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Catamenial pneumothorax and endometriosis-related pneumothorax: clinical features and risk factors

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BACKGROUND: Catamenial pneumothorax and thoracic endometriosis (TE) are still under diagnosed. The purpose of this study is to increase the diagnostic accuracy for these conditions in patients with spontaneous pneumothorax and to identify their risk factors.

METHODS: We conducted a retrospective study on all consecutive women of reproductive age referred to our Centre for surgical treatment of spontaneous pneumothorax between July 2000 and January 2009.

RESULTS: The study population comprised 156 premenopausal women of whom 49 (31.4%) had catamenial and/or TE-related pneumothorax. Over a quarter of these 49 patients had a previous history of recurrent thoracic or scapular catamenial pain. They experienced their first pneumothorax episode at an older age (mean ± SD) (34.0 years ± 6.7) than women with idiopathic pneumothorax (28.7 ± 6.1 years, $P < 0.001$). Pelvic endometriosis was found in 51% of women with catamenial and/or TE-related pneumothorax. After adjustment for confounding factors by multiple logistic regression analysis, the results show that, infertility [odd ratio (OR) = 4.21, 95% confidence interval (CI) = 1.28–13.88] and a history of pelvic surgery with a uterine procedure and/or uterine scraping (OR = 2.85, 95% CI = 1.12–7.26) were the strongest predictors of catamenial and/or TE-related pneumothorax.

CONCLUSIONS: Infertility and uterine procedures are significantly associated with catamenial and/or TE-related pneumothorax. Scapular or thoracic pain during menses often precedes the occurrence of pneumothorax and is highly specific for the diagnosis of TE. Our results suggest that in women with pelvic endometriosis, these symptoms should be systematically investigated for an earlier diagnosis of TE.

Key words: spontaneous pneumothorax / scapular pain / pelvic endometriosis / uterine scraping / infertility

Introduction

Until recently, catamenial pneumothorax (CP) and thoracic endometriosis (TE) have been considered as unusual clinical conditions. Despite an increased interest in the medical community, the conditions remain under diagnosed (Alifano et al., 2006). Some systematic reviews based on small series and case reports collected from the literature have been published on the physiopathology and the management of patients with CP or TE (Joseph and Sahn, 1996; Korom et al., 2004; Channabasavaiah and Joseph, 2010). However, clinical presentations and possible-associated predictive factors have not been described in detail. In particular, the link between CP/TE; reproductive, anthropometric and lifestyle factors; and gynecological aspects have been poorly studied so far.

We present a series of 156 women admitted for surgical treatment of spontaneous pneumothorax. The purpose of the study was to compare the clinical characteristics and possible risk factors of women with catamenial and/or TE-related pneumothorax with those observed in idiopathic pneumothorax. Identification of possible differences could increase the accuracy of diagnosis and its specificity. This may help us to optimize the management of this potentially severe and recurrent disease.

Materials and Methods

Patients

The clinical and pathologic files of all the women of reproductive age referred to our Centre for surgical treatment of spontaneous pneumothorax between July 2000 and January 2009 were retrospectively reviewed. Data on a possible temporal relationship between episodes of pneumothorax and menses, as well as demographic and anthropometric factors were collected. Patients were interviewed by phone to obtain data on lifestyle factors, symptoms, past gynecologic history and long-term outcome. A questionnaire was filled in for every patient. Informed consent was obtained from all women participating in the study, and the research procedures were in accordance with the recommendations of the Helsinki Declaration of 1975 and its successive modifications. Part of this series, focusing on surgical management and pathologic aspects of the disease, has been previously published (Alifano et al., 2007).

Definition used

CP was defined as recurrent episodes of pneumothorax (at least two) occurring between the day before and within 72 h from the onset of menses.

