Surgical treatment of catamenial pneumothorax: a single centre experience

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Abstract

We retrospectively reviewed our experience with catamenial pneumothorax (CP) in terms of treatment and follow-up. From 1993 to 2008, ten women presented at our department with CP. CP was right-sided in all patients: seven presented diaphragmatic defects including one endometriosis, five had apical bulla or blebs that in three patients were the only pathological findings. Surgical approach was thoracoscopic with a muscle-sparing thoracotomy when diaphragmatic defects were present. All patients underwent apical resection and apical pleurectomy associated in seven cases with diaphragmatic plication and chemical pleurodesis. After surgery nine patients underwent apical pleurectomy were performed in all patients. Chem-

Keywords: Catamenial pneumothorax; Thoracic endometriosis; Hormonal therapy

1. Introduction

Catamenial pneumothorax (CP) is a kind of spontaneous recurring pneumothorax that occurs in women and is correlated with menses.

Since the first report appeared in literature in 1958 [1], it has always been considered an unusual condition [2]. In the last decade the recognition of CP has improved and its frequency now has been reported, reaching almost one-third of all spontaneous pneumothorax in women [3, 4].

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

Correct management of CP also remains unclear as the traditional treatment for pneumothorax often seems to be unsuccessful. In the light of these uncertainties we reviewed our experience with CP in terms of treatment and follow-up.

2. Materials and methods

From January 1993 to January 2008, among 56 women surgically treated for spontaneous pneumothorax in our department, ten patients (17.8%) were diagnosed as having CP. The clinical criteria for suspecting CP in absence of histologically confirmed endometriosis or diaphragmatic defects, was recurrent pneumothorax documented by chest radiograph during or following menstruation (within 24–72 h). Patients’ mean age was 32 ± 11 years (range 21–44). Preoperative assessment included a chest X-ray, routine blood tests, ECG and blood gas analysis. Seven patients also underwent a CT scan. Two women had previously been treated through video-assisted thoracoscopy (VATS) elsewhere. VATS was the surgical approach of choice. It was performed under general anaesthesia with double-lumen bronchial intubation and three ports. The lung was inspected for blebs or bulla and possible air leaks were identified by inflating the lung under saline solution. Signs of thoracic endometriosis were sought by inspection of the lung, pleura and diaphragm. The last one was also carefully inspected for the presence of defects. When diaphragmatic lesions were found, a muscle-sparing video-assisted thoracotomy was performed to resect and to repair the diaphragm that was obtained by means of plication. Apical resection and apical pleurectomy were performed in all patients. Chem-
ical basal pleurodesis with talc (1 g) was given to patients with diaphragmatic defects. Thoracic endometriosis was considered histologically ‘proven’ when endometrial glands and stroma were present. Before discharge all patients were referred to a gynaecologist for further investigations and medical therapy.

3. Results

Pneumothorax occurred within 24 h of the onset of menses in three patients, within 48 h in four, and within 72 h in three patients. Symptoms included chest pain in all patients, cough in three, shortness of breath in two cases and hemotorax in one patient. The number of preoperative episodes varied from zero to four (mean 2 ± 2). Pneumothoraces were unilateral, recurrent and right-sided in all ten patients; one patient had also a history of left-sided pneumothorax treated conservatively elsewhere. Eight patients had a chest tube before surgery. Associated comorbidity included pelvic endometriosis in two patients. Chest computed tomography showed an endometrial implant only in one patient (Fig. 1).

Diaphragmatic defects were observed in seven patients (70%) (Fig. 2) one of whom was found to have a diaphragmatic endometrial implant which was histologically confirmed (patient 4; Fig. 3). Apical bulla or blebs alone were detected in three patients (30%). Postoperative complications consisted in one case of prolonged air leak (patient 6). The mean duration of chest tube drainage was 4±1 days (range 3–9). None of the patients underwent hormonal therapy before surgery. Postoperative hormonal treatment was proposed to all patients but one patient refused. Three patients at the beginning of our experience were put on hormonal estrogen–progesterone complex and six received gonadotropin-releasing hormone agonist (GnRH agonist) for six months in all cases. One patient was unable to tolerate the six-month treatment and suspended it after two months.

