

DOI: 10.4172/2471-9803.1000135

Long-term Follow-up of a Patient with Repeated Pneumothorax during Pregnancy and a History of Catamenial Pneumothorax: A Case Report

Daisuke Katsura^{1*}, Yoshihiko Hayashi², Takashi Hanatani³, Tomoya Kono⁴, Fuminori Kimura¹ and Takashi Murakami¹

¹Department of Obstetrics and Gynecology, Shiga University of Medical Science Hospital, Japan

²Department of Obstetrics and Gynecology, Nagahama Municipal Hospital, Japan

³Department of Respiratory Medicine, Nagahama Municipal Hospital, Japan

⁴Department of Thoracic Surgery, Nagahama Municipal Hospital, Japan

* **Corresponding author:** Daisuke Katsura, Department of Obstetrics and Gynecology, Shiga University of Medical Science, Setatsukinowa-cho, Otsu, Shiga 520-2192, Japan, Tel: +81-77-548-2267; Fax: +81-77-548-2406; E-mail: katsuo14@belle.shiga-med.ac.jp

Received date: October 15, 2016; **Accepted date:** October 31, 2016; **Published date:** November 09, 2016

Copyright: © 2016 Katsura D et al. This is an open-access article distributed under the terms of the Creative Commons Attribution License, which permits unrestricted use, distribution, and reproduction in any medium, provided the original author and source are credited.

Citation: Katsura D, Hayashi Y, Hanatani T. Long-term follow-up of a patient with repeated pneumothorax during pregnancy and a history of catamenial pneumothorax: A case report. *Crit Care Obst&Gyne.* 2016, 2:27.

Abstract

We report the subsequent course of a previously-reported (Japanese abstract) case of catamenial pneumothorax (CPT) involving seven recurrences of pneumothorax during pregnancy, treated by thoracoscopic surgery and then small thoracotomy segmental resection and pleural covering with polyglycolic acid felt and fibrin glue. The patient took an oral contraceptive (OC) from 6 months postpartum until discontinuation after 2 years to become pregnant. Pregnancy occurred after 1 year and an elective caesarean section was performed in the 37th week of pregnancy. During that period, there was no evidence of recurrence of pneumothorax.

Endometriosis-related pneumothorax during pregnancy recurs with high probability following medical management by thoracic drainage. For pneumothorax during pregnancy occurring on the right side followed by early recurrence after medical management by thoracic drainage, endometriosis should be considered to be a possible cause, particularly when there is a past history of endometriosis. In such cases, we recommend early thoracoscopic surgery comprising not only resection of the endometriosis lesions after careful evaluation but also pleurodesis and/or covering with polyglycolic acid felt and fibrin glue. Further studies are needed to clarify and develop appropriate management of pneumothorax in pregnant women with a history of CPT.

Keywords: Catamenial pneumothorax; Recurrence; Pregnancy; Endometriosis-related pneumothorax; Pneumothorax of pregnancy

Introduction

Catamenial pneumothorax (CPT) is a rare disease in which pneumothorax recurs in a pattern that corresponds with the menstrual cycle and is generally defined as not occurring during pregnancy. Most CPT involves the right side, and it is said to account for more than 20% of pneumothorax in women. Recently, CPT was reported to be primarily caused by the thoracic endometriosis syndrome (TES), and TES may develop without correspondence to the menstrual cycle. The diagnosis of TES is confirmed by observation of intrathoracic endometrial lesions and histopathological evidence of endometrial gland or stromal tissue; however, it is reportedly not found in 1/3 of CPT cases. If considering a diagnosis of CPT because of the clinical course, we often perform hormone therapy; the recurrence rate in these patients is approximately 50%. Although complete resection of endometrial lesions by thoracoscopic surgery is desirable, it is very difficult in CPT cases. The recurrence rate is approximately 30% following incomplete surgical resection. Therefore, combination therapy comprising thoracoscopic surgery using polyglycolic acid felt and fibrin glue, pleurodesis and hormone therapy, is considered [1-5].

To our knowledge, currently there are only eight case reports (including that of the current patient's first pregnancy) regarding endometriosis-related pneumothorax during pregnancy. None of these documented the subsequent pregnancy history of the patients. Here we report a second full-term pregnancy in a patient who had recurrent pneumothorax during the first full-term pregnancy (as previously reported in a Japanese abstract) and propose a management strategy for endometriosis-related pneumothorax during pregnancy.

