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RESEARCH ARTICLE

HYDATID PULMONARY EMBOLISM: AN EXTREMELY RARE COMPLICATION OF HYDATID DISEASE

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Abstract

The location of hydatid cysts inside the pulmonary artery is extremely rare. The cause is usually the rupture of intracardiac cysts. Only a few case reports of hydatid pulmonary embolism are reported in the literature.Herein, we describe the clinical case of a young 30-year-old patient, functionally asymptomatic, in whom multiple hydatid pulmonary embolisms were discovered on a CT scan which was done as part of the extension assessment of a hydatid cyst. We will discuss the pathophysiology as well as the management of this case. Recognition of the unique radiologic findings of hydatid cyst embolization to the pulmonary arteries is critical for prompt diagnosis and institution of appropriate therapy.

Introduction:-

Hydatid pulmonary embolism is a very rare manifestation of *Echinococcus* infection. Only a few case reports of this condition are reported in the literature.

Herein, we describe the clinical case of a young 30-year-old patient, functionally asymptomatic, in whom multiple hydatid pulmonary embolisms were discovered on a CT scan which was done as part of the extension assessment of a hydatid cyst. We will discuss thepathophysiology as well as the management of this case.

Recognition of the unique radiologic findings of hydatid cyst embolization to the pulmonary arteries is critical for prompt diagnosis and institution of appropriate therapy.

Case Report:-

This is the case of a 30-year-old man with medical past history of hepatic hydatid cyst operated 2 years ago, he underwent a conservative surgery for a liver cyst located at the fourth segment of the liver, with good immediate post operative results.

Two years later, during a pre-recruitment medical examination, a hepatomegaly was found without evidence of hepatic insufficiency, portal hypertension or heart failure signs. Clinical examination found a normotensive patient with a respiratory rate of 17/min. Lung auscultation and the remainder of the physical examination were uneventful.

Pertinent laboratory findings were a WBC of 11,600/µL with an elevated eosinophil count of 1,500/µL and elevated liver transaminases with ALT and AST of 88 and 53, respectively.

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CT scan was performed which revealed fluid attenuation filling defects in the two main pulmonary arteries extending into and completely obstructing the two right lower lobe arteries and segmental pulmonary arteries and extending into the left upper lobe and segmental arteries (figure 1) with peripheral wedge-shaped pulmonary consolidations corresponding to pulmonary infarction. Large hepatic cysts typical of hydatid disease of the liver were also visualized on images of the upper abdomen adjacent to and compressing the intrahepatic inferior vena cava (figure 2).

Transthoracic echocardiography did not find a cardiac localization of the hydatid cyst; the right ventricle was slightly dilated with preserved systolic function, hemodynamic evaluation did not reveal any signs of pulmonary hypertension, pulmonary artery systolic pressure was estimated at 29mmHg, and the inferior vena cava was thin and compliant.

The patient was diagnosed with hydatid pulmonary embolism secondary to intravascular rupture of hepatic hydatid disease.

After multidisciplinary discussion, a surgical procedure was rejected given the large number of the hydatid cysts, their extension into the distal divisions of the pulmonary arteries, and the significant operative risk for an uncertain result. The patient was discharged under treatment with albendazole at a dose of 400 mg twice daily with courses of 28 days separated by intervals of 15 days,followed by an evaluation every 6 months.

A clinical follow-up 12months later showed no deterioration of the clinical picture.

