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Catamenial pneumothorax: surgical repair of the diaphragm and hormone treatment

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ABSTRACT

Catamenial pneumothorax is defined as spontaneous pneumothoraces occurring within 72 h before or after onset of menstruation. It is rare but clinical index of suspicion should be high in ovulating women with spontaneous pneumothoraces. The mechanism is unclear but is thought to involve pre-existing or acquired diaphragmatic defects and endometrial implants. Traditional therapy involving hormonal treatment or surgical pleurodesis alone is associated with high rates of recurrence. A series of four patients with catamenial pneumothorax managed at our institution is presented to highlight the condition to various surgical specialties to whom it may present, and to emphasise the importance of both surgical and hormonal interventions in preventing recurrence. Each patient underwent video-assisted thoracoscopic inspection of the diaphragm, mechanical pleurodesis and, most importantly, repair of diaphragmatic defects with an artificial mesh. Surgical treatment was strictly followed by a course of gonadotrophin-releasing hormone analogue therapy in three patients, with no recurrence to date (longest follow-up 45 months). The fourth patient suffered a postoperative recurrence when hormonal treatment was delayed for 6 weeks, stressing the importance of hormonal treatment in conjunction with surgery.

KEYWORDS

Catamenial pneumothorax – Videothoracoscopy – Diaphragm – Artificial mesh – Endometriosis

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A retrospective review of all female patients undergoing surgery for spontaneous pneumothorax at our tertiary referral centre from June 1999 to October 2003 revealed four women with recurrent spontaneous pneumothoraces, (aged 34–40 years; mean, 37 years) who reported symptoms coinciding with the onset of menstruation (Table 1). Each patient had suffered at least two recurrences; one with as many as fourteen. Previous treatment had been exclusively tube thoracostomy at their referring institutions. Clinical suspicion for catamenial pneumothorax was high as each patient had suffered from pelvic endometriosis and two had been laparoscopically diagnosed with abdominal endometriosis. Two patients suffered relapses after stopping the oral contraceptive pill.

At surgery, diaphragmatic fenestrations measuring more than 10 mm were identified in each patient (Fig. 1). Endometrial nodules in the visceral pleura were visualised in one patient. The diaphragmatic defects were repaired with a polyglactin (Vicryl®) or polypropylene mesh (Prolene®). Three were secured with endoscopic sutures, one with surgical steel clips (Endoclips®) and the last with glue (Tisseel®). All had pleural abrasion or full pleurectomies. Three patients also underwent talc poudrage to encourage the formation of fibrotic adhesions between diaphragmatic and lung surfaces. Pulmonary blebs or bullae were not visualised. Three weeks' postoperatively, three patients commenced gonadotrophin releasing hormone (GnRH) analogue (goserelin) therapy for 6 months. To date, follow-up has reached 49 months (mean, 33 months) with no recurrence. The remaining patient, who had a polypropylene mesh secured over her diaphragm with Tisseel® glue and talc pleurodesis, suffered a recurrent basal pneumothorax 6 weeks' post-operatively, 2 days before she was due to commence GnRH treatment. This was managed conservatively with tube thoracostomy and a subsequent 6-month course of GnRH therapy, with no further recurrence (follow-up 16 months).

Discussion

Catamenial pneumothorax is rare but should be suspected in ovulating women with spontaneous pneumothorax, even if they do not have a previous history of endometriosis.[1] The timing of recurrences with onset of menstruation is classic,

Table 1 Patient details

Patient no.	Age (yr)	Pelvic endomet. (P); abdominal endomet. (A)	Previous surgical pleuro-desis	Side of recur-rence	No. of recur-rences	Pathological findings	Surgical treatment (all VATS approach)	Hormonal treatment (months)	Follow-up (months)
1	34	P & A	No	Right	14	Diaphragmatic fenestrations	Talc pleurodesis; pleurectomy; diaphragm cover with vicryl mesh (secured with sutures)	6	45
2	35	P & A (previous colonic resection due to endometrioma)	No	Right	4	Diaphragmatic fenestrations	Talc pleurodesis; pleural abrasion; diaphragm cover with vicryl mesh (secured with sutures)	6	40
3	40	P	No	Right	4	Diaphragmatic fenestrations	Talc pleurodesis; pleural abrasion; diaphragm cover with prolene mesh (secured with clips)	6	19
4	38	P	No	Right	2	Diaphragmatic fenestrations	Pleural abrasion; diaphragm cover with vicryl mesh (secured with Tisseel glue)	6	16

often occurring within 72 h. A gynaecological history and assessment of the menstrual cycle should be taken for every woman presenting with recurrent spontaneous pneumothoraces. Recent reports indicate that its incidence may be more common than historically thought.[2–4] Before the advent of video-assisted thoracoscopy in 1990, surgical management of recurrent spontaneous pneumothorax was axillary thoracotomy and apical pleurectomy, which did not always allow appropriate inspection of the diaphragm.[5] The diagnosis of catamenial pneumothorax may have thus been missed, perhaps explaining its low incidence pre-1991.