Case Presentation

A 34 year-old female patient undergoing pregnancy checkups was admitted to our hospital because of dyspnea in the 24th

week of her third pregnancy. The past history included laparoscopic cystectomy for bilateral endometrial cysts and thoracoscopic lung and diaphragm segmental resection for right-sided CPT at 30 years of age. At 31 years of age, she developed a right-side pneumothorax and began taking an oral contraceptive (OC) after thoracic drainage. At 33 years of age, after discontinuing the OC to become pregnant, a rare, right inguinal endometrial mass was extracted, and after this she had a spontaneous abortion.

At the current admission, she was diagnosed with a right pneumothorax by chest X-ray. Because the lungs expanded after thoracic drainage and air leakage resolved, the drain was removed after 3 days. However, pneumothorax recurred the next day and thoracic drainage was performed again. Minor leakage persisted. She then underwent thoracoscopic surgery in the 27th week of pregnancy. In spite of careful, close-up observation, no clear bulla or air leakage was noted. Because air leakage resolved, the drain was removed and she was discharged on the 3rd postoperative day. However, pneumothorax recurred in the 28th, 30th (twice), 31st, and 33rd weeks of pregnancy, and she was admitted for thoracic drainage accordingly. Finally, without removing the thoracic drain placed to treat a recurrence during the 35th week of pregnancy, an elective caesarean section was performed in the 37th week.

After the delivery, air leakage persisted during the puerperium. She then underwent thoracoscopic surgery, followed by a small thoracotomy on the 15th postoperative day. The red plaques in the pleura of the interlobar surface of the right lower lobe and air leakage from around three lobes merging at a section of the right middle lobe were found, resected, and covered with polyglycolic acid felt and fibrin glue. Interlobar surfaces of the upper, middle and lower lobes were covered with surgical sheets using fibrin glue. By histopathological examination, fibrosis and granulation tissue formation were demonstrated, but there was no evidence of endometriosis. However, because of the clinical course and surgical history, we think that collapse of the pleura from an endometrial lesion was the cause of this pneumothorax.

Postoperative recovery was uneventful and the patient was discharged on the 5th postoperative day. She took an OC from 6 months to 2 years postpartum, and had no pneumothorax relapses during withdrawal bleeding and subsequent spontaneous menstruation. At age 37, she conceived again. No pneumothorax occurred during the course of that pregnancy. After a prenatal checkup revealed no problems, an elective caesarean section was performed in the 37th week of that pregnancy at age 38. There was no evidence of recurrence of pneumothorax during the puerperal period.

Discussion

This patient's prior pregnancy was reported previously as an abstract in Japanese; here we report the subsequent course,

including a subsequent pregnancy. The final thoracoscopic surgery after the first childbirth was very effective, and there was no evidence of recurrence of pneumothorax for 5 years, including during the next pregnancy. With the inclusion of this case, to our knowledge, there are only eight reported cases regarding CPT, endometriosis and pregnancy, by searches in English of MEDLINE and PubMed, and in Japanese of IGAKU-CHUO ZASSHI [6-13]. **Table 1** shows the eight patients' ages, the past history of endometriosis, gestational age of pneumothorax onset, relapse-free interval, onset side, initial and final treatments, causes, postpartum recurrence, and modes of delivery. The women's ages ranged from 27 to 37 years (median 32 years); five cases were multipara and three cases were primipara. Including clinical diagnoses, six cases (75%) had a past history of endometriosis, five (83%) of which were CPT. Of these, two CPT cases took an OC; in our case there was no recurrence during the use of the OC, but in the other case pneumothorax recurred twice on the OC. All cases of CPT, except for ours, were in remission following spontaneous or thoracic drainage, and thoracoscopic surgery was not performed. The patient having pleural effusion and ascites retention in her past history was treated surgically, but the details were not documented. Regarding pneumothorax during pregnancy, five cases (62.5%) developed pneumothorax in the second trimester, two cases (25%) in the third trimester and one case (12.5%) in the first trimester (the 8th week). In all cases, pneumothorax onset was on the right side. Recurrence occurred early, from 1 day to 47 days (median 13 days) later, even if managed medically by thoracic drainage (in our case it recurred 7 times); and ultimately, surgery was needed in all except one case, in which lung biopsy was performed (details were not documented). Thoracoscopic surgery was performed during the pregnancy in five cases, during the puerperium in one case, during both in our case, and was not documented in one case. In our case, thoracoscopic surgery during the prior full-term pregnancy resulted in no resection or pleurodesis, and there were repeated pneumothorax recurrences soon after the surgery. In CPT patients, postoperative recurrence is often seen [3] because it is difficult to detect microscopic lesions during surgery. In our case, the additional small thoracotomy or the combination of subsequent covering and pleurodesis may have helped to prevent recurrence [4]. As for final treatments, of the five cases with reported details that underwent thoracoscopic segmental lung resection, one had pilmorrhaphy, two underwent pleurodesis, two underwent covering, and two underwent hormone therapy in combination with the other therapies. There was no evidence of pneumothorax recurrence in most cases during a short follow-up period, and Flieder et al. reported a follow-up phase more than 5 years for one patient; however, to our knowledge, this is first report of a long follow-up period including the next pregnancy outcome.

