right side.

Thoracic ultrasound showed multiple cystic formations, with floating membrane realizing an "water lilly sign" (figure 2 A), the abdominal ultrasound revealed two cystic lesion within the liver with detachment of the endocyst realizing a serpent sign suggestive of hydatid disease (figure 2 B), which was later confirmed by abdominal CT scan that showed two multiseptale lesions, one at the hepatic dome (123 x 96 x 90 mm) and the second straddling segment VI and VII (123 x 123 x 128 mm). The one in the dome communicated with the large right pleural cavity via a breach (6mm), thus feeding a right pleural effusion of great abundance (figue 3).

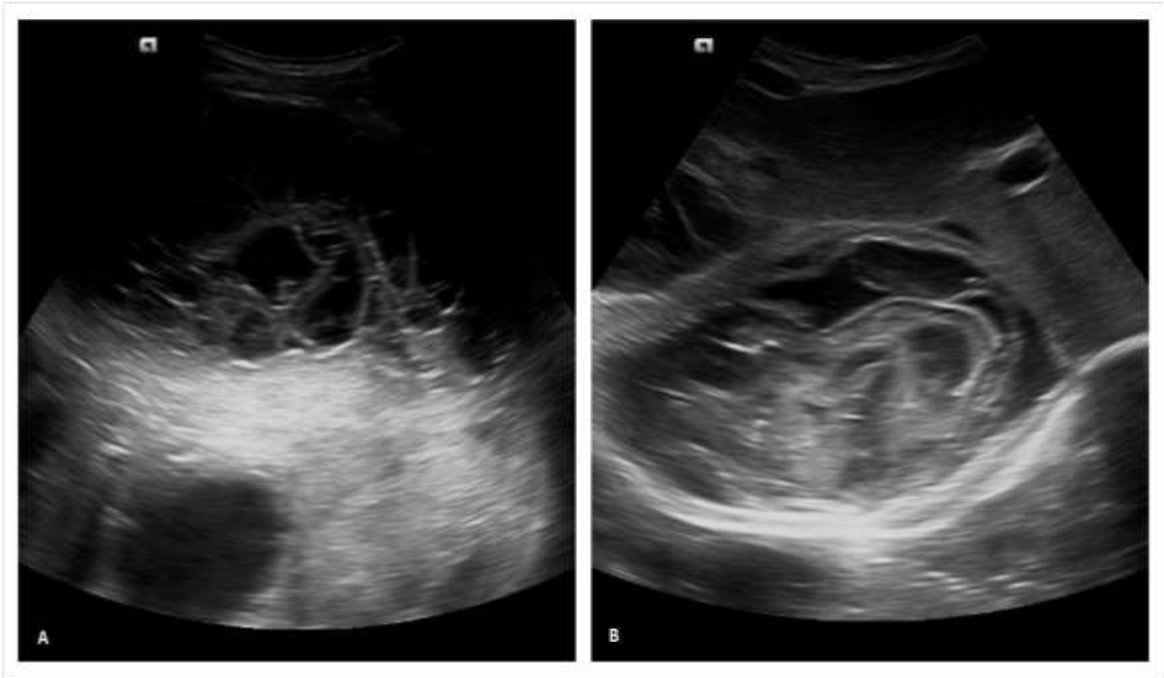

Figure 2:- A thoracic ultrasound: multiple cystic formations filling up the entire right pleural cavity. B: abdominal ultrasound demonstrates the detached folded endocyst.

