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Case Report

A case of intra-operative anaphylactic shock in hepatic hydatidosis with inadequate chemotherapy and proximity to hepatic vessels

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ABSTRACT

Incidence of intraoperative Hydatid anaphylaxis is 0.2-3.3% Reporting of such rare cases is crucial for future reference and study. 52 years old lady presented with hepatic hydatidosis. Computed Tomography showed 2 cystic lesions; one in segment 6, 7 in proximity to right hepatic vein; multiseptated cyst with multiple daughter cysts in segment 4 adjoining the middle hepatic vein with intracystic vessel. No rupture seen. Preoperatively she was prescribed Albendazole 400mg twice-daily for 3months. She was given Hydrocortisone before surgery. 10% povidone iodine mops to prevent intraperitoneal spillage and hypertonic saline used to flush the cyst. One hour after anesthesia when the Segment IV cyst close to middle hepatic vein was punctured she had anaphylactic shock, resuscitated. Marsupialization of both the cysts with omentoplasty and intracystic drains done. She recovered well and discharged with Albendazole for 1 month. Growing cyst produces complex echinococcal antigens, increased cellular immune response, Th2 balanced with Th1; elevated immunoglobulin levels. In dead cysts Th2 responses drop rapidly. Albendazole 10mg/kg for 3 months causes good cyst wall degeneration, less viability of protoscoleces and cyst, less echinococcal antigen production. After the surgery we found that she had stopped albendazole in the preoperative period, which she did not reveal pre-operatively fearing delay in surgery. Inadequate albendazole, close proximity of cysts to vascular structures with high intracystic pressure would have predisposed to develop anaphylactic shock during cyst handling. Preoperative Albendazole can reduce anaphylaxis, morbidity and mortality due to hydatid, making hydatid surgery safer.

Keywords: Anaphylaxis, Albendazole, Echinococcus, Hydatid, Marsupialization

INTRODUCTION

Hydatid disease is found worldwide, particularly in cattle raising countries like India where agriculture is a major occupation. Echinococcosis a zoonotic disease caused by metacestode of the genus Echinococcus, where the Classical Cystic Echinococcosis is caused by E. granulosis and Alveolar polycystic Echinococcosis is caused by *E. multilocularis* and *E. vogeli*. Majority of the hydatid cysts are seen in the liver (50-93%) which acts as a filter for all cysts in the portal system. Hepatic hydatid cysts remain asymptomatic for years. Patient can manifest with jaundice, rupture or secondary infection. Liver hydatidosis without any intervention can develop biliary fistulas, rupture with peritoneal seedling, development of daughter cysts within or rarely die.[1] Anaphylactic shock due to hydatid is associated with inter-individual variation and type I IgE mediated hypersensitivity while the severity and speed depends on the amount of specific antibodies.[2]

CASE REPORT

A 52 years old lady weighing 53 kgs, from West Bengal with hypothyroidism under control, presented with complaints of upper abdominal pain. She had no history of jaundice, fever and no previous surgeries in the past. Blood investigations showed mild hyperbilirubinemia of 1.51 mg/dl, raised Alanine Transaminase of 108 U/L, alkaline phosphatase of 146 U/L, gamma glutamyl transferase of 195 U/L with a normal Lactate dehydrogenase. On evaluation she was found to have hepatic hydatid cysts. Patient's cardiac and pulmonary evaluation was within normal limits. Contrast Enhanced Computed Tomography showed enlarged liver of 16 cm with two cystic lesions. A well encapsulated cystic lesion measuring 6.1x6.5x8.3 cm noted in segment 6, 7 of liver in proximity to right hepatic vein (Figure 1a, 1b, 2c).

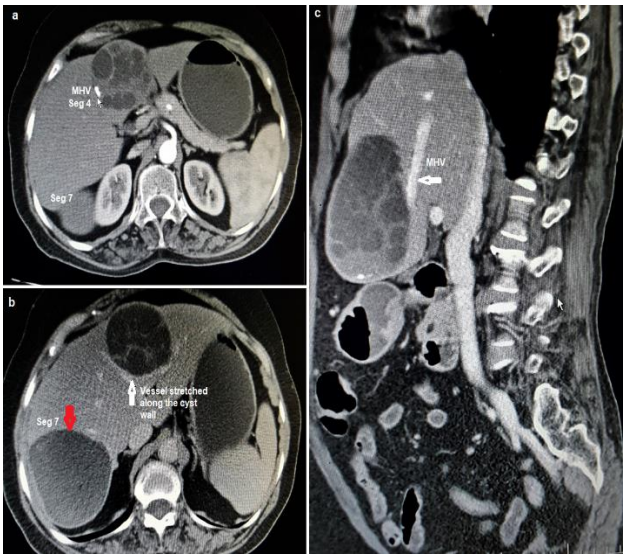

Figure 1: a. Segment 4 hydatid cyst with Middle Hepatic Vein (MHV) Branch; b. MHV branch stretched along the Cyst wall, Segment 6,7 hydtaid cyst (Red arrow); c. Lateral view Segment 4 cyst with adjoining MHV.