The mean follow-up was 52±32 months (range 14–168). Recurrences occurred in four patients (40%) that were all initially put on estrogen–progesterone treatment. After recurrence, however, these patients received GnRH agonist therapy. Recurrence was localized in the lower side in two patients and laterally in the other two. Three patients were managed positioning a chest tube and one was treated conservatively. At the present time all ten patients are well with no recurrence of CP. Pathologic findings, treatment and outcome are presented in Table 1.

4. Discussion

CP has always been considered a rare cause of spontaneous pneumothorax with a reported incidence in the past of 2.8–5.6% [9]. Recent series suggest that this incidence in the reality is around 30% [2, 9] probably due to an increased awareness of this pathology.

In this study, according to the literature [2, 3, 5, 9] we defined CP as recurrent spontaneous pneumothorax occurring within 24 h before and 72 h after the onset of menses. Diagnosis of CP is most often made clinically although in some cases computed tomography and magnetic resonance imaging have been reported to be useful [10].

A number of hypotheses have been suggested to explain the aetiology of catamenial pneumothorax. According to one hypothesis the open connection between the atmosphere and the peritoneal cavity during menstruation allows air to migrate into the thoracic cavity through diaphragmatic fenestrations and porosities [4, 6]. Another hypoth-
Table 1
Patient characteristics, treatment and outcome

<table>
<thead>
<tr>
<th>Patients</th>
<th>Age</th>
<th>Intraop. findings</th>
<th>Treatment</th>
<th>Chest tube (days)</th>
<th>Hormonal treatment</th>
<th>Recurrence</th>
<th>Recurrence treatment</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>37</td>
<td>Diaphragm defect</td>
<td>DR, AP, AR, CP</td>
<td>4</td>
<td>GnRH agonist</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>2</td>
<td>36</td>
<td>Diaphragm defect</td>
<td>DR, AP, AR, CP</td>
<td>4</td>
<td>GnRH agonist</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>3</td>
<td>44</td>
<td>Blebs or bulla</td>
<td>AP, AR</td>
<td>3</td>
<td>Refused</td>
<td>Yes</td>
<td>Chest tube</td>
</tr>
<tr>
<td>4</td>
<td>25</td>
<td>Diaphragm defect, endometriosis</td>
<td>DR, AP, AR, CP</td>
<td>3</td>
<td>Estrogen–progesterone</td>
<td>Yes</td>
<td>Conservative treatment</td>
</tr>
<tr>
<td>5</td>
<td>26</td>
<td>Blebs or bulla</td>
<td>AP, AR</td>
<td>3</td>
<td>Not tolerated</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>6</td>
<td>26</td>
<td>Diaphragm defect</td>
<td>DR, AP, AR, CP</td>
<td>9</td>
<td>Estrogen–progesterone</td>
<td>Yes</td>
<td>Chest tube</td>
</tr>
<tr>
<td>7</td>
<td>42</td>
<td>Diaphragm defect</td>
<td>DR, AP</td>
<td>4</td>
<td>GnRH agonist</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>8</td>
<td>21</td>
<td>Blebs or bulla</td>
<td>AP, AR</td>
<td>3</td>
<td>Estrogen–progesterone</td>
<td>Yes</td>
<td>Chest tube</td>
</tr>
<tr>
<td>9</td>
<td>41</td>
<td>Diaphragm defect + blebs</td>
<td>DR, AP, AR, CP</td>
<td>4</td>
<td>GnRH agonist</td>
<td>No</td>
<td></td>
</tr>
<tr>
<td>10</td>
<td>34</td>
<td>Diaphragm defect + blebs</td>
<td>DR, AP, AR, CP</td>
<td>3</td>
<td>GnRH agonist</td>
<td>No</td>
<td></td>
</tr>
</tbody>
</table>

DR, diaphragmatic repair; AP, apical pleurectomy; AR, apical resection; CP, chemical pleurodesis; GnRH agonist, gonadotropin-releasing hormone agonist.

Diaphragmatic defects are caused by endometriosis and the third involves a metastatic spreading of endometriosis through the uterine veins into the venous system [7]. Lastly, prostaglandin F2, a potent constrictor of bronchioles and vascular structures which can be found in the plasma of some women during menstruation, may destroy alveolar tissue due to vasospasm, thus leading to pneumothorax [4, 6–8].

Endometriosis seems to play the most important role in the development of CP, although the association with pelvic endometriosis has been reported in 20–70% of patients [11] which is close to the percentage of patients in our series (20%) with a history of pelvic endometriosis. It seems that diaphragmatic defects are strictly correlated with endometriosis.