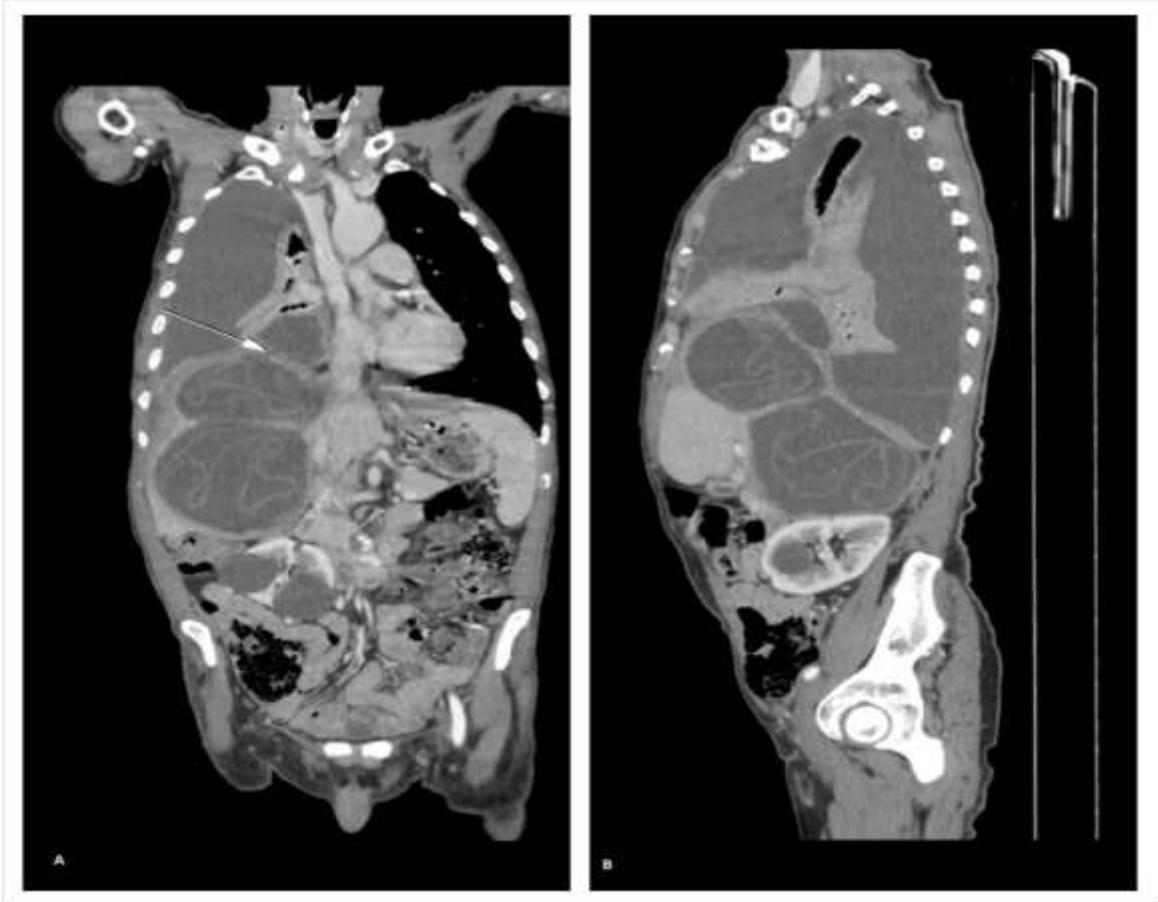

Figure 3:- CT scan (axial and coronal sections) showing two hydatid cysts, one at the hepatic dome communicated with the large right pleural cavity via a breach (6mm), thus feeding a right pleural effusion of great abundance.

The patient then underwent surgery: A posterolateral thoracotomy was performed, the pleural cavity was opened, a yellow-colored fluid was drained, and it was filled up with too many laminated membrane and daughter cysts. The pleural cavity was completely evacuated then repeatedly washed, the lung was trapped. After surgical decortication, the collapsed right lung was fully re-expanded. Then phrenotomy and cystectomy of the two liver cysts was performed, without visualization of a biliary fistula (figure 4).

As a postoperative follow up, chest X-ray was performed and the lung was fully re-expanded. The patient was discharged from the hospital after 6 days with the indication of continuing his medical treatment with Albendazole for 1 year.