Diagnosis of TE

The surgical treatment consisted of video-assisted thoracoscopy, which allowed an exploration of the whole thoracic cavity. Signs of TE were also carefully searched for. Visceral and parietal pleura were examined to detect nodular brown lesions, whereas the diaphragm was systematically inspected to search for holes and/or endometrial implants. When abnormalities were found (i.e. holes or brown implants), their resection was performed. Pathologic slides of tissues obtained at surgery for pneumothorax were examined by trained pathologists. Pathologic criteria for TE were: the presence of both endometrial glands and stroma, or isolated endometrial stroma staining positively with estrogen/progesterone receptors.

Gynecological investigation

Data on symptoms suggesting pelvic endometriosis were collected in all women. In cases of suspicion of CP and/or TE-related pneumothorax (CP/TE), a clinical gynecological examination followed by pelvic magnetic resonance imaging (MRI) was performed. The MRI criteria for the diagnosis of pelvic endometriosis were as follows. For ovarian endometriosis, diagnosis required the typical features of endometrioma or endometriotic implants with high signal intensity at both T1 and T2-weighted images (or with gradual variation of signal at T2-weighted images as 'shading') persisting at subsequent fat-suppressed T1-weighted images (Togashi et al., 1991). For deep endometriosis, the diagnosis was suggested in case of the presence of any more or less irregular nodule with low or isosignal intensity on T2-weighted images and/or high signal intensity spots in the lesion on T1-weighted images and fat-suppressed T1-weighted images according to the anatomic location of deep endometriosis (Kinkel et al., 1999). No attempt was made to diagnose superficial implants or adhesions. Laparoscopy was performed only if indicated for treatment of pelvic endometriosis.

Statistical analysis

Baseline distributions of certain characteristics, with respect to type of pneumothorax, CP or CP/TE versus non-catamenial and non-TE-related pneumothorax, were evaluated. Results are presented as mean ($\pm$ SD) or as percentages where appropriate. χ^2 tests or, if necessary, the Fisher exact test (for categorical variables) and t-tests (for continuous variables) were done to determine whether these characteristics differed according to the pneumothorax status. We performed logistic regression analyses of data. Age at first pneumothorax, BMI and smoking habits were considered a priori as potential confounding variables and they were included in a multivariate analysis. Odd ratios (ORs) and 95% confidence interval (95% CI) were calculated. Statistical analyses were done using SAS statistical software (version 9.1).

Results

The study population comprised 156 premenopausal women undergoing surgical treatment for spontaneous pneumothorax. Mean age at inclusion was 31.8 $\pm$ 8.7 years. We were able to contact 131 women by phone.

Prevalence of CP and TE in the population

Among the 156 women operated on for pneumothorax, 37 (23.7%) presented with CP. According to the pathologic criteria, the frequency of TE among all patients was 23.1% (n = 36). Among the patients with CP, pathologically proven TE reached 64.9% (n = 24), while 13 patients (8.3%) with CP had no TE. Twelve other patients with TE had non-CP. Combining clinical and pathological criteria, 49 (31.4%) of the patients had CP and/or TE-related pneumothorax (CP/TE); the remaining 107 patients had non-catamenial and non-TE-related pneumothorax (Fig. 1).