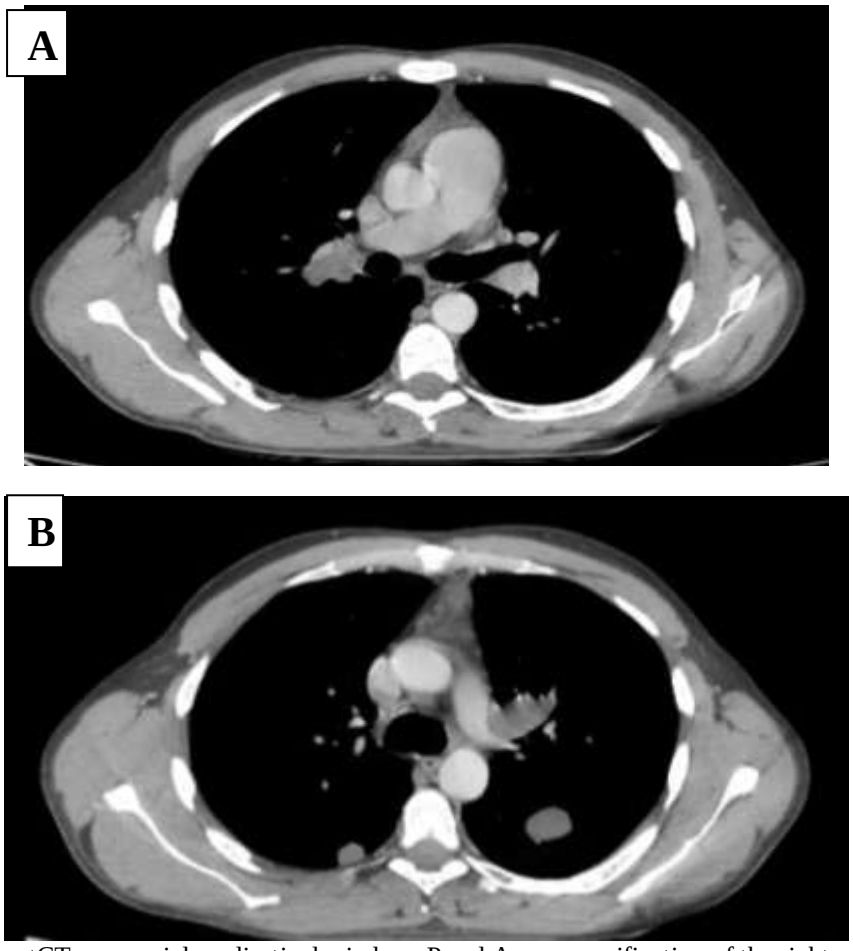

Figure 1:- ChestCT scan, axial mediastinal window: Panel A: nonopacification of the right main pulmonary artery and its branches. Panel B: intra-arterial cysts extending along the left lobar and segmental pulmonary arteries of lingula.

