

Pneumologia

First report of a case of negative-pressure pulmonary edema during pregnancy

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Abstract

English:

Negative-pressure pulmonary edema (NPPE) or postobstructive pulmonary edema (POPE) is a rare but life-threatening complication in the perioperative and critical care settings. In this study, we present the first case of NPPE in a pregnant woman.

Case presentation: A 39-year-old 31-week pregnant woman underwent transurethral lithotripsy. She had respiratory distress about 10 minutes after extubation. Chest imaging confirmed pulmonary edema. The patient was managed in the intensive care unit (ICU) with intravenous furosemide supplemental oxygen. Evaluations showed no heart failure, but laboratory tests confirmed the presence of primary hyperparathyroidism. She was discharged on 5th day without complication.

Conclusion: In this study, we reported the first case of NPPE in a pregnant woman. NPPE should be considered in differential diagnosis of respiratory distress during pregnancy.

Keywords

Negative pressure pulmonary edema • postextubation • pregnancy • hyperparathyroidism

Prima raportare a unui caz de edem pulmonar cu presiune negativă în timpul sarcinii

Rezumat

Romanian:

Edemul pulmonar cu presiune negativă (EPPN) sau edemul pulmonar postobstructiv (EPPO) este o complicație rară, dar care pune viața în pericol în mediile perioperatorii și de terapie intensivă. În acest studiu, prezentăm primul caz de NPPE la o femeie însărcinată.

Prezentarea cazului: O femeie însărcinată în vârstă de 39 de ani, în săptămâna 31 de sarcină a suferit litotripsie transuretrală. Pacienta a dezvoltat detresă respiratorie la aproximativ 10 minute după extubare. Imagistica toracică a confirmat edemul pulmonar. Pacienta a fost tratată în unitatea de terapie intensivă (UTI) cu furosemid administrat intravenos și cu oxigen suplimentar. Evaluările nu au arătat insuficiență cardiacă, dar testele de laborator au confirmat prezența hiperparatiroidismului primar. A fost externată în a 5-a zi fără complicații.

Concluzie: În acest studiu, am raportat primul caz de NPPE la o femeie însărcinată. NPPE trebuie luată în considerare în diagnosticul diferențial al detresei respiratorii în timpul sarcinii.

Cuvinte-cheie

edem pulmonar cu presiune negativă • postextubare • sarcină • hiperparatiroidism

Introduction

Negative-pressure pulmonary edema (NPPE) or postobstructive pulmonary edema (POPE) is a life-threatening condition in the perioperative and critical care

settings. (1). Tsi *et al.* (2) reported NPPE as a relatively rare problem with an incidence rate of 0.019% among 85,561 patients receiving general anesthesia with endotracheal

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intubation. The most common cause of NPPE in adults is post-extubation laryngospasm (3), accounting for as much as 50% of adult cases. It has also been reported with choking (4), hanging (5), recurrent polychondritis, aspiration of foreign material, obstructive sleep apnea (6), epiglottitis and croup (especially in children), and biting of the endotracheal tube (7).

As far as the researchers of this study investigated, no study has reported NPPE in a pregnant woman so far. In this case report, we present NPPE in a 39-year-old pregnant woman undergoing transurethral lithotripsy.

Case presentation

A 39-year-old 31-week pregnant woman underwent general anesthesia with endotracheal intubation for transurethral lithotripsy on January 09, 2022. She was extubated and transferred to the recovery room. In the recovery room, she found respiratory distress about 10 minutes after extubation. Physical examination revealed tachycardia, tachypnea, and bilateral crackles. In pulse oximetry, SPO₂ dropped to 70%. We prepared the patient for reintubation, but the SPO₂ rose to 92% and the patient got better with supplemental oxygen and intravenous furosemide. Then, she was transferred to the intensive care unit (ICU) for further management. The portable anteroposterior chest radiography of the patient was consistent with pulmonary edema (Figure 1A). There was no pneumothorax or pleural effusion. ECG demonstrated sinus tachycardia without any sign of ischemia, infarction, or arrhythmia.

Echocardiography was performed three times, which showed normal left ventricular size with ejection fraction 55-60%, no pericardial effusion, no valvular disease, and right ventricular systolic pressure (RVSP) of 35 mmHg.

