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Encountering the unknown: anesthetic management of a rare case of massive abdominopelvic hydatid cyst - a case report

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ABSTRACT:

- ***Background:*** Hydatid disease is a zoonosis caused by parasitic tapeworms of *Echinococcus spp*. The two most frequently impacted organs are the liver and lungs. Disseminated hydatid disease, presenting as a massive abdominopelvic cyst with accompanying cysts in the lungs, is very rare, as most patients usually have fatal anaphylaxis due to cyst rupture during disease progression.
- ***Case Report:*** We report the successful anesthetic management of a 24-year-old patient with a massive abdominopelvic hydatid cyst. She underwent exploratory laparotomy for evacuation of cysts under general anesthesia.
- ***Conclusions:*** Anesthetic management of patients with abdominopelvic hydatid cysts undergoing cystectomy is challenging. It involves taking into account the patient's clinical status, prevention and prompt management of anaphylaxis and control of hemodynamic disturbances during cyst evacuation.

- ***Keywords:*** Hydatid cyst, *Echinococcus*, Abdominal, Pelvic, Cysts, Anaphylaxis, Excision.

INTRODUCTION

Hydatid disease is caused by the parasite *Echinococcus spp.* commonly *E. granulosus*. Livestock are intermediate hosts and dogs are definitive hosts. It is transmitted to humans through the consumption of food contaminated with dog feces (containing parasitic eggs). The first-stage larva (oncosphere) hatches in the duodenum, from where it enters the portal circulation and liver (first filter). From the liver, it passes into the right heart and then the lungs (second filter). If the oncosphere passes the lung filter, it can enter the systemic circulation. The most often affected organs are the lungs (18-35%) and liver (55-70%). Peritoneal spread is rare (2-5%) and usually occurs due to the dissemination of the germinative layer of the cyst[1,2].

During development, cysts cause mechanical compression of the adjacent tissue (which forms the outermost layer or pericyst) and spread rapidly in the surrounding areas. They cause anaphylaxis on rupture. The abdomen, being a spacious cavity, allows cysts to grow without much tissue impedance and form daughter cysts until they encounter resistance and cause pressure effects. Treatment involves surgical evacuation of cysts from the peritoneal cavity *via* a midline laparotomy followed by peritoneal lavage with scolicidal agents. The surgery is challenging since massive numbers of cysts make removal *in toto* difficult. Perioperative cyst rupture can have drastic consequences such as an acute abdomen, sepsis and anaphylactic shock. Anesthetic management is likewise challenging due to ventilatory and hemodynamic problems caused by the compression effects of cysts and anaphylactic shock[3].

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We report the anesthetic management of a rare case of disseminated hydatid disease with a massive abdominopelvic cyst who underwent exploratory laparotomy for surgical evacuation of cysts.

CASE PRESENTATION

A 24-year-old female from North India (weighing 40 kg, body mass index 16.8) presented with progressive abdominal distension and breathlessness for 8 months (initially confused as pregnancy) (Figure 1). Subsequently, developed a cough with expectoration and fever for three months. A computed tomography (CT) scan of the abdomen showed a large abdominopelvic multiloculated cystic lesion extending from the xiphisternum to the pelvis. Multiple cysts were also present in the right lung along with the right hydropneumothorax. Serology was positive for *E. granulosus*. She was admitted and started on albendazole 400 mg BD. An intercostal drain was inserted for the hydropneumothorax. Meanwhile, she suffered from an episode of anaphylaxis, which was managed with pheniramine maleate 45.5 mg intravenous (IV) and hydrocortisone 100 mg IV. She was then taken for exploratory laparotomy (cystectomy). The pre-anesthetic evaluation showed cachexia with a grossly distended abdomen. There was no prior history of food or drug allergy. Her vitals were stable, but she was mildly tachypneic and auscultation revealed right basal decreased air entry and wheeze. Her blood parameters were within normal limits. A written informed consent was obtained from the patient after thoroughly explaining the risks of anaphylaxis and anaphylactic shock, the need for vasopressors, and post-operative mechanical ventilation. On the morning of surgery, she was administered ranitidine 50 mg IV and pheniramine maleate 44.5 mg IV and nebulized with salbutamol and hydrocortisone.

