Case Report

Successful Pregnancy in Chronic Obstructive Pulmonary Hypertension Secondary to Pulmonary and Cardiac Hydatid Cysts: A Case Report

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Highlights

- Pregnancy is typically contraindicated in women with pulmonary hypertension (PH) due to severe mortality risks.
- A 31-year-old woman with PH, secondary to previous hydatid cysts, conceived against medical advice while on medical therapy. She successfully delivered via emergency C-section at 37 weeks.
- Her case highlights those high-risk pregnancies complicated by PH can be successfully managed through prompt, multidisciplinary care at a specialized tertiary center.

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ABSTRACT

Background: Pregnancy in women with pulmonary hypertension (PH) is contraindicated because of high mortality rates associated with physiological changes, particularly in the peripartum and postpartum periods. Termination is recommended early if pregnancy occurs. Consequently, clinical experience with continued pregnancy in patients with PH is scarce.

Case Presentation: A 31-year-old woman was diagnosed with multiple bilateral pulmonary and right ventricular hydatid cysts. After surgical and pharmacologic treatments, she developed PH due to pulmonary artery obstruction 8 years later. Because of her condition, she was ineligible for surgery and received medical therapy, including tadalafil, bosentan, and eplerenone. After 3 years, she conceived despite advice to avoid pregnancy. At 37 weeks' gestation, she underwent successful emergency cesarean delivery because of maternal dyspnea and tachycardia.

Conclusion: Prompt treatment and regular follow-up in a tertiary care center using a multidisciplinary approach are crucial for managing pregnant patients with PH.

Keywords: Case Report; Heart; Hydatid Cyst; Lung; Pregnancy; Pulmonary Hypertension

Background

The 2 most common types of hydatid disease are cystic and alveolar, caused by Echinococcus granulosus and Echinococcus multilocularis, respectively.[1]

These are endemic in several parts of the world, including the Middle East, Eastern Europe, South America, and the Mediterranean.[2] The liver (70%) and lungs (20%) are the most typical sites for these cysts; nonetheless, they can spread to any organ. Brain, heart, bone, and spleen involvement is uncommon.[1,3] Among previous cases, heart involvement is reported in 0.5% to 2% of cases.[4-6]

Although all parts of the heart can be involved, cysts are found in the free wall of the left ventricle in more than half of cases.[4,7] Other involved areas include the interventricular septum (10%–20%), right ventricular myocardium (15%), left atrium (8%), pericardium (8%), pulmonary artery (7%), and right atrium (3–4%).[7] To diagnose hydatid cysts, echocardiography is noninvasive, sensitive, and simple.[8] Subsequently, computed tomography and magnetic resonance imaging, as well as serologic testing, are conducted to confirm the diagnosis.[9,10] Surgery is the primary treatment for ventricular hydatid cysts.[8] Removal techniques include left or right thoracotomy with cardiopulmonary bypass, moderate hypothermia, and aortic cross-clamping.[11]

Hydatid cyst–related pulmonary emboli are uncommon and often have a dismal outcome. Surgery remains the gold standard for treating pulmonary emboli produced by hydatid cysts. Aneurysms, hydatid dispersion, new embolus creation, anaphylactic shock, and ruptured hydatid cysts or arteries are serious complications that may arise during surgery.[12] Additionally, pulmonary hypertension (PH) can be caused by pulmonary artery embolization in cardiac cysts.[13] PH is a heterogeneous condition defined by a mean pulmonary artery pressure of 20 mm Hg or more.[14]

This case report describes a woman with PH due to hydatid cyst–related pulmonary emboli undergoing a successful delivery via emergency cesarean section.

Case Presentation

A 31-year-old woman was admitted in 2008 with chest pain and dyspnea (functional class 3). Chest CT and CT angiography revealed multiple bilateral pulmonary hydatid cysts. Albendazole treatment continued for 2 years. At 3-year follow-up, the patient presented with cough, nonmassive hemoptysis, dyspnea (functional class 3), and weight loss. Transesophageal echocardiography revealed a right ventricular hydatid cyst with an ejection fraction of 45%.

She underwent surgery to evacuate the cardiac cyst (Figure 1, Video 1, and Supplementary File 1). Postoperative albendazole was administered for 6 months. One year later, she presented with worsening dyspnea (functional class 4). Transesophageal echocardiography and right heart catheterization confirmed chronic obstructive PH with systolic pulmonary artery pressure of 70 mm Hg. The patient was inoperable because of distal-type chronic thromboembolic PH. She was followed up with echocardiography and medical treatment, including tadalafil (10 mg twice daily), bosentan (10 mg daily), and eplerenone (25 mg daily). That year, she became pregnant and refused termination. A multidisciplinary team monitored the pregnancy; treatment continued with digoxin (0.125 mg daily) and enoxaparin (60 mg subcutaneously twice daily). Bosentan was discontinued. Monthly echocardiography was performed.

At 37 weeks' gestation, emergency cesarean delivery was performed under general anesthesia because of maternal tachycardia and worsening dyspnea. A live male newborn (weight, 2100 g; height, 50 cm; Apgar score, 9) was delivered without congenital malformations. The patient was transferred to the ICU and received a drip of prostaglandin, milrinone, dobutamine, and intravenous iloprost for 72 hours. Blood pressure during and after delivery remained within the normal range. Before and during pregnancy, echocardiograms showed no significant changes, with an ejection fraction of approximately 55%.

