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RESEARCH ARTICLE
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A CASE OF SPLENIC HYDATID CYST -A RARE DIAGNOSIS.

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Abstract
Hydatid disease or cystic echinococcus is caused by a parasitic tape worm called as Echinococcus granulosus. The disease presents with the formation of hydatid cysts usually in the liver and lungs, however, an isolated splenic hydatid cyst is a rare occurence with a worldwide incidence of 0.5-4%. Herein, we present the case of a 16-year-old female with an isolated splenic hydatid cyst. The patient presented with chronic and nonradiating pain in her left hypochondrium. Ultrasonography showed a large cystic lesion with septations in the spleen which was confirmed with a Computerized tomography (CT) scan showing a cystic lesion in splenic parenchyma with numerous internal enhancing septations. She was successfully treated surgically with a splenectomy along with the cyst being excised completely intact. At six months follow-up the patient was asymptomatic.

Keywords: Spleen, Hydatid cyst, Echinococcus, Splenectomy, Vaccines

Introduction

Human echinococcosis (hydatidosis, hydatid disease) is a systemic zoonotic disease caused by larval stages of restores (tapeworms) of the genus Echinococcus. The disease is globally distributed and is more common in sheep and cattle raising areas of the world. Sheepdogs are the definitive hosts and sheep, cattle, goats etc are the intermediate hosts.[1]. Infection in humans occurs accidentally by the ingestion of parasite eggs excreted in dog feces through hand-to-mouth transfer. Primary splenic involvement by hydatid cyst is rare and accounts for less than 2% of patients [2]. Berlot was first to introduce Primary splenic hydatidosis in 1790 [3].

The liver acts as the first filter in trapping the embryos which then develop into hydatid cysts in 55 – 70% cases, followed by the lungs being 2nd filter in 18-35% cases. Incidence of unusual sites is about 8-10% and various sites are like spleen, psoas muscle, pelvic cavity, peritoneum, mesentery, brain, kidneys, bones, muscles and soft tissues. Hydatid cysts involving the spleen are rare (2.5%). Splenic hydatid, being a rare entity, can occur primarily or in association with hepatic, pulmonary or multi-organ hydatidosis [4].

The disease may be asymptomatic and discovered coincidently on postmortem or when an ultrasonography or a CT scan is done for some other condition. However, the main presentation of splenic hydatid disease is a slowly growing, asymptomatic cystic mass, although it may manifest as

a painful bulge in the left hypochondrium. Dyspnea and anaphylactic responses may occur due to cyst rupture. Spleenic hydatosis being a rare entity requires a high index of suspicion and differential diagnosis includes pseudocysts, epidermoid cysts, splenic abscesses, cystic neoplasms, and hematomas of spleen. Ultimately, the diagnosis is made by a combination of good history and clinical examination supplemented by serology and imaging.

Case Presentation

A 16 year old unmarried girl presented in the Surgical OPD with a history of pain in upper abdomen since 1 month, which was associated with intermittent vomiting and fever. The pain was localized to the left hypochondrium, gradual in onset, moderate in intensity, dull aching in character, progressive in nature and was associated with a feeling of heaviness in the left upper abdomen. There was no history of jaundice, bowel and bladder complaints. No history of known comorbidities. No significant past history. No history of contact with dogs or sheep.

On General physical examination- revealed a blood pressure of 110/ 70 mmHg, pulse 80 beats/min with no signs of anemia, jaundice, cyanosis.

Per abdomen examination- Soft, non-distended, an irregular soft to firm non -tender mass was present, moving with respiration and occupying whole of the left hypochondrium of about 7-8 cm from the costal margin. Bowel sound was present. The rest of the systemic examination was unremarkable. A clinical diagnosis of cystic lesion of spleen was made.

On investigations; TLC: was raised to 12,300; Eosinophil count was high (8%), haemoglobin (Hb): 13.4 g%, platelet count: 227000/cumm, total bilirubin: 0.26 mg/dl, direct bilirubin 0.09mg/dl, serum glutamic oxaloacetic transaminase 16 U/l, serum glutamic pyruvic transaminase 17 U/l, alkaline phosphatase 111 U/l, T. protein: 6.4 g/dl, albumin: 3.7, urea: 21 mg/dl, creatinine: 0.8 mg/dl, S. sodium: 131 meq/l, potassium: 4.3meq/l and chloride: 104meq/l. Indirect haemagglutinaton test for echinococcus was positive. X-Ray chest was normal.