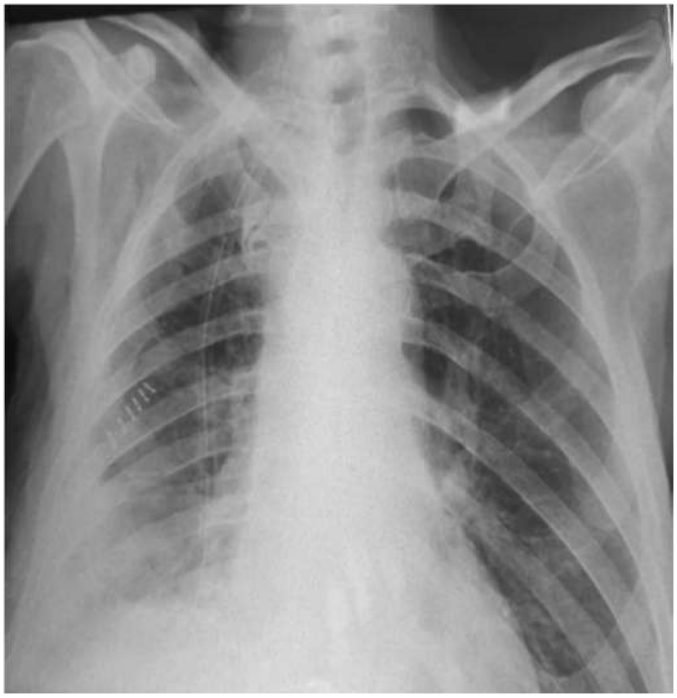

Figure 4:- Patient's chest X-ray 3 days postoperatively, a fully re-expanded right lung.

Discussion:-

Cystic echinocccosis (CE) also known as hydatid disease is an anthropozoonosis caused by Echinococcus granulosus. It is still endemic in developing countries [6]. Liver is the most commonly affected organ; the lungs are the second most frequent location of hematogenous spread in adults. Other organs are less commonly affected [7].

Intrathoracic rupture of hepatic hydatid cyst is rare, with a reported frequency of 0.6 to 1.6% in adult population [8]. The rupture into the pleural cavity is less common than in the bronchial tree [9]. This complication results from the long evolution of a hydatid cyst of the hepatic dome. The mechanical compression exerted by the cyst on the diaphragm leading to ischemia and muscles erosion.

Pleural hydatidosis can mimic the clinical presentation of pleural effusions such as dyspnoea, chest pain, dry cough, as it can be asymptomatic. Chest radiography is the primary method to evaluate pulmonary HCs because of its common usage, but it is inadequate for assessment of complications and spread, and for diagnosis of intrathoracic extrapulmonary locations, as pleural hydatidosis. In our case it shows a right pleural effusion, of wich the thoracocentesis showed eosinophilic pleural effusion. As an underlying cause, we thoughts of malignant causes, we also thought of TB as we are in endemic country. So, two pleural biopsies were performed, as well as tuberculosis testing. Then thoracic ultrasound was performed and it revealed suggestive aspect of hydatid disease.

Ultrasonography imaging is the gold standard for the diagnosis of hydatid liver cysts. Some radiological signs are pathognomonic, such as the presence of daughter cysts, water-lily sign which is the result of collapsed membranes floating on the surface of the cyst, and snowflake sign or serpent sign which is a result of the detachment of the endocyst. There are various classifications of sonographic appearance of HC, the first and most commonly used

being proposed by Gharbi in 1981 [10]. In 2001, the World Health Organization proposed another US classification based on the active-transitional-inactive status of the cyst as suggested by its sonographic appearance [10]. In this classification, six cyst stages have been assigned to three clinical groups (figure 5).

Therefore, in pleural hydatidosis ultrasound might reveal suggestive aspects, in our case ultrasonography exam showds multiple cystic formations filling up the entire right pleural cavity with “water lilly sign” suggestives of hydatid disease.