In the Operating Room, standard monitoring was instituted, and wide-bore IV access was secured. Once the antibiotic sensitivity was confirmed, 500 mg metronidazole and 1.5 g IV ceftriaxone were given. After inserting an arterial line and central venous catheter, modified Rapid Sequence Induction (RSI) was done with fentanyl 2 mcg/kg IV, propofol 2 mg/kg IV and succinylcholine 1-1.5 mg/kg IV. An oxygen-air mixture (1:1) with isoflurane was used for the maintenance of anesthesia, and muscle relaxation was maintained with cis-atracurium. Post-induction and intubation, we noted hypotension [fall in mean arterial pressure (MAP) up to 40 mm Hg], high peak airway pressures (45-50 cm H_2O) and reduced chest compliance. Chest auscultation showed right basal wheeze and decreased air entry. End-tidal carbon dioxide (Et CO_2) levels were normal. The endotracheal tube position was checked by laryngoscopy. IV infusion of noradrenaline was started at 10 mcg/min and titrated in accordance with MAP. Hydrocortisone 100 mg IV *statim* and salbutamol puffs *via* the endotracheal tube were given. However, airway pressures still remained high. Upon opening the abdomen and excising the mother cyst membrane, numerous daughter cysts were removed (Figures 2 and 3). When all the daughter cysts had been removed, we noticed that the peak airway pressures decreased, lung compliance improved and vasopressor requirement reduced. During the removal of the

Figure 1. Massively distended abdomen of the young female patient with massive abdominopelvic hydatid cyst.

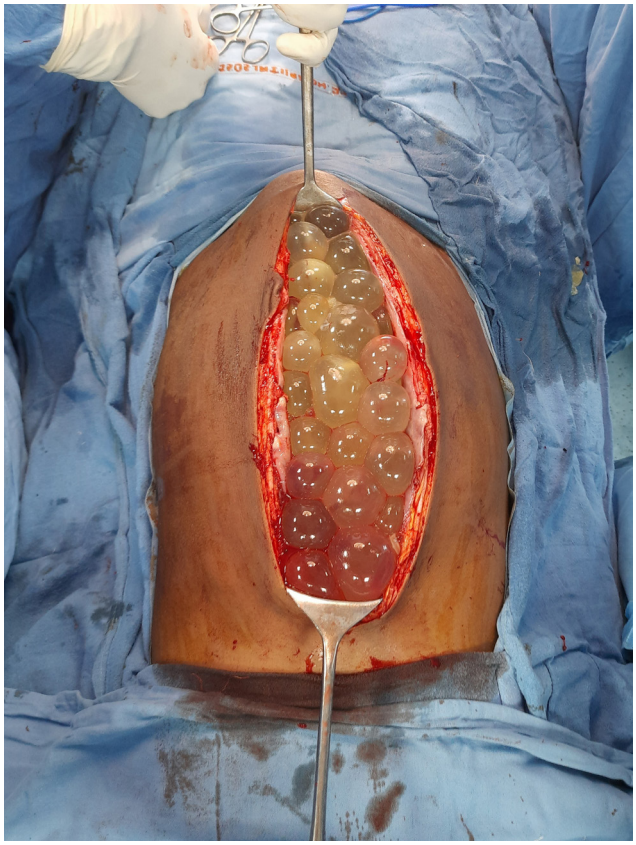

Figure 2. Numerous daughter cysts seen inside the abdominal cavity after a midline laparotomy incision on the patient with a massive abdominopelvic hydatid cyst.