Both the mother and the neonate were discharged with stable vital signs. The patient's treatment continued with macitentan (10 mg twice daily), digoxin (0.125 mg daily), and eplerenone (25 mg daily). Breastfeeding was not

recommended. At 12-month follow-up, the child showed normal growth and development.

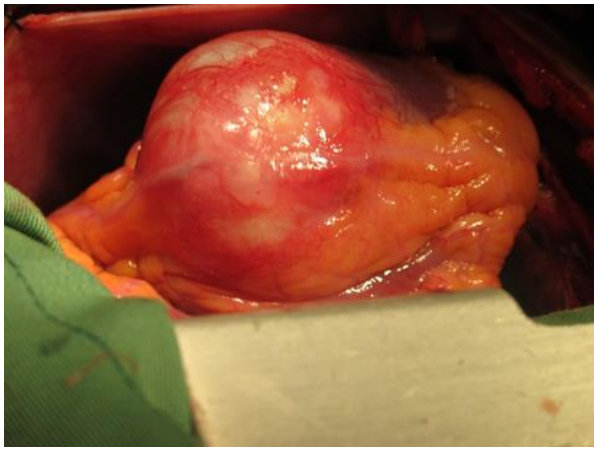

Figure 1. Cardiac hydatid cyst during surgery.

Discussion

A case of successful pregnancy and delivery is presented in a patient with chronic obstructive PH following treatment for pulmonary and cardiac hydatid cysts.

Cardiac hydatid cysts are rare, affecting 2% of patients.[9] Because of left coronary artery dominance, cardiac hydatid cysts occur more frequently in the left ventricle. Nevertheless, right ventricular hydatid cysts are more likely to rupture, which can result in anaphylaxis, pulmonary embolism, and sudden death.

Typically, hydatid cyst symptoms do not appear until the cysts enlarge.[16,17] Even in asymptomatic patients, cardiac hydatid cysts should be surgically removed because of the significant risk of complications.[18] The use of anthelmintic chemotherapy before cardiac hydatid cyst surgery is controversial.[19] De Moraes et al[20] reported that preoperative therapy is generally not advised because it increases membrane friability, which increases the likelihood of rupture during surgery. In contrast, Wadhawa et al[21] reported that 8 patients with cardiac hydatid cysts received albendazole treatment for 2 weeks preoperatively, and this treatment was associated with positive outcomes. However, albendazole treatment for 3 to 6 months following removal has been recommended in most studies.[20,22]

Hydatid cysts can also cause pulmonary embolism when they rupture or detach from the right atrium and right ventricle, allowing cyst contents to enter the pulmonary artery and its branches.[12] The clinical presentation of pulmonary embolism can lead to 3 potential outcomes: acute embolism-related abrupt death, subacute PH, and chronic PH cases.[23] Patients diagnosed with acute or chronic echinococcal pulmonary emboli have a poor prognosis, with mortality occurring within months.[15] In addition, PH can be caused by embolization of the pulmonary artery in cardiac cysts.[13]

Diagnosing PH in pregnant individuals is crucial and can have fatal consequences.[24] According to current guidelines, a woman with PH should not become pregnant.[14] Care for women with pulmonary arterial hypertension (PAH) should begin with contraceptive education because the maternal mortality rate during pregnancy and delivery is high, ranging from 30% to 35%.[25] In case of pregnancy, termination should be considered; if the patient refuses, optimizing therapy, close monitoring, avoiding teratogenic drugs, and a multidisciplinary team approach are necessary. The primary goal of patient-specific treatment plans is the prevention of systemic hypotension, hypoxemia, and acidosis while maintaining euvolemia.[14] The lateral decubitus position is recommended to reduce the likelihood of inferior vena cava obstruction. The 2 main types of pharmacologic treatments are anticoagulation and specific PH therapies. For hereditary, idiopathic PAH, and chronic thromboembolic PH, anticoagulation with low-molecular-weight heparin is recommended. Low-dose aspirin may also reduce the risk of preeclampsia.[14] Sildenafil and tadalafil are phosphodiesterase 5 inhibitors that have been successfully used by pregnant women.[26]

Guidelines for the preferred method of delivery in patients with PAH are not established, and the optimal approach remains disputed. The Pulmonary Vascular Research Institute has stated that elective cesarean section is the preferred approach, although vaginal delivery has been proposed as an alternative.[27] Meng et al[28] showed a higher mortality rate after cesarean section compared with vaginal delivery. Still, other reports cite cesarean section as the preferred delivery method.[28,29] Therefore, the mode of delivery in patients with PH remains controversial, and the choice is currently a case-by-case decision made by multidisciplinary teams.

Conclusions

Although pregnancy is contraindicated in women with PH, some patients conceive and refuse termination. Safe and successful pregnancy outcomes are achieved through cooperation between a multidisciplinary medical team and the patient. In such cases, regular follow-up in specialized centers using a multidisciplinary approach is recommended.

Declarations:
Ethical Approval

This study followed the principles of the Declaration of Helsinki and was approved by the medical ethics committee.

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Conflict of Interest

The authors declare no conflicts of interest

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