Ultrasonography examination abdomen showed a large cystic lesion with septa (? Floating membranes) in the splenic parenchyma which was suggestive of spleenic hydatid cyst. A 1.3× 1.1 cm hypoechoic area was noted in left ovary - ? Ovarian cyst. Rest all organs were normal. Computed enhanced tomography (CECT) - showed a large well-defined cystic lesion of size 84 × 86 mm sewn in the sleenic parenchyma with thin separations, confirming the diagnosis of a splenic hydatid cyst. The patient was planned for an elective splenectomy before which she was advised Tablet Albendazole 400mg twice daily for 1 month in order to stabilize the cyst and decrease the chances of rupture during surgery.

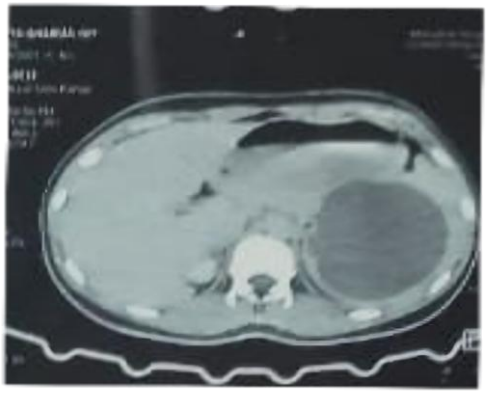
She was also prophylactically vaccinated against capsulated organisms (Pneumococcal, Meningococcal and Haemophilus influenzae) to prevent Overwhelming Postsplenectomy Infection (OPSI). Exploratory laparotomy was done and intraoperative findings revealed spleen with hydatid cyst of 5×4cm. Splenic vein and artery were identified and ligated and cut. Spleen and hydatid cyst was separated completely from the surrounding tissues extracted out en masse. Abdominal drain was placed and abdomen closed in layers. Postoperative stay was uneventful and she was discharged on Post-operative day 8 in a stable condition. In the postoperative period, the patient continued on tablet Albendazole for a month.

Histopathology report

Gross description:-

Received already cut open splenectomy specimen measuring 11x7x4.5cm. Cut surface shows a large cyst at medial part of spleen including hilum measuring 5.5x5.5x4cm, shows prominent vascular markings. Rest of the spleen shows normal splenic parenchyma. On cutting through cyst mucoid material came out. Wall thickness 0.1 to 0.5 cm.

Microscopic Description:- Multiple section examined reveal eosinophilic lamellated cyst wall with degenerated scolex. Moderate mononucleosis inflammatory infiltrate is seen in the cyst lining. Splenic parenchymal reveal thicken trabeculae with expansion of red pulp and normal appearing white pulp. Overall findings were in favour of hydatid cyst of spleen.



Figurre 1

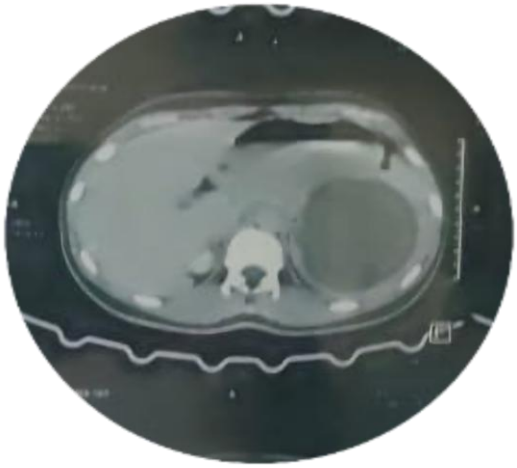


Figure 2

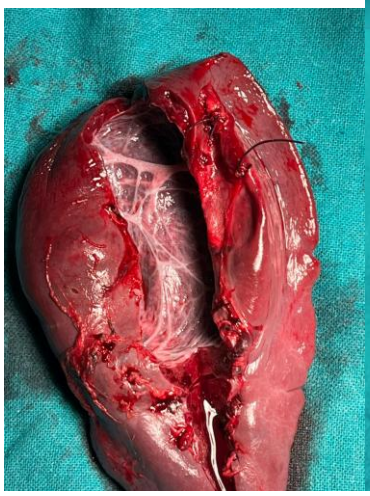


Figure 3.

