CASE REPORT

"Anesthetic Management of Rare Case of Lung and Liver Hydatid Cyst with Bilateral Pheochromocytoma" – A Case Report

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ABSTRACT

Hydatidosis is a parasitic infection caused by the encysted larvae of *Echinococcus granulosus* known as hydatid cyst. It involves almost all organs. However, it affects liver (55–70%) followed by lungs (18–35%). Pheochromocytoma is a catecholamine secreting tumor that typically occurs in patients of 30–50 years age. Pheochromocytoma represents very significant challenges to the anesthetist. A 48-year-old male patient presented with dyspnea, cough, weakness, chest, and abdominal pain. Clinical examination and investigations revealed hydatid cyst of right lung and right lobe of liver. Accident findings on computed tomography scan abdomen were bilateral suprarenal mass. The patient was scheduled for excision of hydatid cyst of liver, lung, and excision of bilateral suprarenal tumor. The anesthetic technique used was general anaesthesia with epidural analgesia. During thoracotomy, one lung anesthesia was done with double lumen endotracheal tube.

Key words: Anesthetic management, double lumen tube, *Echinococcus granulosus*, hydatid cyst, one lung ventilation, pheochromocytoma

INTRODUCTION

Hydatidosis is a parasitic infection caused by the encysted larvae of *Echinococcus granulosus*, commonly called as hydatid cyst. Most commonly, it affects liver (55–70%) followed by lungs (18–35%). Thoracic complications of hepatic hydatid cyst are seen in approximately 0.6–16% cases.[1,2] The surgery and anesthetic management become very challenging if hydatid cysts are in or near the vicinity of vital organs, such as heart.[3]

Pheochromocytoma is a catecholamine secreting tumor that originates in the adrenal medulla or in chromaffin tissues along the paravertebral sympathetic chain extending from the pelvis to the base of the skull.[4] More than 95% of all pheochromocytomas are found in the abdominal cavity and 90% originate in the adrenal medulla. 10% of these tumors involve both the adrenal glands. Pheochromocytoma typically occurs in patients of 30–50 years age.[5]

We are reporting anesthetic management of a rare case of pulmonary and hepatic hydatid cyst with accidently diagnosed bilateral pheochromocytoma. Thoracotomy for lung hydatid cyst excision and laparoscopy for liver hydatid cyst excision along with bilateral adrenal mass excision was successfully managed with general anesthesia and epidural analgesia.

CASE REPORT

A 48-year-old, 52 kg, male patient was admitted with a history of fever, chronic dry cough, difficulty in breathing on exertion, pain on left scapular region, and intermittent abdominal pain for 2 months.

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During preanesthetic evaluation, his heart rate (HR) was 86/min, regular, blood pressure (BP) 130/82 mmHg, and on auscultation of chest, there was decreased air entry over left middle and lower zones. Cardiovascular examinations, including electrocardiograph (ECG) and echocardiography, were normal. On palpation of abdomen, tenderness was present over left hypochondrium and epigastric region without any associated organomegalies. Airway examination revealed normal parameters with Mallampati Score Grade II. Chest X-ray posteroanterior view revealed a cystic lesion with air and fluid level at left middle and lower zones [Figure 1].

Ultrasound sonography abdomen showed 7 × 6 cm cyst in left lobe of liver, 4 × 3 cm solid lesion in right suprarenal region, and 3 × 3 cm solid lesion in left suprarenal region. Computed tomography (CT) chest showed a 7.4 × 6.9 × 7.9 cm well-defined cystic lesion in apical and posterolateral basal segment of left lower lobe. CT upper abdomen showed 5.9 × 8.1 × 6.2 cm well-defined thin walled cystic lesion in segment II and III of left lobe of liver. CT abdomen also showed two hypervascular soft-tissue lesions involving both adrenal glands. On the right side, it measures 3.3 × 4.1 × 3.6 cm, and on the left side, it measures about 2.6 × 3.3 × 2.8 cm [Figure 2].