A total of 431 pneumothorax episodes occurred in the 156 patients, giving a mean of 2.7 $\pm$ 2.4 episodes (range 1–25) per patient. The mean follow-up was 5.4 $\pm$ 3.8 years after the first occurrence of pneumothorax. Women with CP/TE experienced their first episode at the mean age of 34.0 $\pm$ 6.7 years, which was significantly later than for women without CP/TE who had a mean age of 28.7 $\pm$ 6.1 (P < 0.001). The mean number of episodes before
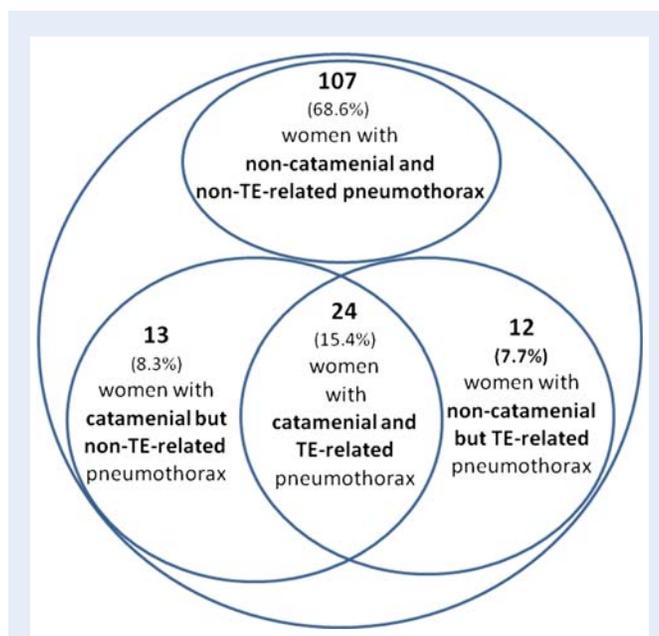

Figure 1 Prevalence of TE and CP among 156 women undergoing surgical treatment of spontaneous pneumothorax.

surgery was not significantly different between patients with CP/TE [2.2 ± 0.9 (range 1−5)], and patients without CP/TE [2.0 ± 0.8 (range 1−5)] ($P = 0.18$). However, the mean total number of episodes (before surgery and during follow-up) was significantly higher in patients with CP/TE [4.0 ± 3.6 (range 1−25)] than in patients without CP/TE [2.2 ± 1.2 (range 1−6)] ($P < 0.001$). Furthermore, among women with TE, the mean interval between the first pneumothorax and the diagnosis of TE was 18.9 ± 27.0 months, with a wide range (0−132 months), and there was a mean number of 3 ± 7.6 (range 1−25) pneumothorax episodes before TE was diagnosed.

Clinical characteristics of pneumothorax

Up to 13 (26.5%) of the 49 patients with CP/TE experienced recurrent thoracic or scapular catamenial pain before the first episode of pneumothorax. This pain usually started several months before the onset of the first pneumothorax, and over a year before for seven patients. One of the patients had suffered from thoracic pain during every menses since her menarche.

All the cases of CP/TE were right-sided except in three patients with bilateral presentation and one with a left-sided pneumothorax only. In contrast, only 58.7% of non-CP and non-TE-related pneumothorax were right-sided, which is compatible with the normal relative contribution of the right lung (55%) to the whole pulmonary mass.

The date of the episode according to the menstrual cycle was available for 336 of the 431 pneumothorax. Their time distribution is presented in Fig. 2. Among patients with non-catamenial and non-TE-related pneumothorax, only 11.5% of the episodes occurred during the menstrual period (maximally once) (Fig. 2D). Among patients with CP/TE, 65.9% of the episodes occurred between Days −1 and +3 from the onset of menses, and 68.7% between Days −3 and +4 from the onset of menses. Furthermore, half occurred on Day 1 of the cycle (Fig. 2A and 1B and C). An additional group of eight pneumothorax episodes occurred during breakthrough bleeding or spotting. Among the women with CP/TE, 29.6% episodes occurred in the intermenstrual period: seven patients with TE, with a total of 17 episodes, did not experience any pneumothorax episode during the menstrual period (Fig. 2C), and 18 women who otherwise experienced recurrent CP also experienced 33 episodes in the intermenstrual period (Fig. 2A and 1B).