The results of laboratory tests in the ICU were as follows: white blood cells count (WBC) 9400 cells/mcL, Hb 11.1g/dL, platelets 150000/mcL, total bilirubin 0.8 mg/dL, urea 27 mg/dL, creatinine 0.89 mg/dL, Na 138 mEq/mL, and potassium 3.2 mEq/mL. Calcium level was checked three times, which showed hypercalcemia with total calcium 2.5 mmol/dL (ionized 1.41 mmol/dL), 12.3 mg/dL (ionized 1.73 mmol/dL), and 12.1 mg/dL (ionized 1.5 mmol/dL). Magnesium level was 1.3 mg/dL (normal: 1.8-2.6), phosphorus 1.7 mg/dL (normal: 2.5-5), 25 dihydroxy Vitamin D 19 ng/mL, iPTH 314.9 Pg/mL (normal 12-65), and T4 10.7 mL, and TSH 2.34 mIU/mL (normal 0.3-5.5). Finally, primary hyperparathyroidism was diagnosed.

Follow-up CXR on January 12, 2022 showed decreased pulmonary edema (Figure 1B). We did not perform CT scan due to pregnancy. The patient was discharged from ICU on

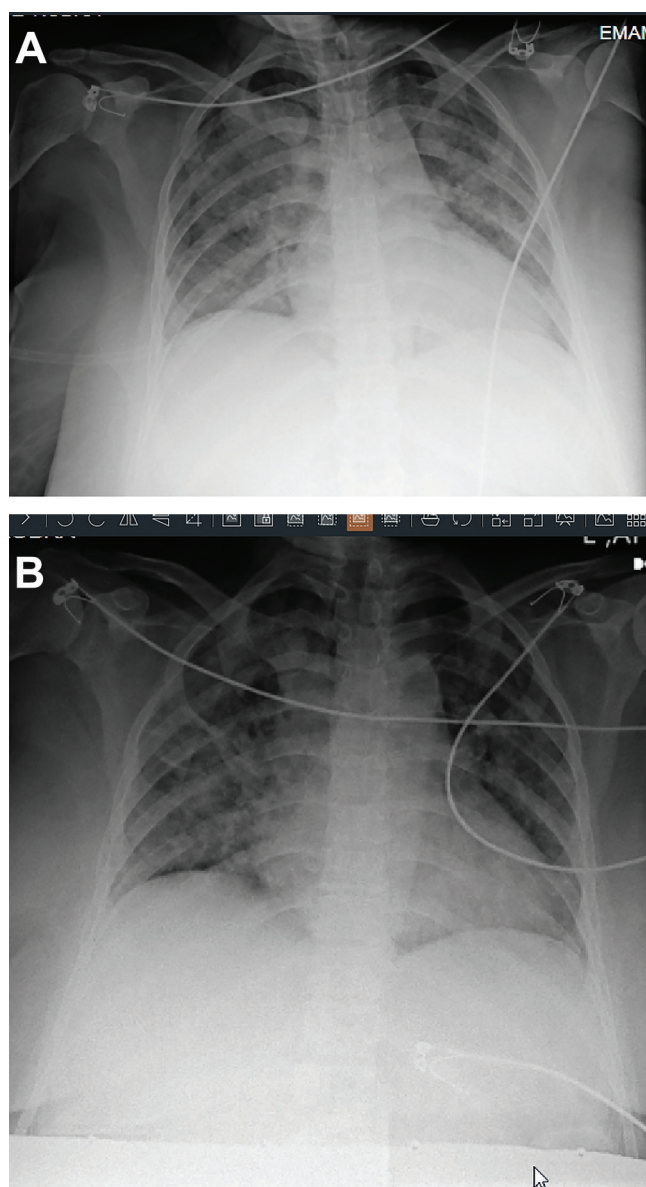


Figure 1. A: Chest X-ray Shows bilateral pulmonary edema, **B:** decreased edema with intravenous diuretics. White arrow shows abdominal lead shield to decrease fetal radiation exposure.

the 5th postoperative day without any complication. We also recommended her to continue pregnancy with follow-up for hyperparathyroidism.

Discussion

To the best of our knowledge, we report the first case of NPPE during pregnancy. There are two types of NPPE. Type

I occurs after relief of sudden and severe episode of upper airway obstruction and type II develops after relief of a chronic partial airway obstruction (8).

The reported risk factors of post-extubation NPPE include male sex, obesity, use of irritant volatile agents, difficult intubation, operation on the head and neck region, and recent upper airway infection (2). Laryngospasm can be triggered by stimuli in the throat such as foreign body, secretion blood, gastric juice, and cold temperate.