The aetiology of catamenial pneumothorax is unknown: the spontaneous rupture of blebs during hormonal changes, alveolar rupture due to prostaglandin-induced bronchiolar constriction, and sloughing of ectopic endometrial implants in the visceral pleura during menstruation with resultant air

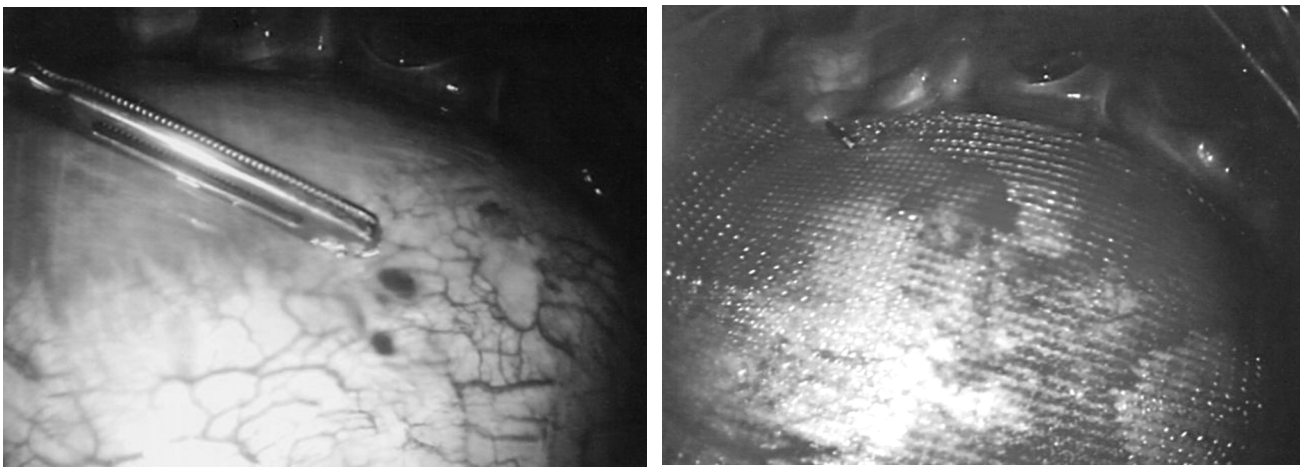

Figure 1 (a) Diaphragmatic fenestrations and endometrial implants in the central tendon of the right hemidiaphragm. (b) Diaphragmatic defects covered with Prolene® mesh.

leaks, may all play a role.[2] An absent cervical mucous plug may lead to air tracking from the genital tract into the peritoneum and through diaphragmatic fenestrations into the pleural space.[3] A clear relationship with endometriosis has not been established; certainly, many patients with catamenial pneumothorax do not have a pre-operative diagnosis of endometriosis.[4] Catamenial pneumothorax predominantly occurs on the right side, as the clockwise peritoneal circulation from the pelvis to the right subphrenic space sweeps endometrial implants to the right diaphragm. The 'piston effect' of the liver then transmits intraperitoneal pressure spikes through pre-existing diaphragmatic defects, forcing air into the pleural space.[2]

At our institution, video-assisted thoracoscopy is carried out with the patient positioned for a standard posterolateral thoracotomy. Complete exploration of the thoracic cavity is undertaken to visualise and repair diaphragmatic fenestrations. Surgical identification would ideally occur during menstruation because cyclical hormone changes cause pleuro-diaphragmatic endometrial implants to bleed and undergo necrosis.[1] Visible lesions should be excised and the defects repaired. Endoscopic resection of the involved area of hemidiaphragm with an endolinear stapler has been reported, but there is risk of secondary diaphragm rupture.[3] We advocate the systematic placement of an artificial mesh over diaphragmatic fenestrations for all patients, as mechanical pleurodesis, pleural abrasion or pleurectomy alone may not be sufficient to prevent recurrence.

Given the systemic nature of the disease, surgical intervention should, in all cases, be followed by gonadotrophin-releasing hormone (GnRH) analogue therapy to reduce the rate of recurrence.[4] The last patient suffered a postoperative recurrence when hormonal treatment was delayed for 6 weeks, emphasising the importance of starting hormonal therapy swiftly after surgery.

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