The clinical presentation of pneumothorax was partially different between patients with or without TE, although for most it included chest pain (90.5%), shortness of breath (72.9%) and cough (7.4%).
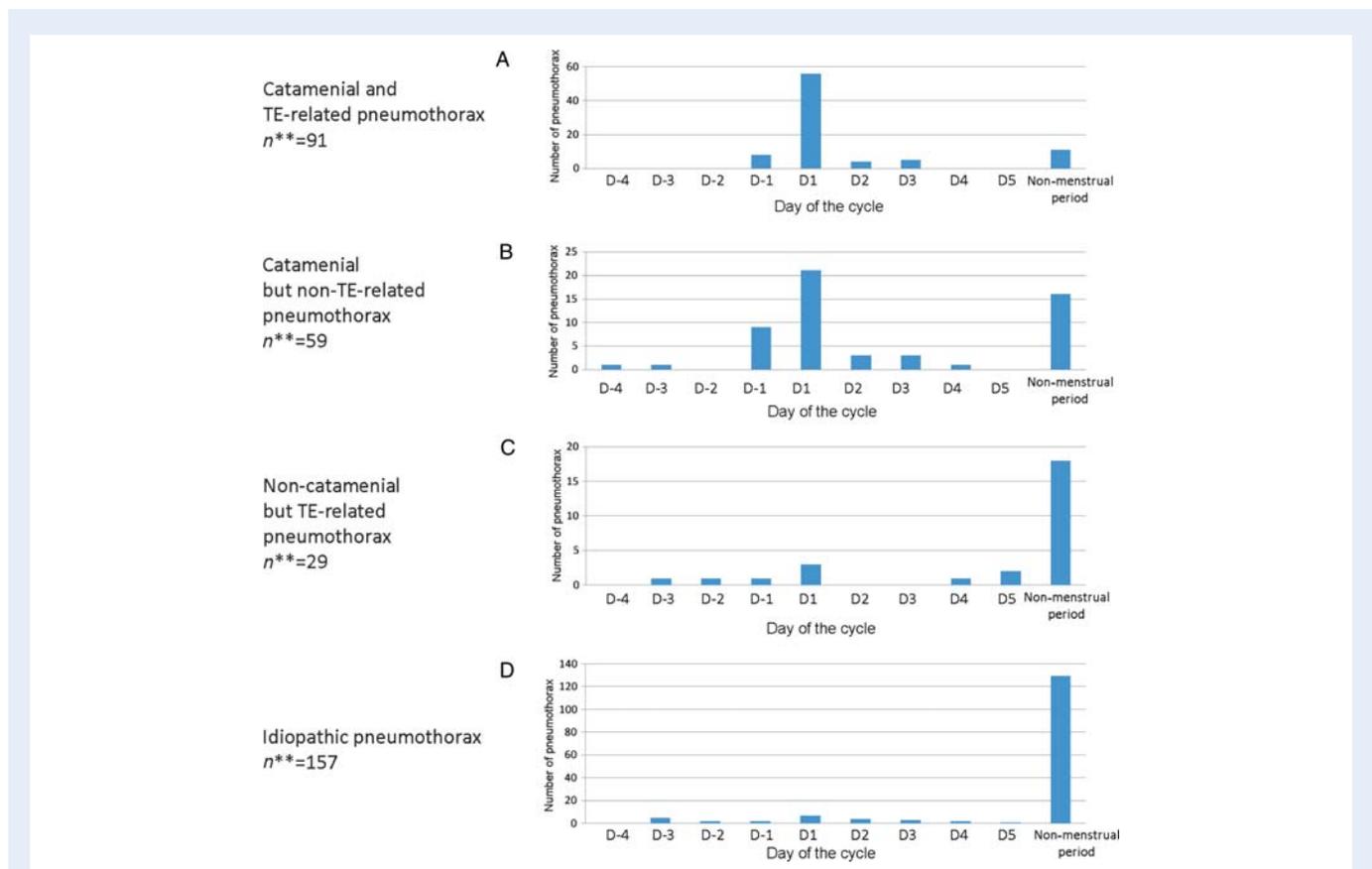

Figure 2 Distribution of pneumothorax episodes in the different groups delineated in Fig. 1 according to the menstrual cycle (D1: Day 1 of the cycle). Pneumothorax in women with catamenial and TE-related penumothorax ($n^* = 24$). Pneumothorax in women with catamenial but non-TE-related pneumothorax ($n^* = 13$). Pneumothorax in women with non-catamenial but TE-related pneumothorax ($n^* = 12$). Pneumothorax in women with idiopathic pneumothorax ($n^* = 107$). CP was defined as recurrent episodes of pneumothorax (at least two) occurring between the day before and within 72 h from the onset of menses. Thus, some patients who experienced only one episode during the menstruation were not categorized as CP, even in case of TE. $^*n$, number of women; $^{**}n$, number of pneumothorax.