The patient was diagnosed having hydatid cyst of liver and left lung (middle and lower lobe) and bilateral pheochromocytoma. All routine investigations including hemogram, renal function tests, hepatic function tests, blood glucose, 24 h urine vanillylmandelic acid (VMA) level, and urine routine examination were within the normal limit. Preoperative incentive spirometry started. Left thoracotomy and laparoscopy for liver cyst and excision of bilateral adrenal mass were planned as the surgical intervention. General anesthesia and double-lumen endotracheal tube (DLT) intubation for isolation of lungs and epidural analgesia for intra- and postoperative pain relief were planned.

Written and informed consent for the American Society of Anesthesiologists status III was taken. Preoperatively, tablet ranitidine 150 mg and tablet alprazolam 0.25 mg were administered on previous night. In operation theater, monitors were attached for ECG, noninvasive BP (NIBP), invasive BP (IBP), end-tidal carbon dioxide ($ETCO_2$), and pulse oximetry (SpO_2). Intravenous (IV) access was secured with two large bore cannulae in both the upper limbs. Injection hydrocortisone 100 mg IV and injection chlorpheniramine 25 mg IV were injected before induction of anesthesia in anticipation of anaphylaxis to possible spillage of contents of the cysts during surgery. Under all aseptic conditions, epidural catheter was inserted at T11–T12 space and fixed at 5 cm into the epidural space. After negative aspiration for blood and cerebrospinal fluid, 2 mL of 2% lignocaine with 15 μg adrenaline was injected as test dose. 5 min after the test dose, 10 mL of 0.25% bupivacaine with dexona 8 mg

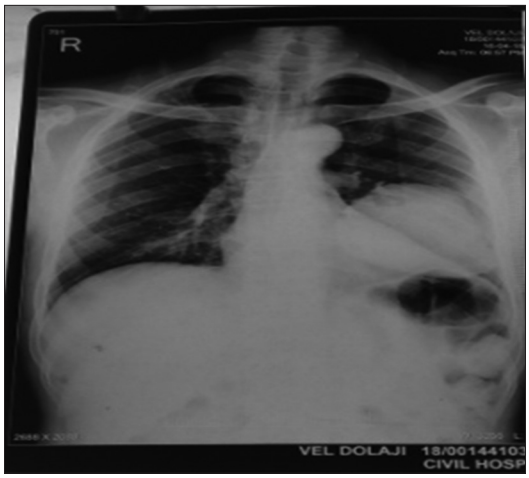

Figure 1: X-ray chest

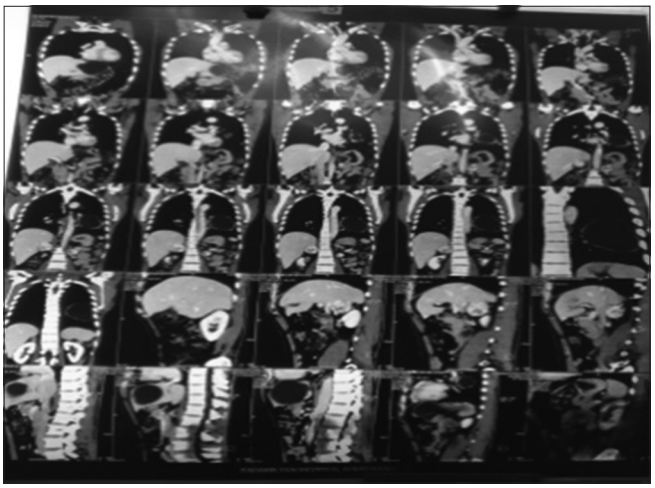

Figure 2: Contrast-enhanced computed tomography chest and abdomen

was injected into epidural space. As part of pretreatment before induction of anesthesia, injection midazolam 1 mg IV, injection glycopyrrolate 0.2 mg IV, and injection fentanyl 100 μg IV were administered. Induction of anesthesia was carried out with injection propofol 160 mg, complete muscle relaxation was achieved with injection suxamethonium 100 mg, and then, trachea was intubated with 37 Fr left-sided double lumen tube. Bilateral equal air entry was checked and confirmed while ventilation for each lung was checked individually after blocking bronchial and tracheal lumen alternatively [Figure 3]. Anesthesia was maintained with O_2, N_2O, sevoflurane, and injection vecuronium bromide.