Another multiseptated cystic lesion measuring 5.1x5.5x8.1 cm noted in segment 4 with daughter cysts within; the medial cyst wall adjoining the middle hepatic vein. The middle hepatic vein stretched along this cyst wall with its branches entering into the cyst (Figure 1, 2a, 2b). Wall and septal calcifications noted within. No evidence of rupture seen. Intrahepatic biliary radical are normal. Preoperatively patient was started on Albendazole 400mg twice daily for 3months with drug holidays in-between. Later in the postoperative period it was found that the patient was not compliant with albendazole. Patient was started preoperatively on hydrocortisone injection 100mg Intra Venous (IV) from the day before the surgery. She was taken for marsupialization of hydatid cysts under general anesthesia. Patient was pre-oxygenated, induced with fentanyl and propofol. Neuromuscular blockade achieved with atracurium.

Figure 2: a. MHV branch adjoining the cyst; b. Vessel within the cyst (thin arrow) and MHV coursing along the cyst wall (thick arrow); c. Segment 6, 7 with adjacent Right Hepatic Vein (red arrow).

She was intubated, maintained with isoflurane, nitrous oxide, oxygen, atracurium and intermittent positive pressure ventilation. Patient was catheterized per urethra and urine output monitored. A kocher's incision was made, with adequate retraction and exposure, segment 4 cyst was handled first. 10% povidone iodine solution soaked mops were used to prevent intraperitoneal spillage and spread, hypertonic saline used to flush the cyst cavity. One hour after general anesthesia, after taking adequate precautions when the segment 4 cyst was punctured, patient had sudden hypotension with non-recordable blood pressure, pulse rate of 86 per min, oxygen saturation SpO2 95%, Pmax 29 cm of H_2O and $EtCO_2$ of 22 mm Hg suggestive of anaphylactic shock. She was immediately resuscitated with 100 mg hydrocortisone IV, Ephedrine 6mg IV, Pheniramine maleate 45mg IV, 0.5mg adrenaline IV, 100% oxygen and IV bolus fluids. After 5 minutes she continued to be in shock. She was given another cycle of Adrenaline, Ephedrine and Ketamine 10mg IV. An additional 18G wide bore IV canula was initiated. After 10 minutes her blood pressure and SpO_2 was not recordable. Noradrenaline (8mg/500ml) infusion started at the rate of 1ml per hour with fluid resuscitation. After 15 min her blood pressure was 190/100 mm Hg maintaining and noradrenaline infusion was stopped. Once she was stabilized, surgery was resumed. Segment 4 cyst contents evacuated, no biliary communication noted and the cavity flushed, packed with omentum. After releasing the attachments liver was pushed down and retracted anteriorly, segment 6, 7 hydatid cyst located posteriorly was evacuated, flushed and a 28F intracystic drain placed and brought out through the abdominal wall along with another 28F subhepatic drain. Patient was shifted to ICU for postoperative monitoring with swollen tongue, heart rate of 60/min, blood pressure of 120/80 mm Hg, SpO_2 100% with oxygen. She was monitored for 2 days, she recovered well, and she was shifted to the ward, started

on normal diet. Hydrocortisone was continued for 5 days totally. Patient was discharged on oral Albendazole 400mg twice daily for one month. Histopathological examination of the cyst showed fragments of necrosed lamellated membranes of hydatid cyst with germinative layer and scolices with rostrum and hooklets. Hydatid cyst fluid analysed by microbiologists showed yellow coloured fluid which was not blood stained; Wet mount examination showed numerous proto scolices which were circular to oval structures showing rows of hooklets within the protoscolex arranged in a circular fashion (Figure 3). A presumptive identification of Echinococcus species was made based on the microscopic findings. The fluid sample was subjected to conventional PCR for both *E. multilocularis* and *E. granulosus* targeting mitochondrial 12S rRNA gene. The species was identified as *Echinococcus granulosus*. Patient came for follow-up after one month and is doing well.