Interestingly, basithoracic pain occurred significantly more often in patients with CP/TE (9.15%) than in patients with idiopathic pneumothorax (3.6%) ($P = 0.036$). Only one patient presented with hemoptysis. A triggering factor was found in 26.2% of cases, and was significantly more frequent in non-catamenial, non-TE-related pneumothorax (39%) and in non-catamenial but TE-related pneumothorax (18.8%), compared with 12.7% in CP cases ($P < 0.00001$ and $P = 0.02$ respectively). The triggering factor was heavy load carriage (31.3%) or other physical exercise (12.7%) the day before the pneumothorax episode, a Valsalva maneuver equivalent (4%) or cough (5.9%). A context of stress (mostly professional) was noted in 27.5% of cases, with no difference according to the type of pneumothorax ($P = 0.17$).

Association with pelvic endometriosis

Among the 131 women who were interviewed, the prevalence of pelvic endometriosis symptoms (dysmenorrhea, dyspareunia) was significantly higher in women with TE (58.8%) (20/34) than in women without TE (19.6%) (19/97) ($P < 0.001$), as well as in women with CP (55.9%) (19/34) than without CP (20.6%) (20/97) ($P < 0.005$). Thus, in this population of women with recurrent pneumothorax, the positive predictive value of dysmenorrhea or dyspareunia for the diagnosis of TE was 51.3% (20/39).

Pelvic MRI and/or laparoscopy were performed in 75.5% (37/49) of patients with CP/TE. In these patients, pelvic endometriosis was found in 60.0% (18/30) of women with TE and in 67.6% (25/37) of women with TE/CP. Laparoscopy, which was performed in symptomatic women only, was positive for pelvic endometriosis in 80% of women with TE. There were 22 women with CP/TE who were asymptomatic for pelvic endometriosis but an MRI performed in 13 of them found pelvic endometriosis in 6 (46.1%). Overall, pelvic endometriosis was found in 50% (18/36) of women with TE and in 51% (25/49) of women with CP/TE. Of women with non-TE-related CP, 53.8% (7/13) were shown to have pelvic endometriosis. Different forms of the disease were detected: peritoneal superficial endometriosis (7/25), peritoneal deep endometriosis with posterior cul de sac obliteration and/or uterosacral ligaments infiltration (15/25) [associated with endometrioma (1/15) or adenomyosis (2/15)], isolated endometriomas (2/25) and isolated adenomyosis (1/25).

The sensitivity and specificity of different clinical patterns for the diagnosis of TE are presented in Table I. The occurrence of CP [recurrent episodes of pneumothorax (at least two) occurring between the day before and within 72 h from the onset of menses] were highly specific for the diagnosis of TE. Strikingly, a previous history of recurrent thoracic or scapular catamenial pain was extremely specific for the diagnosis of TE. Symptoms suggesting pelvic endometriosis had lower sensitivity and specificity than CP for the diagnosis of TE. However, the combination of CP with pelvic endometriosis symptoms increased the specificity to >90%, as did the combination of recurrent thoracic or scapular catamenial pain with pelvic endometriosis symptoms (Table I).

Other risk factors

We then investigated whether there was a correlation between different anthropometric, hormonal, lifestyle factors or gynecologic history and the presence or absence of TE and/or CP (Table II).

Table I Sensitivity and specificity of different clinical patterns for the diagnosis of TE.