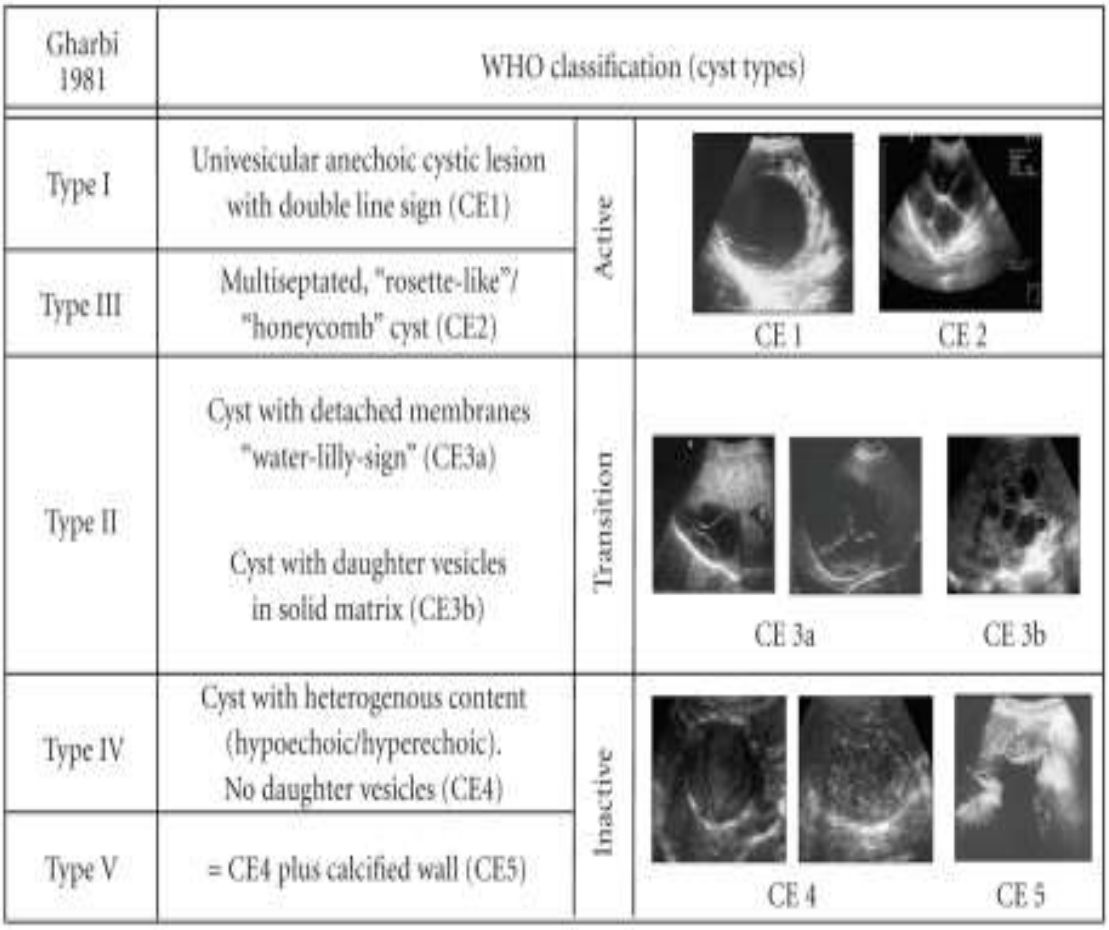

Figure 5:- Who Us Classification Of Hydatid Cysts [11].

CT would be useful when the involved area is inaccessible for ultrasound, and is indispensable for evaluating lung and bone. It can demonstrate features, size, number, and the presence of calcifications in the cyst; unenhanced CT is the modality of choice to assess calcifications. It also plays a key role in the assessment of complications. Furthmore CT can play a role in follow up and evaluating efficacy of treatment by demonstrating changes in the size, number, and shape of HCs [10, 12].

MRI has been accepted as superior to CT in many circumstances; MRI cholangiography is preferred in complicated cases of communication or rupture into the biliary system. As well It is useful in evaluation and depicting its close connection with adjacent structures.

On top of that it provides information about the internal structure of HCs; MRI with a T2-weighted sequence is better than CT for characterizing the internal structures of echinococcal cysts, reproducing better the ultrasound-defined features of HC. MRI may, also, be indicated when US is insufficient and CT is contraindicated [10, 12].