Thoracotomy was done in right lateral decubitus position after clamping bronchial lumen and ventilating only the right lung. Surgical procedure went uneventful. In supine position, laparoscopy for drainage and excision of liver hydatid and excision of bilateral adrenal mass was carried out. During this phase of surgery, ventilation of both the lungs was carried out

Figure 3: Double-lumen endotracheal tube in place

simultaneously. Throughout surgery, continuous monitoring of HR, ECG, NIBP, IBP, SPO_2, $ETCO_2$, and urine output was done. Intermittently, arterial blood gas analysis was done to know about the metabolic profile and oxygenation status. At the end of surgery, residual neuromuscular blockade was reversed with injection neostigmine 2.5 mg and injection glycopyrrolate 0.04 mg. Trachea was extubated when the patient became fully conscious and started obeying verbal commands. Thereafter, the patient was shifted to intensive care unit (ICU) for further observation. In the ICU, pain relief was managed by epidural bolus dosage of 80 mg tramadol in 8 ml saline as and when required. Postoperative recovery was uneventful, and the patient was shifted to the surgical ward next day.

DISCUSSION

Hydatid disease caused by the larval stage of *Echinococcus* produces unilocular cysts. It most commonly affects liver and the lungs. Hepatic hydatid cyst manifests as abdominal pain or a palpable mass in the right upper quadrant. Pulmonary hydatid cysts may rupture into the bronchial tree or pleural cavity and produce cough, chest pain, or hemoptysis.[6] Surgery is mainstay method of treatment, but the risks involved during surgery include dissemination of infectious scolices from leakage of fluid and anaphylaxis.[6]

Anesthetic challenges are associated with one lung ventilation (OLV) and rupture of the cyst and dissemination. Cyst content can enter in bronchial tree, if cyst is connected with bronchus or during surgical manipulation which can result in airway obstruction or spillage of pleura and bronchioles. These can be avoided by OLV method using DLT tubes. Problems faced with OLV are V/Q mismatch, malposition of DLT, and air trapping. Confirmation by fiberoptic bronchoscopy can significantly reduce such malpositioning.[7] DLT helps to control ventilation and prevent flooding of the contralateral healthy lung.[8,9] The magnitude of allergic reactions ranges from mild hypersensitivity to fatal anaphylactic shock.[10,11]

Pheochromocytoma secretes predominantly norepinephrine. Sustained severe hypertension is often the most common presentation of pheochromocytoma.[12] Diagnosis of pheochromocytoma is confirmed by CT, magnetic resonance imaging, and raised urinary catecholamine and VMA in 24 h urine.[13,14]

Primary goal is the delivery of an anesthetic which provides stable hemodynamics in the face of catecholamine surges (especially at laryngoscopy, peritoneal insufflation, surgical stimulation, and tumor handling) followed by the opposite scenario following tumor ligation.[13,15]

Roizen criteria to objectively gauge the efficacy of adequate preoperative alpha-blockade include no in-hospital BP >160/90 mmHg for 24 h prior to surgery, no orthostatic hypotension with BP <80/45 mmHg, no ST or T-wave changes for 1 week before surgery, and no more than 5 premature ventricular contractions per minute.[13,15] The alpha-adrenergic blockade is typically administered starting 10–14 days preoperatively which normalizes BP and also aids in expanding the highly contracted intravascular volume. Successful alpha-blockade is reflected by normalizing BP with mild orthostasis.[13,15]

Preoperatively, the patient had normal BP and antihypertensive not started in our patient. As our patient had hydatid cyst in the lung and liver and also bilateral adrenal mass, we preferred general anaesthesia and lower thoracic epidural for analgesia for our patient as it was a thoracotomy and laparoscopic approach.

Premedication should avoid drug which releases histamine.[16] Here, in our case, we used benzodiazepine as premedication to reduce anxiety induced activation of sympathetic nervous system. Invasive BP monitoring during surgery has many advantages. It allows continuous beat-to-beat monitoring so close hemodynamic monitoring and if needed rapid pharmacologic intervention can be done during induction and intraoperatively.

Central venous cannulation allows for fluid management as per central venous pressure and as per need delivery of vasopressors or vasoactive agents. We monitored noninvasive and IBP and also CVP.