Figure 3: Microscopic image of hydatid cyst fluid under 40X. Protoscolex with rows of hooklets seen.

DISCUSSION

Surgery is the primary choice of treatment for hepatic hydatid cysts. Surgery can be conservative like marsupialization, endocystectomy or radical procedures like pericystectomy, wedge hepatic resection, segmentectomy. The concerns during the surgery are intraoperative spillage of cyst contents, spread of viable scoleces, overlooked small cysts and anaphylaxis. The incidence of anaphylaxis during hydatid surgery ranges from 0.2-3.3%.[3] During surgery, anaphylaxis is marked by hypotension, tachycardia, and arrhythmias whereas the usual signs of rashes, flushing, urticaria are masked by the surgical draping. Bronchospasm is less sensitive and infrequent after general anesthesia. In hydatid surgery, other causes of anaphylaxis should be eliminated before attributing it to hydatid fluid. In our case, other causes are not considered because the anaphylaxis happened after a long time from induction, exactly coinciding with the handling of segment 4 cyst which is adjoining the middle hepatic vein.

Albendazole is used for prophylaxis preoperatively to reduce the recurrence and viability of hydatid.[4] Eliminating viability in the hydatid cysts preoperatively would reduce the recurrence rates and anaphylaxis. Larger the cyst means more the quantity of antigenic fluid which can cause anaphylaxis during surgery. Medical therapy reduces the size of the cyst thereby reducing the amount of antigenic fluid in the cyst, sterilizes the cyst.[5] Size of the cyst is independently associated with the incidence of perioperative anaphylaxis.[2] A growing cyst produces complex echinococcal Antigens to overcome the host, mainly cellular mediated immune response. These Antigens stimulate complex immune responses including polarized Th2 responses balanced with Th1 responses; IgG1, and IgG4, IgE, and IgM levels are elevated. When the cyst is dead, dying, or surgically removed, the Th2 responses drop rapidly whereas the Th1 responses drop slowly, then becoming polarized.[6] Thus a dead or dying cyst would elicit less immunological response which would be helpful during the intraoperative period.

Albendazole is the commonly used drug with better absorption. It has variable absorption with 5-20% drug absorption, mostly from the small intestine. It gets converted to albendazole sulphoxide which is an active antihelminthic. Increasing the systemic bioavailability of albendazole has been tried with co-administering with a fatty meal, using liposomal preparations and with soya bean oil emulsions.[1] Duration of treatment less than 3 months produces suboptimal response. A randomized control study among patients undergoing immediate surgery and patients undergoing surgery after albendazole therapy demonstrated the efficacy of chemotherapy with albendazole on hydatid cyst through ultrasonographic changes, post-surgical analysis of the cyst by assessing the viability of protoscolices on microscopic analysis, supravital staining and inoculation into mice; it was found that albendazole at a dose of 10mg/kg for 3 months is most effective against uncomplicated hydatid equivalent to surgery with good cyst wall degeneration, less viability of protoscolex and cyst.[7] If viable scoleces are still identified after preoperative albendazole with surgery, then one month of postoperative albendazole is advised.[4]

On retrospective analysis following surgery, it was found that our patient was not compliant with albendazole in the preoperative period which she did not reveal earlier fearing postponement of surgery, also the hydatid cysts were in close proximity to hepatic veins as seen in CT which could have predisposed the patient to develop anaphylactic shock during the cyst handling. Combination of antigenic fluid under high intracystic pressure with veins in the hydatid cyst cavity like in our case can cause leakage of contents into the vein during blunt dissection.[8] Multiple loculated lesions are seen with

E. multilocularis and rarely with *E. granulosus*. Also *E. multilocularis* is very rare in India. Only one case has been reported from Chandigarh so far.[9] We identified the species as *Echinococcus granulosus*. Hence it is a rare case finding where *E. granulosus* has caused multiple loculated lesions.

CONCLUSION

Anaphylactic shock during surgery can be due to muscle relaxants (60-70%), latex (10-20%), antibiotics (5-20%), colloids and induction agents (3-5%). All other causes of anaphylactic shock must be excluded before attributing anaphylactic reaction/shock to hydatid cyst contents during surgery. Hydatid anaphylaxis in the perioperative period is very rare and reporting of such cases is crucial for future reference and study. An early diagnosis and meticulous management remains the mainstay in management of anaphylaxis due to Hydatid cyst surgery. Preoperative chemotherapy for atleast 3 months in elective cases would increase the non-viability of cysts and less echinococcal antigen production.

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