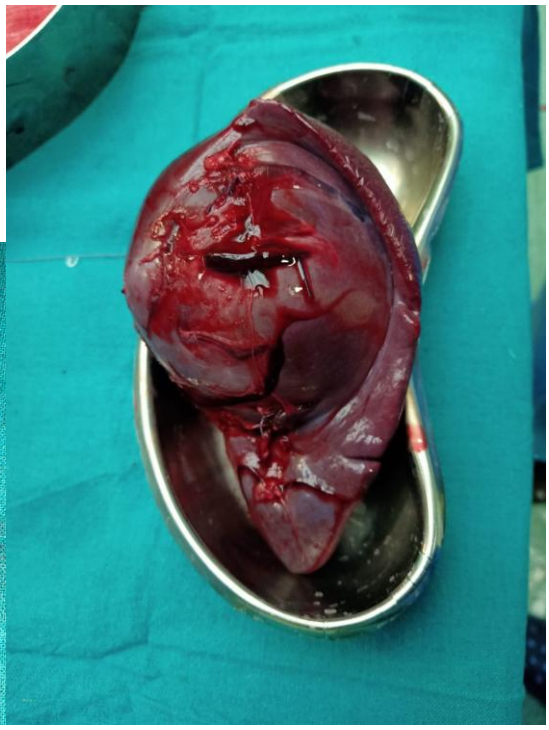


Figure 4.

Figure 1 & 2 - Preoperative CECT - showing the large splenic cyst

Figure 3 - Resected Spleen with hydatid cyst - cut section showing the visible daughter cysts

Figure 4 - hydatid cyst en masse with spleen.

Discussion

Hydatid disease is a serious public health problem especially in the endemic zones of Middle-east, Australia, South America and India. In India the annual incidence of hydatid cysts varies from 1 to 200 per 100,000 people. An isolated splenic hydatid cyst is an extremely rare manifestation, even in endemic countries, with a worldwide incidence rate of 0.5%-4% [5].

The mechanism of primary splenic involvement is either through the splenic artery after bypassing the lung and liver or through the splenic vein by retrograde involvement [2]. The cyst is characterized by three layers: an outer pericyst, which is derived from compressed host organ tissues; an intermediate hyaline ectocyst, which is non-infective; and and inner endocyst, which is the germinal membrane and contains viable parasites that can separate, forming daughter cysts. The patient may be asymptomatic for long time or can present with non - specific dull aching left hypochondriac pain or mass, hypersplenism, dyspepsia, heart burn, constipation and left sided portal hypertension, rupture or fistula formation to colon [2] ,[6-8]. Differential diagnosis for splenic hydatidosis include cystic spleen lesions like abscesses, epidermoid cysts, hematoma, post-traumatic pseudocysts, and neoplasms such as lymphangioma and hemangioma.

Investigations show a raised Eosinophil count; serological tests, such as ELISA and immunoelectrophoresis, point towards the diagnosis. Radiological diagnosis through plain X-ray, ultrasonography (USG), CT, and MRI serves as a valuable method for identifying hydatidosis. In abdominal or chest radiographs, the presence of marginal or crumpled eggshell-like calcifications in the splenic region indicates potential splenic hydatidosis. Ultrasonography and CT scan are the investigations of choice. The CT scan shows a smooth space-occupying lesion with several septa.

Splenectomy is the conventional treatment, with the choice of either total or partial splenectomy depending upon the degree of involvement of splenic parenchyma [9]. Cyst fluid can be drained with puncture and aspiration to reduce the intracystic pressure, but splenectomy without puncturing the cyst is preferable [10, 11]. The World Health Organisation (WHO) recommends one month prior or four days preoperative treatment with tablet albendazole to reduce intraoperative and postoperative complications [12]. Other surgical approaches include cyst enucleation, and unroofing with omentoplasty.

Contamination of the peritoneal cavity at the time of surgery with active hydatid daughters should be avoided by continuing drug therapy with albendazole (10-15 mg/kg/day for one month) or mebendazole (40-50 mg/kg/day for 3-6 months). Additionally, praziquantel (40mg/kg/week for 2 weeks pre and postoperative) is administered to minimize the risk of anaphylactic shock and decrease tension in the cyst wall. This should be combined with packing of the peritoneal cavity with 20% hypertonic saline-soaked packs and instilling 20% hypertonic saline into the cyst before it's opened. Gil-Grande et al reported that albendazole effectively renders 72.3% of cysts sterile within the initial month, with a further increase to 94% by the conclusion of a three-month treatment period [13]. In our case the patient underwent an elective total splenectomy without puncturing the cysts, before which the patient had received tablet Albendazole for a month and was vaccinated against Streptococcus pneumoniae, Haemophilus influenza type b, and Neisseria meningitidis. Post-operatively the patient additionally was prescribed Tab. Albendazole.

Conclusion

Hydatid cyst of spleen being a rare entity (0.5%-4%) requires a high degree of suspicion for diagnosis particularly in non-endemic zones. The difficulty of diagnosis and scarcity of available literature make prompt, effective diagnosis and intervention challenging. CT scan is the imaging of choice with a high level of sensitivity. Although a variety of medical and surgical managements have been tried and tested and appropriate management is customized according to the particular patient, a total splenectomy followed by adjuvant anti helminthic therapy is the best curative procedure and also reducing the chances of recurrence.

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