In our case, induction of anesthesia was done with propofol and fentanyl a potent short acting opioid as analgesic. Both agents can modify the hemodynamic effect of laryngoscopy and intubation. Lidocaine (preservative free) 1.5 mg/kg I.V. was administered 1 min before laryngoscopy to attenuate pressure response of laryngoscopy and intubation. Vecuronium

was used as muscle relaxant for maintenance of anesthesia due to its cardiovascular stability and inability to release histamine. Hydrocortisone replacement was also given. Glucocorticoid and mineralocorticoid cover is mandatory for patients undergoing bilateral adrenalectomy.[13-15]

Anesthetic implications in our case included high chances of hemodynamic instability due to bilateral pheochromocytoma and close proximity of the hydatid cyst to the heart and surgery over the anatomical site possibly causing rupture of cysts and leading to anaphylaxis. Throughout the surgery, hemodynamic stability was maintained. Laparoscopic adrenal mass resection provides better hemodynamic stability.

In conclusion, the management of a case of hydatid disease of the lung and liver along with bilateral pheochromocytoma for thoracotomy and laparoscopy surgery includes thorough understanding of the respiratory physiology of the OLV and the proper positioning of the double-lumen endobronchial tube and continuous monitoring of the saturation, ventilation, hemodynamics, and blood gases to prevent any associated complications and adequate fluid replacement.

REFERENCES

1. Gómez R, Moreno E, Loinaz C, De la Calle A, Castellon C, Manzanera M, *et al.* Diaphragmatic or transdiaphragmatic thoracic involvement in hepatic hydatid disease: Surgical trends and classification. World J Surg 1995;19:714-9.
2. Kilani T, El Hammami S, Horchani H, Ben Miled-Mrad K, Hantous S, Mestiri I, *et al.* Hydatid disease of the liver with thoracic involvement. World J Surg 2001;25:40-5.
3. Bajwa SJ, Panda A, Bajwa SK, Kaur J, Singh A. Anesthetic challenges in the simultaneous management of pulmonary and hepatic hydatid cyst. Anesth Essays Res 2011;5:105-8.
4. Hull CJ. Phaeochromocytoma. Diagnosis, preoperative preparation and anaesthetic management. Br J Anaesth 1986;58:1453-68.
5. Thomas JL, Bernardino ME. Pheochromocytoma in multiple endocrine adenomatosis. Efficacy of computer tomography. JAMA 1981;245:1467-9.
6. White C Jr., Weller PF. In Harrison's Principles of Internal Medicine *Cestodes*: Echinococcosis. 15th ed. United States: Mcgraw Hill medical Publishing Division; 2001. p. 250-1.
7. Esme H, Fidan H, Cekirdekci A. The problems and advantages of one lung ventilation during surgical intervention in pulmonary hydatid Cyst Disease. Indian J Thorac Cardiovasc Surg. 2006;22:137-40.
8. Bickel A, Loberant N, Singer-Jordan J, Goldfeld M, Daud G, Eitan A, *et al.* The laparoscopic approach to abdominal hydatid cysts: A prospective nonselective study using the isolated hypobaric technique. Arch Surg 2001;136:789-95.
9. Smego RA Jr. Sebanego P. Treatment options for hepatic cystic echinococcosis. Int J Infect Dis 2005;9:69-76.
10. Wellhoener P, Weitz G, Bechstein W, Djonlagic H, Dodt C. Severe anaphylactic shock in a patient with a cystic liver lesion. Intensive Care Med 2000;26:1578.
11. Anthi A, Katsenos C, Georgopoulou S, Mandragos K. Massive rupture of a hepatic hydatid cyst associated with mechanical ventilation. Anesth Analg 2004;98:796-7.
12. Magner WM, Jun G. Phaeochromocytoma overview. In: Swabs JD, editor. Text book of Hypertension. Oxford: Blackwell Scientific; 1994. p. 941-58.
13. Prys-Roberts C. Phaeochromocytoma recent progress in its management. Br J Anaesth 2000;85:44-57.
14. Pullerits J, Ein S, Balfe JW. Anaesthesia for phaeochromocytoma. Can J Anaesth 1988;35:526-34.
15. Pace N, Buttigieg M. Phaeochromocytoma. BJA CEPD Rev 2003;3:20-3.
16. Wallece JM, Gill DP. Prazosin in diagnosis and management of phaeochromocytoma. JAMA 1978;240:2752-3.

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