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CASE REPORT

Surgical treatment of catamenial pneumothorax: Report of three cases

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Summary Catamenial pneumothorax (CP) is a rare entity of spontaneous, recurring pneumothorax in females. Although it has been known to be associated with thoracic endometriosis, varying clinical course and the lack of consistent intraoperative findings have led to conflicting etiological theories. We herein discuss the etiology, clinical course, and surgical treatment of three patients with CP. Three females (aged 40 years, 28 years, and 34 years) had recurrent right-sided spontaneous pneumothoraces that coincided with their menses. They had undergone video-assisted thoracoscopic surgery (VATS) previously. Blueberry spots in the right diaphragm were detected in all three cases. Two patients had recurrence, postoperatively. The other patient, who received luteinizing hormone-releasing hormone analog therapy for an abdominal endometriosis in the perioperative period and postoperative chemical pleurodesis to prevent recurrence, has been free of recurrence for 15 months, postoperatively. However, pelvic endometriosis was detected in this patient only. Therefore, CP should be suspected in ovulating females with spontaneous pneumothorax, even in the absence of any symptoms associated with pelvic endometriosis. In addition, while performing VATS, careful inspection of the diaphragmatic surface is important. In complicated cases, hormonal suppression therapy and chemical pleurodesis might also be helpful adjunct modalities.

1. Introduction

Catamenial pneumothorax (CP) is a type of spontaneous recurring pneumothorax that coincides with the menses in females. The first report on CP was published in 1958,[1] and since then it has always been considered as a usual

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condition for various reasons, especially due to its high recurrence rate.[2] In the past decade, the accuracy of detecting CP has improved and subsequently more number of cases are being reported, comprising almost one-third of all spontaneous pneumothorax cases in females.[3,4]

The CP is generally considered to be the most frequent presentation of thoracic endometriosis syndrome, although histological findings of endometriosis during surgery are rare.[5] The etiology of CP is most likely to be multifactorial in origin, involving a combination of different mechanisms.[6–8]

The optimal management of CP also remains unclear, as the traditional treatments for pneumothorax are often unsuccessful. The purpose of this study is to report three typical cases of CP and to discuss the pathogenesis of CP.

2. Case reports

We performed a retrospective review from January 2001 to December 2011 of all female patients undergoing surgery for spontaneous pneumothorax at the Kitakyushu Municipal Medical Center (Fukuoka, Japan). Fourteen patients with recurrent spontaneous pneumothorax received surgical treatment. Of these, three (21.4%) were classified as having CP. The diagnosis of CP was made based on preoperative symptoms that coincided with their menses as well as on the intraoperative pathological findings.

2.1. Case 1

In December 2008, a healthy 40-year-old female had a right-sided spontaneous pneumothorax and was treated with a thoracic tube. Because of a recurrence after conservative treatment, video-assisted thoracoscopic surgery (VATS) was performed. We noted a fenestration-like lesion in the tendinous part of the diaphragm (Fig. 1A) and a subpleural bulla in the apex of the right lung. A partial resection of the diaphragm and a bullectomy were performed. A histological examination of the resected tissues confirmed an endometrial implant in the bulla, but not in the diaphragm. The fenestration-like lesion had no fenestration microscopically, and only some hemosiderin-laden macrophages were observed. Immunohistochemical staining was positive for the estrogen receptor within the stroma of the endometrial implant in the bulla (Fig. 1B). The patient had a recurrence that coincided with her menses in February 2009. Chemical pleurodesis using OK432 was performed twice after thoracic drainage. After recovery, the patient has been symptom-free for 47 months.

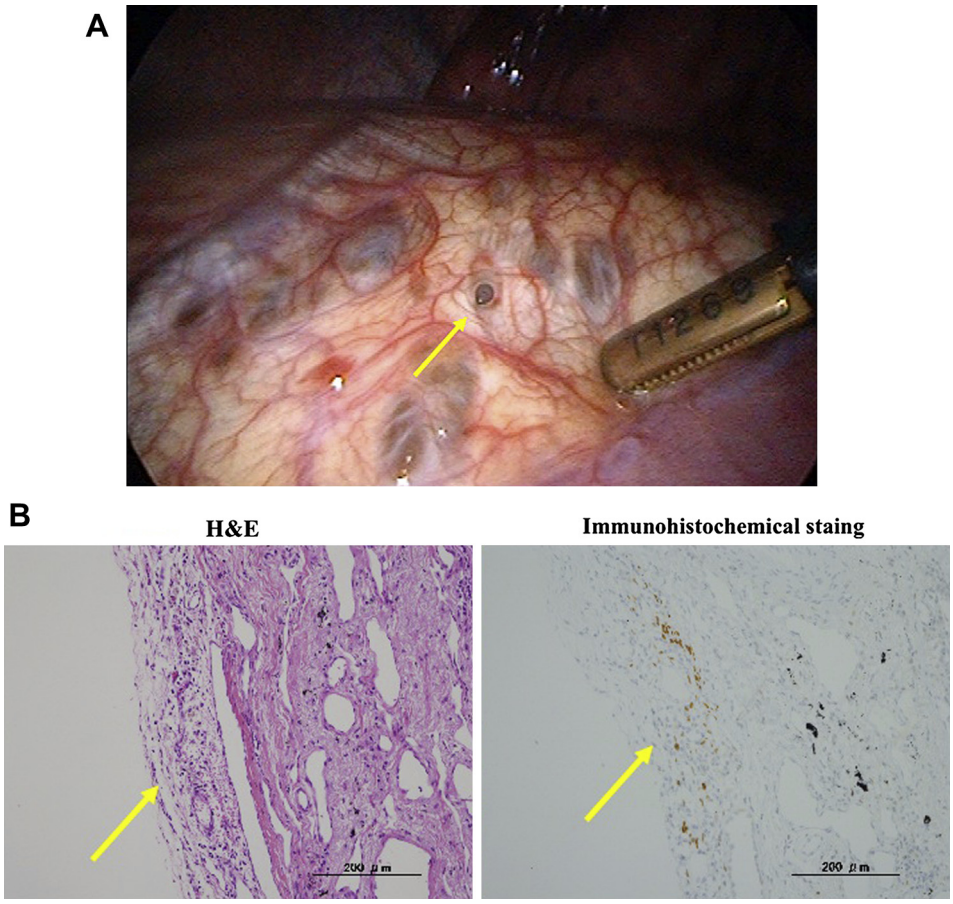

Figure 1 (A) Intraoperative findings of a perforation-like lesion in the tendinous part of the diaphragm. (B) A focus of an endometrial implant of the visceral pleura. Immunohistochemical analysis showed strong nuclear staining for the estrogen receptor (100×). H&E = hematoxylin and eosin.