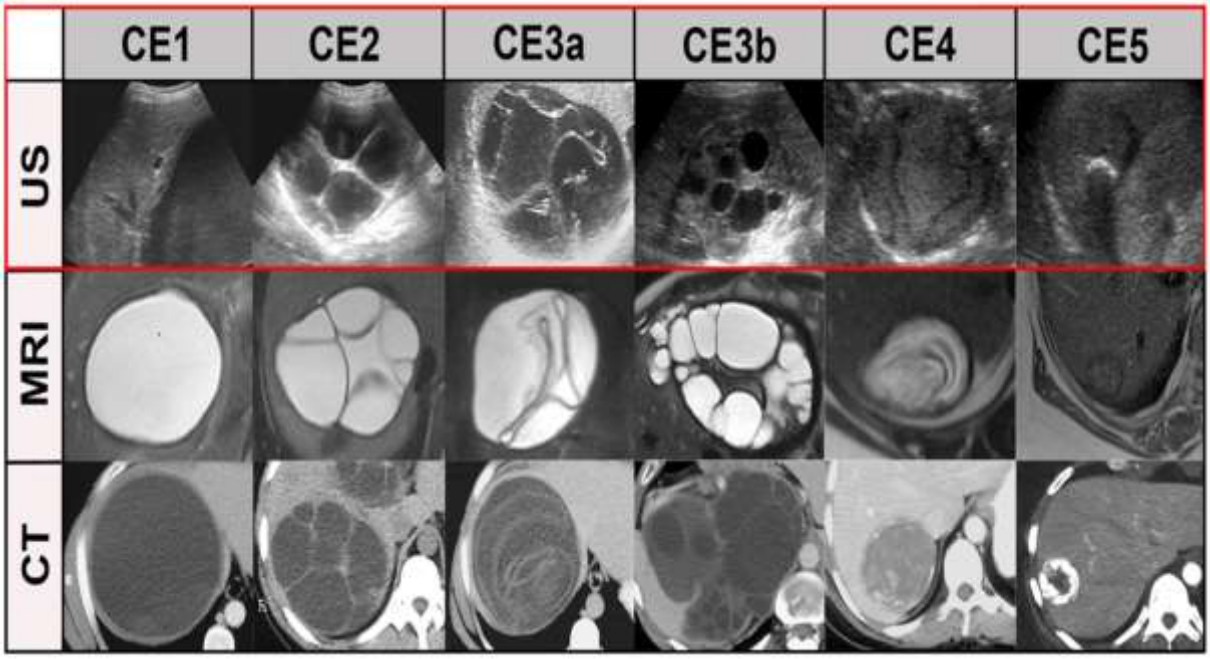

Figure 6:- Classification of hydatid cysts: How Do CT and MRI Perform in Comparison to Ultrasound? [12].

The Diagnosis of hydatid disease is based on imaging studies, such as Ultrasound, Ct-scann, or MRI. In suspected cases, hydatid serology by ELISA with compatible lesion identified by imaging, confirms the diagnosis.

The primary treatment of hydatid disease is surgical intervention. Nonetheless chemotherapy is used as a complement to surgical treatment, or performed in only inoperable cases. Thoracophrenotomy with cystectomy, subdiaphragmatic drainage, and thoracic decortication can be chosen as the surgical procedure in the management of cysts in the liver dome with pleural complication. We have followed the same approach with favorable evolution. A year later, there weren't any recurrence.

Conclusion:-

Pleural parasitic infestation **is** an extremely **rare pleural disease, it is** rarely responsible for eosinophilic pleural effusion, they are the subject of isolated clinical cases, and do not appear in the causes of the large European series, especially echinococcus granulosis. However, in an endemic country, in the presence of eosinophilic pleural effusion, secondary pleural echinococcosis must be evoked, especially since the diagnosis is easy, and imaging appearances are suggestive of diagnosis, and in most cases they are pathognomonics.

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