



# Spontaneous Recurrent Pneumothorax during Pregnancy Secondary to Ectopic Deciduosis

Ali A. Hakimi, BS<sup>1</sup> Morgan M. Sellers, MD<sup>2</sup> Alexander P. Morton, MD<sup>3,4</sup>

<sup>1</sup>University of Colorado Anschutz School of Medicine, Aurora, CO

<sup>2</sup>Department of Surgery, Ernest E. Moore Shock Trauma Center at Denver Health, CO, USA

<sup>3</sup>Division of GI, Trauma, and Endocrine Surgery, Department of Surgery, University of Colorado School of Medicine, Aurora, USA

<sup>4</sup>Department of Surgery, Denver Health Medical Center, University of Colorado School of Medicine, Denver, Colorado

**Address for correspondence** Ali A. Hakimi, University of Colorado - Anschutz Medical Campus, 13001 E 17th Pl, Aurora, Colorado 80045-2559, United States (e-mail: ali.hakimi@cuanschutz.edu).

Thorac Cardiovasc Surg Rep 2022;11:e67–e69.

## Abstract

### Keywords

- ▶ pneumothorax
- ▶ diaphragm
- ▶ pathology
- ▶ pleural disease
- ▶ pregnancy
- ▶ thoracoscopy/VATS

**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.

**Conclusion** Ectopic deciduosis is a rare cause of recurrent pneumothorax in pregnancy and should be considered when evaluating these patients.

## 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.<sup>1</sup> Ectopic decidualization most commonly affects the omentum and ovary.<sup>1</sup> 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

received  
July 3, 2022  
accepted after revision  
July 25, 2022

DOI <https://doi.org/10.1055/s-0042-1758115>.  
ISSN 2194-7635.

© 2022. The Author(s).

This is an open access article published by Thieme under the terms of the Creative Commons Attribution-NonDerivative-NonCommercial-License, permitting copying and reproduction so long as the original work is given appropriate credit. Contents may not be used for commercial purposes, or adapted, remixed, transformed or built upon. (<https://creativecommons.org/licenses/by-nc-nd/4.0/>)

Georg Thieme Verlag KG, Rüdigerstraße 14, 70469 Stuttgart, Germany



**Fig. 1** Friable endometrioid lesions on the diaphragm.

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 did not require additional intervention. The patient had an uncomplicated vaginal delivery at 38 weeks and has recovered uneventfully with a normal chest radiograph at 10 weeks postpartum.

## Discussion

Decidualization refers to the morphologic transformation of endometrial tissue as the uterus prepares for implantation, while decidual tissue found outside the uterine cavity is known as ectopic deciduosis.<sup>2</sup> This process is often compared to endometriosis, which can similarly present with ectopic endometrial-like glands and stroma. However, ectopic deciduosis is a unique diagnosis in that it occurs specifically

during pregnancy, is most commonly benign, and generally has complete resolution in the postpartum period.<sup>1</sup>

Furthermore, unlike the growing awareness of thoracic endometriosis syndrome,<sup>3</sup> pleuropulmonary manifestations of ectopic deciduosis are exceedingly rare. To our knowledge, this is the fifth reported case in the English literature, following the review work of Dudek et al in 2014.<sup>4-7</sup> Furthermore, our pathological findings of progesterone receptor positivity and absence of HMB45, are consistent with prior pathological cases and deciduosis criteria.<sup>8</sup> However, this is the first case to also document PAX8 presence in the decidual tissue.<sup>4-7</sup> Given the role of PAX8 in organogenesis of the Müllerian system,<sup>9,10</sup> this recent finding may further reassure the involvement of decidual tissue and provide an opportunity for further scientific inquiry. Due to its uncommon presentation and increased risks associated with operating on a gravid patient, it is likely that pneumothoraces secondary to ectopic deciduosis may be underdiagnosed or mislabeled as an irregular presentation of catamenial pneumothorax.

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.<sup>11-13</sup> 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

- Mendes J, Costa A. Ectopic decidualization: a forgotten entity [in Portuguese]. *Acta Med Port* 2016;29(01):63-72
- Mangla M, Nautiyal R, Shirazi N, Pati B. Ectopic cervical deciduosis: a rare cause of antepartum hemorrhage in mid trimester. *Eurasian J Med* 2021;53(02):152-154
- Soares T, Oliveira MA, Panisset K, et al. Diaphragmatic endometriosis and thoracic endometriosis syndrome: a review on diagnosis and treatment. *Horm Mol Biol Clin Investig* 2021;•••. Doi: 10.1515/hmbci-2020-0066
- Dudek W, Schreiner W, Strehl J, Sirbu H. Spontaneous pneumothorax due to ectopic deciduosis: a case report. *Thorac Cardiovasc Surg Rep* 2014;3(01):58-60

- 5 Flieder DB, Moran CA, Travis WD, Koss MN, Mark EJ. Pleuro-pulmonary endometriosis and pulmonary ectopic deciduosus: a clinicopathologic and immunohistochemical study of 10 cases with emphasis on diagnostic pitfalls. *Hum Pathol* 1998;29(12): 1495–1503
- 6 Kim YD, Min KO, Moon SW. Thoracoscopic treatment of recurrent pneumothorax in a pregnant woman: a case of ectopic deciduosus. *Thorac Cardiovasc Surg* 2010;58(07):429–430
- 7 Yoshioka H, Fukui T, Mori S, Usami N, Nagasaka T, Yokoi K. Catamenial pneumothorax in a pregnant patient. *Jpn J Thorac Cardiovasc Surg* 2005;53(05):280–282
- 8 Sorokin P, Nikiforchin A, Panin A, Zhukov A, Gushchin V, Kurtser M. Diffuse ectopic deciduosus imitating peritoneal carcinomatosis with acute abdomen presentation: a case report and literature review. *Case Rep Obstet Gynecol* 2020;2020:8847082
- 9 Gokulnath P, Soriano AA, de Cristofaro T, Di Palma T, Zannini M. PAX8, an emerging player in ovarian cancer. *Adv Exp Med Biol* 2021;1330:95–112
- 10 Khizer K, Padda J, Khedr A, et al. Paired-Box Gene 8 (PAX8) and its association with epithelial carcinomas. *Cureus* 2021;13(08):e17208
- 11 Garg R, , Sanjay, Das V, Usman K, Rungta S, Prasad R. Spontaneous pneumothorax: an unusual complication of pregnancy—a case report and review of literature. *Ann Thorac Med* 2008;3(03): 104–105
- 12 Georgopapadakos N, Lioumpas D, Mpenakis G, Tsochrinis A, Tsarna E. Primary spontaneous pneumothorax in a 32-week complicated pregnancy. *Cureus* 2021;13(05):e15037
- 13 Jain P, Goswami K. Recurrent spontaneous pneumothorax during pregnancy: a case report. *J Med Case Reports* 2009;3:81