mother cyst membrane (Figure 4), there was sudden hypotension (MAP 40-45 mm Hg), tachycardia (130-140 beats/min) and development of urticaria. On auscultation, there was a bilateral wheeze, and the peak airway pressures rose again (40-45 cm H_2O). Suspecting anaphylaxis, the patient was immediately given 100% oxygen. Manual ventilation was used to check for adequacy of ventilation. The plane of anesthesia was deepened. Fluid resuscitation was started. Pheniramine maleate 45.5 mg IV *statim* and two boluses of epinephrine 100 mcg IV *statim* were given. Noradrenaline support was increased and epinephrine IV infusion at 0.1 mcg/kg/min was started to maintain blood pressure. This led to a stabilization of the hemodynamics, and the remaining surgery was completed successfully. The patient was not extubated due to dual vasopressor support and the risk of recurrence of anaphylactic shock. On completion of surgery, she was sent to the Intensive Care Unit for post-operative mechanical ventilation and monitoring for recurrence of anaphylaxis. Her vasopressors were tapered, and she was extubated on postoperative day 1. She was discharged after a week with the plan to excise the pulmonary hydatid cysts at a later date.

DISCUSSION

Disseminated hydatid disease presenting as a massive abdominopelvic hydatid cyst is very rare, es-

Figure 3. Daughter hydatid cysts being removed *in toto* from the abdominal cavity of the patient.

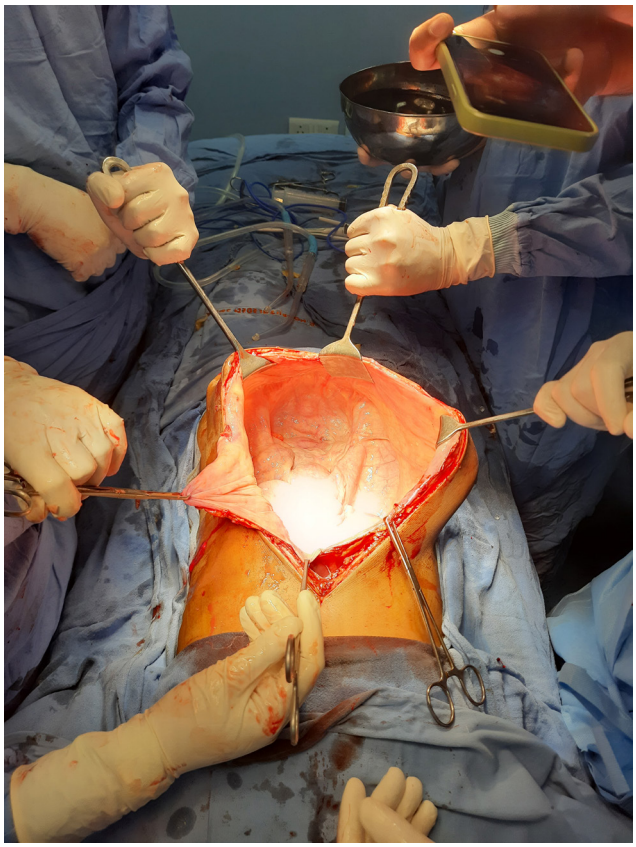

Figure 4. Empty mother hydatid cyst cavity seen in the peritoneal space after daughter cyst removal.

pecially in the North Indian population[4]. Laparoscopic approaches for cyst excision have better outcomes and reduced rates of complications but are very difficult to perform in massive peritoneal hydatid cysts. Open surgery through a midline laparotomy incision allows easier removal of the cysts but has a greater risk of complications secondary to cyst rupture[3]. These patients present with multiple issues which affect the perioperative outcomes. Malnutrition is present due to long-standing parasitic infection. In addition, the presence of pulmonary cysts increases the risk of postoperative morbidity and pulmonary complications[5,6]. Optimization of the nutritional and pulmonary condition of these patients is challenging. We nebulized the patient with salbutamol and hydrocortisone pre-operatively, which helped reduce her breathlessness due to its bronchodilator and anti-inflammatory actions, respectively. Gross abdominal distension increases the risk of aspiration. Hence, anti-aspiration prophylaxis and RSI are recommended. These patients also have poor cardiopulmonary reserve and are prone to desaturation during airway maneuvers. Therefore, adequate preoxygenation should always be done prior to RSI, and minimum attempts at laryngoscopy and intubation should be made. Apneic oxygenation techniques can also be implemented[7].