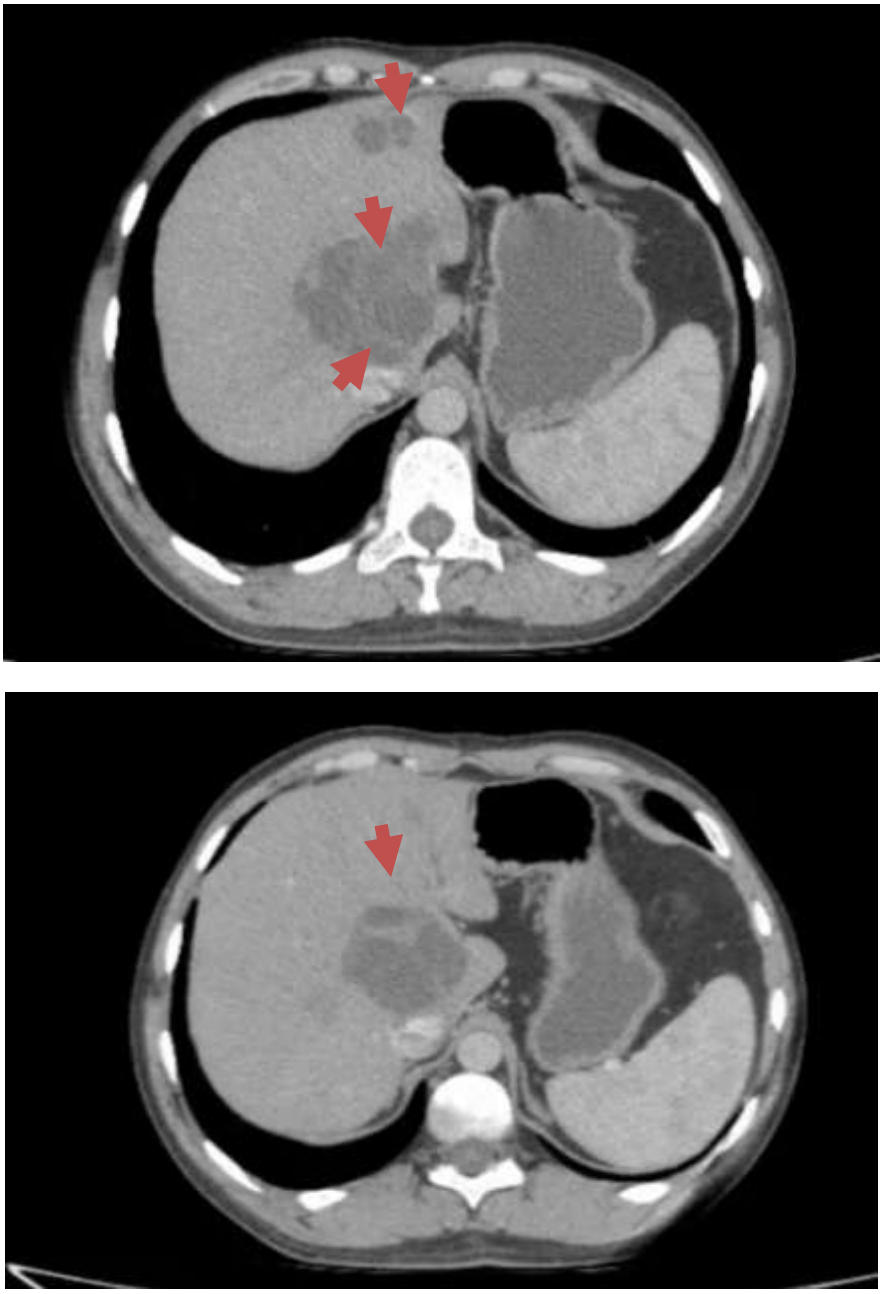

Figure 2:- Axial image through the upper abdomen shows a large, round, low-attenuation lesion in the liver with peripheral daughter cysts (arrows) characteristic of hydatid disease.

Discussion:-

Echinococcosis is a widespread zoonotic parasitic disease which has major medical and socio-economic costs for humans [1,2].

The causal pathogen, *Echinococcus granulosus*, is only known to infect dogs and wild canids as its definitive hosts, following the ingestion of cysts in ungulate viscera where the adult cestode develops. In the intestine, the tapeworm releases eggs that are passed into the environment during defecation. Herbivores act as intermediate hosts for the parasitic larval stage (metacestode) [3].

Human contamination occurs following the ingestion of E. Granulosis eggs through contaminated food, mainly vegetables and water [4] or by direct contact with contaminated dogs that retain eggs on their coat.

Onceconsumed, the hexacanth embryo is released from the egg, larvae hatch in the duodenum and enter the portal system, most often lodging in the liver (in 60–75% of cases). In approximately 15–20% of cases, larvae bypass the liver and travel to the pulmonary circulation in the lungs and have a relatively slow growth of 1–5 cm per year and are usually asymptomatic. If larvae traverse the lungs, they may lodge in any organ including the brain and heart.

Cardiac hydatidosis is uncommon and occurs in only 0.5–3% of cases. Although the left ventricle is most often involved (60%), cysts may be found in all parts of the heart (5).

A pulmonary-artery cyst may result from the rupture of hepatic or abdominal cysts into the hepatic veins or the inferior vena cava after spontaneous or iatrogenic fistula. In deed, the majority of cases of hydatid embolism described in the literature had a history of hepatic hydatid cyst liver surgery, or it developed directly from the migration of hydatic larvae after the rupture of a cyst in the right cardiac chambers resulting in thedissemination of protoscolices and the formation of secondary cysts in the infected tissues. This mechanism is infrequent. During the initial phase, the cyst causes an erosion of the endartery subsequently causing several aneurysms which increases the risk of hematogenous dissemination and exposes to massive hemoptysis by rupture in the bronchi (6).

Hydatid Pulmonary Embolism can cause long-term complications including pulmonary hypertension, chronic pulmonary heart disease, and bronchopulmonary destruction.

Cardiovascular hydatid cyst complications vary between 0.02% and 2%. (7) Only a few case reports of ruptured hepatic hydatid cyst in the IVC are reported in the literature.