Table 1 Comparison of eight case reports.

Author	Age (Yr)	MP /PP	PHE	GA at pneumothorax			Final treatment	Cause of pneumothorax	PR	Mode of delivery		SI
				Onset weeks	Onset side	IT				GA	Week	
Kono et al. [6]	34	MP	Endometrial cyst, CPT Lung, Diaphragm, Surgery, OC	24, 28, 30, 30, 31, 33, 35	Right	Thoracic drainage	Puerperium Thoracoscopic partial resection of the lung Covering OC	Collapse of the pleura by a suspected stray endometrial lesion	None	CS	37	Maternal adaptation
Kim et al. [7]	34	PP	None	18, 22	Right	Thoracic drainage	GA 22 weeks Thoracoscopic resection of bulla	Diaphragmatic fenestration, Ectopic decidual change of endometriosis	None	VD	39	-
Oonohara et al. [8]	33	PP	Pleural effusion ascites, Pleura, Diaphragm, endometriosis, Surgery	32	Right	Thoracic drainage	Puerperium Thoracoscopic pulmorrhaphy	Weakening of the lung pleura by decidual change of endometriosis	None	CS	37	-
Nakamura et al. [9]	36	MP	Endometrial cyst suspected, Danazol 4 months, CPT s/o, Thoracic drainage	16, 23, 26	Right	Thoracic drainage	GA 29 weeks, Thoracoscopic resection of bulla Pleurodesis (autologous blood)	Cyst of the lung pleura by decidual change of endometriosis	None	VD	39	-
Kaneko et al. [10]	37	MP	Endometrial cyst suspected, Danazol 4 months, CPT s/o, Thoracic drainage	16, 22	Right	Thoracic drainage	GA 28 weeks, Thoracoscopic partial resection of the lung	Cyst of the lung pleura by decidual change of endometriosis	-	-	-	-
Hiromu et al. [11]	29	PP	CPT suspected, Thoracoscopic surgery, No evidence of endometriosis, Danazol 6 months	8	Right	Thoracic drainage	GA 15 weeks Thoracoscopic partial resection of the lung Pleurodesis (OK-432) OC	Cyst of the lung pleura by decidual change of endometriosis	None	AA	AA	-
Schoenfeld et al. [12]	27	MP	CPT suspected, 5 times (twice during use of the OC), 3 times spontaneous remission, Twice thoracic drainage	24	Right	Thoracic drainage	Thoracoscopic surgery	Diaphragmatic fenestration suspected	-	-	-	-
Flieder et al. [13]	27	MP	Pulmonary nodule, 2 years observation	28	Right	Thoracic drainage	Lung biopsy Thoracic drainage	Decidual change of endometriosis in the lung interstitium	None	VD	39	-

GA: Gestational Age, MP: Multipara, PP: Primipara, CPT: Catamenial Pneumothorax, OC: Oral Contraceptive, CS: Cesarean Section, VD: Vaginal Delivery, AA: Artificial Abortion, Covering: Pleural covering with polyglycolic acid felt and fibrin glue, PHE: Past History of Endometriosis, PR: Postpartum Recurrence, SI: Surgical induction, IT: Initial Treatment

Regarding the previously-reported causes of pneumothorax, histopathological analysis showed decidual change of the endometriosis induced by pregnancy in seven cases (82.5%). In the other case, small diaphragmatic fenestrations were observed, but there was no evidence of endometriosis. For the

modes of delivery, three cases were vaginal birth and two cases were caesarean section. In our case, because there was maternal fatigue due to the persistence of pneumothorax, we performed an elective caesarean section. In the other case, the

adaptation of caesarean section was documented. Basically, caesarean section should be performed by obstetric adaptation.