Pregnancy has not been stated as a risk factor for NPPE. NPPE is a noncardiogenic pulmonary edema with two suggested mechanisms for its development. First and most supported mechanism is hydrostatic pulmonary edema which is supported by low edema fluid protein concentration in most patients (9). Forceful inspiration to overcome an obstructed upper airway result in marked increase in negative intrathoracic pressure, and it may even reach to minus one hundred (-100) cmH₂O. Most NPPE cases are reported in otherwise healthy young individuals who are capable of producing such markedly negative intrathoracic pressures. More negative pressure leads to increased right ventricular output due high venous retu. More negative interstitial pressure in the presence of high right ventricular output disturbs Starling equation and promotes fluid movement to interstitial space and alveoli. Our patient was a 31-week pregnant woman without any history of cardiovascular or respiratory disease and she was classified as status one according to the ASA classification of physical status. Young healthy people (ASA I–II) are at a greater risk for NPPE.

The symptoms of our patient developed in the recovery room. NPPE usually presents rapidly within few minutes after reliving upper airway obstruction. However, delayed onsets have been reported, Liu *et al.* (10) described a 25-year-old woman with NPPE 30 minutes after endoscopic thyroidectomy.

The differential diagnosis for the patient included cardiogenic pulmonary edema, massive fluid infusion, anaphylaxis, pulmonary thromboembolism, and acute respiratory distress syndrome due to obstetric events.

One of the main differential diagnoses was cardiogenic and overloads pulmonary edema. One study reported concomitant NPPE and Takotsubo cardiomyopathy after acute upper airway obstruction [1]. All cardiovascular system examinations and echocardiography results (for three times) were normal in this patient.

Meanwhile, there were no signs of allergy such as flushing urticaria and eosinophilia, and the patient had no massive liquid infusion during surgery. Obstetric events that lead to pulmonary edema should be considered in differential diagnosis in pregnancy. Our patient had no obstetric event at admission, and she was discharged without obstetric events.

Our patient had hypercalcemia with increased PTH. She also had primary hyperparathyroidism, which explains her urolithiasis. We investigated any possible relationship between hypercalcemia and hyperparathyroidism with pulmonary edema. Extensive search in PubMed and Google scholar showed no report of pulmonary edema in patients with mild hypercalcemia. Although there are rare reports of pulmonary edema in hypercalcemia, all the reported cases had severe hypercalcemia such as calcium (22.5 mg/dl) parathyroid crisis (11) associated with malignancy or pancreatitis. There is only one report of pulmonary edema in a patient with primary hyperparathyroidism without malignancy or pancreatitis (12). However, in the mentioned reference, the patient was old (a 72-year-old female) and she suffered from diabetes mellitus, atrial fibrillation (AF) on anticoagulant, and had moderate to severe hypercalcemia (Ca:15.4 mg/dL). So, it seems that there is no relationship between pulmonary edema and mild hypercalcemia. In an animal study, Chen *et al.* (12) showed that hypercalcemia produced a sepsis-like syndrome by increasing free radicals proinflammatory cytokines, plasma nitrate/nitrite, and iNOS activity in rats. Increased nitrate/nitrite ratio and iNOS lead to pulmonary damage.

We used furosemide for treatment. Diuretics is unlikely to be harmful in NPPE and may hasten the resolution of pulmonary edema because pulmonary vascular pressure is elevated in NPPE (13) similar to cardiogenic pulmonary edema (13). NPPE is typically self-limited and most of the reported cases were resolved with supportive measures. However, few studies reported cases requiring reintubation and even ECMO for NPPE (14).

Several methods to reduce laryngeal irritation have been suggested to prevent NPPE. These include intravenous and laryngotracheal lidocaine, appropriate and careful initial intubation, limiting the use of aggressive suctioning during the procedure, as well as extubation under lighter sedation (15). However, none of these techniques have been proven as effective.

The report described a case of NPPE in a pregnant woman. The diagnosis of NPPE requires a high index of suspicion, as the presentation mimics other causes of pulmonary edema, such as cardiogenic pulmonary edema, iatrogenic volume overload, and Mendelson's syndrome during general anesthesia.

Conclusion

We presented the first case of NPPE during pregnancy with good course. NPPE should be suspected and managed early in postoperative dyspneic pregnant patient to prevent unfavorable outcomes. In addition, the patient has primary

hyperparathyroidism, we didn't find its any relation with pulmonary edema.

Informed consent statement

Informed consent was obtained from all subjects involved in this study

Conflict of interest

We have no Conflict of interest.

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