In our patient, the large cyst displaced the bowel upwards, causing increased intra-thoracic and intra-abdominal pressure and reduced the venous return. This was responsible for the initial hypotension and increased airway pressures. We saw a drastic reduction in airway pressures and an improvement in blood pressure after the evacuation of the cysts. Bronchospasm and endobronchial intubation are close differential diagnoses in this scenario. Endobronchial intubation was ruled out by auscultation and checking the position of the black mark of the endotracheal tube by laryngoscopy. Bronchospasm can also be ruled out as the wheeze was already present preoperatively, the depth of anesthesia was adequate, and the airway pressures did not resolve after the administration of beta-agonists and steroids. Also, the Et CO_2 levels would have risen in both of the above situations. When the patient developed anaphylactic shock, bronchospasm seemed to be the more likely diagnosis due to the presence of bilateral wheeze associated with urticaria and hypotension.

Anaphylaxis is common in hydatid disease with an incidence of 1-12.5%[8]. It occurs due to antigenic cyst fluid entering the bloodstream and activating IgE secretion and histamine release. During anesthesia, anaphylactic reactions have an incidence of 0.2-3.3% with a mortality rate between 3-5%[9]. Prevention of anaphylaxis is key. Firstly, a history of any known drug allergy and anaphylactic episodes should be elicited and documented. Secondly, avoiding histamine-releasing drugs like morphine and atracurium is preferable. We ruled out the use of other drugs that can cause anaphylaxis, such as antibiotics and muscle relaxants. Cis-atracurium is a safe choice of muscle relaxant as it does not cause histamine release. Thirdly, prophylactic administration of H1 and H2 blockers and glucocorticoids is recommended[10,11]. Ranitidine, an H2 blocker, serves the dual role of anti-aspiration and anti-anaphylaxis prophylaxis. Lastly, the surgeon should ensure that all the cysts are removed intact (Figure 3). Peritoneal lavage with povidone-iodine, which is a scolicidal agent, was done for decontamination and prevention of anaphylaxis[12]. However, anaphylaxis, even without rupture of the hydatid cyst, has been reported during hydatid cyst surgeries[13]. This was also observed in our case. Prompt clinical diagnosis is the critical step in management. General anesthesia can mask some symptoms of anaphylaxis, so close monitoring is warranted. Serum tryptase levels are of diagnostic importance, but we were unable to perform them due to their unavailability at our hospital. Tailoring the treatment priorities on the basis of the "Airway-Breathing-Circulation" (ABC) approach and early administration of H1 and H2 blockers, glucocorticoids and epinephrine are fundamental. Epinephrine is the gold standard drug in the treatment of anaphylaxis and anaphylactic shock. The anti-anaphylactic effect of epinephrine is due to its action on alpha and beta receptors[14]. The intravenous route is preferred over the intramuscular route intraoperatively due to its faster onset, more predictable absorption, and 100% bioavailability. The dose of epinephrine is 50-100 mcg IV bolus, and extreme precautions should be taken to avoid an overdose, which can have fatal complications[15].

CONCLUSIONS

Massive abdominopelvic hydatid cysts have multiple perioperative concerns. Such patients should receive treatment in tertiary care centers with intensive care units and surgical expertise in handling such cases. Apprehension of the possibility of anaphylaxis is paramount. Prophylactic drugs, close monitoring, and prompt treatment can help prevent adverse outcomes. Anesthesiologists should also be prepared to manage compression symptoms, which impair ventilation and cause hemodynamic disturbances.

Conflict of Interest:
The authors have no conflict of interest to declare.

Informed Consent:
Written and informed consent was obtained from the patient for publication of this case report.

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Authors' Contributions:
Sohini Ray was involved in conception and design of the study, acquisition of data, drafting the article and making critical revisions related to the relevant intellectual content of the manuscript.

Ethics Approval:
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Availability of Data and Materials:
The data used to support the findings in this study are included within the article.

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AI Disclosure:
No form of generative artificial intelligence was used for writing the manuscript.

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