The recurrence rate of spontaneous pneumothorax during pregnancy is about 40% [14]; however all of the reported cases recurred even if treated by thoracic drainage, and ultimately surgery was needed except for one case. For pneumothorax during pregnancy having right side onset and early recurrence after medical management by thoracic drainage, endometriosis should be considered to be a possible cause, particularly in patients with a past history of endometriosis. Care managers serving as a bridge between physicians, specialists, and patients, collaborating with the patients' doctors while working directly with patients. They must be careful about such symptoms of the patient. In such cases, we would like to recommend early thoracoscopic surgery comprising not only resection of the endometriosis lesions after careful evaluation, but also pleurodesis and/or covering with polyglycolic acid felt and fibrin glue. Further studies are needed to clarify and develop appropriate management of pneumothorax of pregnant women with a history of CPT; however, as the long follow-up of the current case shows, if the surgery is successful, recurrence of pneumothorax during the concurrent and subsequent pregnancies may be prevented.

Competing Interests

The authors declare that they have competing interests.

Acknowledgements

It is acknowledged that all authors have contributed significantly and that all authors are in agreement with the content of the manuscript.

References

- Alifano M, Trisolini R, Cancellieri A, Regnard JF (2006) Thoracic endometriosis: Current knowledge. *Ann Thorac Surg* 81: 761-769.
- Alifano M, Jablonski C, Kadiri H, Falcoz P, Gompel A, et al. (2007) Catamenial and non-catamenial. Endometriosis-related or nonendometriosis-related pneumothorax referred for surgery. *Am J Respir Crit Care Med* 176: 1048-1053.
- Bagan P, Barthes FLP, Assouad J, Souilamas R, Riquet M (2003) Catamenial pneumothorax: Retrospective study of surgical treatment. *Ann Thorac Surg* 75: 378-381.
- Alifano M, Legras A, Rousset-Jablonski C, Bobbio A, Magdeleinat P, et al. (2011) Pneumothorax recurrence after surgery in women: clinicopathologic characteristics and management. *Ann Thorac Surg* 92: 322-326.
- Began P, Berna P, Assouad J, Hupertan V, Barthes FLP, et al. (2008) Value of cancer antigen125 for diagnosis of pleural endometriosis in females with recurrent pneumothorax. *Eur Respir J* 31: 140-143.
- Kono T, Matsui C, Takubo Y, Hanatani T, Noguchi T (2012) A case of catamenial pneumothorax during pregnancy. *Shiga-Igaku* 34: 138-139.
- Kim YD, Min KO, Moon SW (2010) Thoracoscopic Treatment of Recurrent Pneumothorax in a Pregnant Woman: A Case of Ectopic Deciduos. *Thorac Cardio Surg* 58: 429-435.
- Onohara Y, Ohata Y, Ikeno S, Okada M, Minagawa Y (2009) A case of pneumothorax caused by thoracic endometriosis during pregnancy. *Tottori-Igaku Zasshi* 37: 47-48.
- Nakamura M, Andoh A, Mizutake S, Asahina H, Saitoh M, et al. (2008) A case of pneumothorax repeated during pregnancy. *Saitama J Obstet Gynecol* 38: 3-6.
- Kaneko K, Adachi A, Koh E, Kadoyama C, Matsushima H, et al. (2007) A case of pneumothorax relapsed during pregnancy. *Jpn J Diagnos Pathol* 24: 260-262.
- Yoshioka H, Fukui T, Mori S, Usami N, Nagasaka T, et al. (2005) Catamenial Pneumothorax in a Pregnant Patient. *Jpn J Thorac Cardio Surg* 53: 280-282.
- Schoenfeld A, Ziv E, Zeelel Y, Ovadia J (1986) Catamenial pneumothorax- a literature review and report of an unusual case. *Obstet Gynecol Surv* 41: 20-24.
- Flieder DB, Moran CA, Travis WD, Koss MN, Mark EJ (1998) Pleuro-pulmonary endometriosis and pulmonary ectopic deciduos: a clinicopathologic and immunohistochemical study of 10 cases with emphasis on diagnostic pitfalls. *Human Pathol* 29: 1495-1503.
- Tanase Y, Yamada T, Kawaryu Y, Yoshida M, Kawai S (2007) A case of pneumothorax during pregnancy and review of the literature. *Kobe J Med Sci* 53: 251-255.