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

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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...
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 in 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 (62.5%) 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.
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

Table 1 Comparison of eight case reports.

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


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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.

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