Diaphragmatic defects in CP range from 29 to 66% of patients [4, 7]. Some authors [9] report concomitant diaphragmatic defects and endometrial implants but the association is not widely observed [12]. In our series, despite seven patients (70%) presenting with diaphragmatic fenes-trations, only in one case it was associated with endome-trial implant. In consideration of this evidence we do not perform routinely chest computed tomography and/or magnetic resonance that would be useful only in case of evident thoracic endometrial implant [10]. Two of our patients had been previously treated elsewhere through a VATS procedure where diaphragmatic defects had been unnoticed and were thus untreated with consequent recurrence of pneumothorax. Only blebs and bulla were found in patients 3, 5 and 8 during surgery and although the diaphragm had been carefully inspected there were no signs of diaphragm defects or porosities. However, it is likely that small diaphragmatic defects were present, since two out of the three patients had recurrences after surgery. In order to avoid recurrences leaving behind small defects, we performed also localized chemical pleurodesis on the diaphragm; some authors, to achieve the same results, propose the apposition of a mesh [7, 13]. Despite diaphragmatic chemical pleurodesis, patients 4 and 6 recurred although recurrence was localized in the lateral side of the pleural cavity. Since only apical pleurectomy was carried out in all patients we suggest subtotal pleurectomy to prevent recurrences not connected with diaphragm defects.

There is general consensus in the literature that the initial approach to CP must be minimally invasive and therefore performed through a thoracoscopy. VATS, in fact, provides magnification and exposure of possible defects that are sometimes better than that provided by thoracotomy. Misdiagnosis may occur, however, especially if the patient is positioned for an axillary thoracotomy since complete visualization of diaphragm is difficult. A better approach seems to be a VATS with the patient positioned for a postero-lateral thoracotomy [12, 14]. In our experience, seven patients underwent a video-assisted muscle-sparing thoracotomy (as a second procedure to repair diaphragmatic defects) and three underwent a VATS treatment alone. If any doubt or if the correction of the diaphragmatic defect is impossible it is better to perform a postero-lateral thoracotomy and not an axillary one. Bullectomy and/or resection of dystrophic apex are useful not only to prevent recurrences but also for histological purposes.

The incidence of recurrence can be drastically reduced if an adequate pleurodesis is scheduled as part of the operation. Authors agree on pleural abrasion [6, 7, 12, 14], while pleurectomy and chemical pleurodesis are not universally accepted [6, 14]. We performed apical pleurectomy in all patients but recurrences also occurred on the lateral side suggesting that subtotal pleurectomy might represent the best solution to prevent recurrence of pneumothorax.

The association of surgery and medical treatment seems to provide better results than surgery alone for the cure of CP [6, 15]. All of our patients were seen postoperatively by a gynaecologist, although some authors [9] do not consider important this cooperation since pelvic endometriosis is not always associated to CP. Results reported in literature suggest otherwise, that the association of surgery and hormonal treatment is the best choice for the cure of CP [2, 10]. Three patients of our series (patients 4, 6 and 8) were put on an estrogen–progesterone therapy after surgery at the beginning of our experience, which was unsuccessful as all these patients had a recurrence. Therapy with GnRH agonist can instead help surgery in CP [6, 7]. The rationale underlying hormonal control is based on the presence of endometrial implants which are hormonal dependent, and substances known to suppress the growth of ectopic endometrium would also be active on the pulmonary implants. Hormonal control has risks and side-
effects and treatment cannot therefore exceed six months. Nevertheless, this interval is considered sufficient to avoid recurrences [15].

In our experience, the estrogen–progesterone treatment was unsuccessful in all cases while GnRH agonist therapy seems to improve the outcome of treatment of CP. After failure with estrogen–progesterone therapy all patients were put on GnRH agonist for six months and no recurrence occurred.

In conclusion, when a young woman presents with a pneumothorax during menses, CP should be suspected and an accurate investigation needs to be carried out to obtain a correct diagnosis. At the time of surgery the diaphragm should be carefully inspected for defects and/or endometriosis: all defects should be corrected and all nodules resected. Standard pleurodesis may not be sufficient to avoid recurrences in CP patients, so we suggest subtotal pleurectomy associated with chemical pleurodesis on the diaphragm as an initial approach. Hormonal treatment with GnRH agonist for six months seems to be effective in enhancing surgical results.

References