In our case, we believe that the starting point of hydatid pulmonary embolism was the daughter cyst migration from the liver during the first operation, since, there was a hepatic hydatid cyst attached to the IVC.

Three types of clinical outcomes from pulmonary hydatid embolism have been reported in the literature: acute fatal embolism, sub-acute embolism resulting in pulmonary hypertension and death in less than 1 year, and chronic pulmonary hypertension. (7)

The cysts mechanically obstruct the blood flow usually without formation of any associated thrombi [8]. Cysts are slow-growing and pulmonary artery obstruction may remain asymptomatic as blood supply can be provided by the bronchial arteries. Eventually, progression of disease can result in dyspnea, hemoptysis, or anaphylactic shock from the leakage of hydatid cyst fluid due to trauma or surgery. [9].

Diagnosis of hydatid cyst can be made using a combination of radiological imaging and serological tests [10].

Echocardiography is the imaging method of choice for cardiac and pericardial cysts, but it rarely enables direct visualization of a pulmonary embolus. Transesophageal echocardiography may visualize massive emboli in the main pulmonary arteries [11].

On CT, the cysts in the arterial lumen appear as rounded intravascular masses with levels of fluid attenuation and smooth borders surrounded by normal lung tissue, with contrast enhancement at the periphery. This was the case in our patient. [12]

MR imaging of the involved vessels depicts oval-shaped lesions containing fluid with homogeneously hypointense signal intensity on T1-weighted images and homogeneously hyperintense signal on T2-weighted images. The cyst walls demonstrate low signal rims easily discernible on the T2weighted images [13].

The differential diagnoses for the observed lesions are pulmonary thromboembolism that can be excluded because of the lesion’s cystic appearance in CT and MRI while thromboembolism will show solid lesion appearance and primary arterial tumors.

The presence of hydatid cysts within the limbs or the liver (as in our case) makes the diagnosis much easier.

Surgical removal of the intra-arterial hydatid cyst under cardiopulmonary bypass, deep hypothermia and circulatory arrest is the therapy of choice. However, it exposes to the risk of rupture of the cysts during surgical manipulation which can lead to the dissemination of the contents of the cysts, and can produce an anaphylactic shock. (14)

The surgical procedure consists first of all in eradicating the embologenic cyst, hence to remove hepatic hydatid cysts located deep or within the neighbourhood of hepatic venous structures.Certain precautionary measures have been recommended. These include clamping of the inferior vena cava, avoidance of traction on the liver, and cavocaval bypass if necessary. (15)

Approaches to the treatment of this rare disease are not standardized, are underreported and must be personalized on a case-by-case basis. Two operational steps are proposed by Koksal et al. [16]: First, the removal of the intra-arterial cysts followed by a gesture on the affected lung either conservatively or by total pneumonectomy.

Antihelminthic medication such as benzimidazole is usually employed in the perioperative period to prevent infection from spillage of cyst contents during surgery, it could be administrated also in case of surgical contraindication, inoperable patient or disseminated hydatidosis, but its effectiveness is inconstant.

The place of anticoagulant therapy remains discussed in the literature and deserves to be further evaluated. More often it is systematically initiated, before confirmation of the hydatic nature of pulmonary embolism [18], while other authors have not seen interest in it and instead have indicated albendazole.

The best management of hydatidosis is preventive. It is based on individual prophylactic measures: avoidance of contact with dogs, systematic hand washing, careful washing of fruits and vegetables intended to be eaten raw. Collective measures are also necessary, such as improving slaughter conditions, veterinary control of meat, especially in rural areas, tackling illegal slaughter.

This case report highlights the fact that adults with a history of hepatic hydatid cyst should be followed regularly in order to act and avoid complications that may arise.

Conclusion:-

The location of echinococcal cysts in the pulmonary artery is extremely rare. The symptoms are not specific. Hydatid cysts within the pulmonary arteries have a characteristic appearance on both CT and MR imaging. Recognition of this disorder is essential to facilitate prompt diagnosis and treatment. The surgical treatment is stringent and the efficiency of the medical treatment is still controversial.

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