Spontaneous Recurrent Pneumothorax during Pregnancy Secondary to Ectopic Deciduosis

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Abstract

Background  Ectopic deciduosis is a benign presence of endometrial tissue outside of the uterus during pregnancy that rarely presents with pleuropulmonary manifestations and recurrent pneumothorax.

Case Description  We report a 35-year-old woman at 15 weeks’ gestation with a history of recurrent intrapartum right pneumothorax found to have pleural, pulmonary, and diaphragmatic lesions and a middle lobe air leak. Wedge resection of the middle lobe and mechanical pleurodesis was performed. Histopathological analysis was progesterone receptor and PAX8 positive consistent with ectopic deciduosis.

Keywords ► pneumothorax ► diaphragm ► pathology ► pleural disease ► pregnancy ► thoracoscopy/VATS

Introduction

Ectopic deciduosis describes the presence of endometrial tissue outside of the uterus during pregnancy. The pathophysiology of these lesions is yet to be fully elucidated; however, the clinical course of ectopic deciduosis is generally benign and resolves in the postpartum period.1 Ectopic decidualization most commonly affects the omentum and ovary.1 Rarely, ectopic endometrial tissue can involve the pleural space and lung, and cause pneumothorax. Intrapartum spontaneous pneumothorax is a rare, but life-threatening clinical condition that requires prompt diagnosis and management, and requires additional consideration to the risks posed to the fetus for any intervention. In this report, we describe a rare case of recurrent intrapartum pneumothorax caused by ectopic deciduosis.

Case Description

A 15-week pregnant 35-year-old multiparous woman presented to the emergency department (ED) with shortness of breath and right-sided chest pain around an existing thoracostomy tube. She reported that the chest tube had been placed abroad a few days earlier in Kenya, just prior to a scheduled international flight. She also reported a history of two prior pneumothoraces during the first trimester. Imaging on presentation demonstrated a malpositioned tube, but no pneumothorax. The chest tube was removed, and she was subsequently discharged after repeat imaging showed a fully expanded lung without pneumothorax.

The patient returned 2 weeks later with similar chest pain and shortness of breath. Imaging demonstrated a recurrent right-sided pneumothorax. The patient was initially
managed conservatively with pigtail thoracostomy and continuous suction. However, she demonstrated a persistent air leak and reaccumulation of pleural air on each attempt at water seal. After extensive discussion with the patient, we proceeded with video-assisted thoracoscopy (VATS) with plan for blebectomy and pleurodesis.

Intraoperatively, we identified innumerable friable, pinkish, frond-like lesions involving the parietal pleura, diaphragm, and all surfaces of each lobe of the right lung (Fig. 1). There was no evidence of blebs or bullae. Due to the diffuse nature of these lesions it was felt that complete removal would not be possible, and we focused on identifying and treating the active air leak. A leak test was performed, and bubbling was noted from a discrete area of the middle lobe with involvement of the described lesions. Wedge resection and mechanical pleurodesis were performed with resolution of the bubbling on leak test. The patient recovered uneventfully and was discharged on postoperative day 4 after chest tube removal the day prior. Pathologic evaluation demonstrated that the tissue was Human Melanoma Black 45 (HMB45) negative, cytokeratin negative, progesterone receptor positive, and weakly positive for PAX8.

The patient was followed closely by both the general surgery and obstetrical teams. She did present to the ED on two occasions with recurrent chest pain and was found to have a small recurrent basilar pneumothorax. In both instances she was managed with 100% oxygen and observation and have a small recurrent basilar pneumothorax. In both instances she was managed with 100% oxygen and observation and had no evidence of blebs or bullae. Due to the diffuse nature of these lesions it was felt that complete removal would not be possible, and we focused on identifying and treating the active air leak. A leak test was performed, and bubbling was noted from a discrete area of the middle lobe with involvement of the described lesions. Wedge resection and mechanical pleurodesis were performed with resolution of the bubbling on leak test. The patient recovered uneventfully and was discharged on postoperative day 4 after chest tube removal the day prior. Pathologic evaluation demonstrated that the tissue was Human Melanoma Black 45 (HMB45) negative, cytokeratin negative, progesterone receptor positive, and weakly positive for PAX8.

Management of pleuropulmonary changes associated with ectopic deciduosis are challenging due to a high rate of pneumothorax recurrence. As documented in the prior cases, we also recommend VATS exploration with blebectomy and mechanical pleurodesis as indicated, in addition to routine chest radiography until delivery to monitor for worsening or recurrence of pneumothorax. Once the diagnosis of ectopic deciduosis is made, a multidisciplinary plan should be made to ensure a safe delivery. In the case of our patient, a multidisciplinary complex delivery conference was held between the department of surgery, obstetrics, and anesthesiology. Additionally, a chest tube kit and other necessary equipment were available at bedside during delivery to facilitate emergent management of pneumothorax recurrence secondary to increased intra-abdominal pressure during vaginal delivery.

Spontaneous pneumothorax during pregnancy is a rare occurrence without clear differential diagnosis or management guidelines. Although uncommon, spontaneous pneumothorax due to ectopic deciduosis is a life-threatening condition that ultimately requires surgical management. Suspicion for this etiology should be heightened particularly in a gravid patient with a history of recurrent pneumothoraces.

**Conflict of Interest**
None declared.

**References**

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