	Sensitivity (%) (*n/n* total)	Specificity (%) (*n/n* total)
CP	66.7% (24/36)	89.2% (107/120)
Occurrence of at least one pneumothorax within the catamenial interval (from the day before to 72 h from the onset of menses)	80.5% (29/36)	74.2% (89/120)
Recurrent pneumothorax occurring within the 'enlarged' catamenial interval (from 3 days before to 5 days from the onset of menses)	66.7% (24/36)	88.3% (106/120)
History of recurrent thoracic or scapular catamenial pain	32.3% (11/34)	95.9% (93/97)
Symptoms suggesting pelvic endometriosis	58.8% (20/34)	80.4% (78/97)
CP and symptoms suggesting pelvic endometriosis	41.1% (14/34)	92.8% (90/97)
History of recurrent thoracic or scapular catamenial pain and symptoms suggesting pelvic endometriosis	17.6% (6/34)	97.9% (95/97)

BMI and current oral contraceptive use were significantly different between the two groups of women (Table II), but these differences disappeared after age adjustment.

There was a significant higher proportion of current smokers among women with idiopathic pneumothorax.

Half of the women with CP/TE had a history of pelvic surgery and/or pelvic scraping (dilatation and curettage for miscarriage, abortion or abnormal bleeding due to endometrial hypertrophy or uterine polyp, manual removal of placenta and uterine revision after cesarian section, hysteroscopy with endometrial biopsy) compared with only 20.7% of the women with idiopathic pneumothorax ($P < 0.001$).

Infertility was significantly more frequent in women with CP/TE [30.6% (15/49)], than in women with idiopathic pneumothorax [7.8% (7/90)] ($P < 0.001$), and this difference was still significant after age adjustment.

A familial history of pelvic endometriosis was more frequent in women with CP/TE, and a familial history of pneumothorax was more frequent in women with idiopathic pneumothorax (12.8%) than in women with CP/TE (2.1%), although these differences did not reach significance.

Table III shows estimated OR in relation to characteristics of pneumothorax. After adjustment for confounding factors by logistic regression analysis, the results show that, infertility (OR = 4.21, 95% CI = 1.28–13.88), and history of pelvic surgery and/or uterine scraping (OR = 2.85, 95% CI = 1.12–7.26) were the strongest predictors of catamenial and/or TE-related pneumothorax. Interaction between infertility and history of pelvic surgery and/or uterine scraping was not significant. After exclusion of four patients with TE and/or CP who had a previous surgery for co-existing pelvic endometriosis, the factor 'history of pelvic surgery or uterine scraping' remained significant (OR = 2.40, 95% CI = 1.0–6.1).

Table II Characteristics of the women according to the pneumothorax status.

	TE and/or CP (n = 49)	Non-catamenial non-endometriosis-related pneumothorax (n = 107)	P
Mean (SD) age (year) at first pneumothorax	34.0 ± 6.7	28.7 ± 9.1	**< 0.001**
Mean (SD) BMI units	20.4 ± 2.7	19.3 ± 2.3	**< 0.01**
Current-smokers	43.7%	68.9 %	**< 0.01**
Mean (SD) age (year) at menarche	13.1 ± 1.6	13.0 ± 1.5	0.73
Estroprogestin contraception			
Past or current users (%)	85.1	89.0	0.51
Mean (SD) duration (years)	9.9 ± 7.0	8.3 ± 7.0	0.20
Current users at first pneumothorax (%)	48.9	68.9	**0.02**
Intra uterine device (past or current users) (%)	10.9	7.9	0.75
Nulliparity (%)	40.8	56.7	0.07
Infertility (%)	30.6	7.8	**< 0.001**
History of pelvic surgery or uterine scraping (%)	50.0	20.7	**< 0.001**
High socio-professional class (%)	37.2	27.3	0.26
Endometriosis in first-degree relatives (%)	10.6	3.5	0.13
History of pneumothorax in first-degree relatives (%)	2.1	12.8	0.055

Table III OR of main clinical characteristics for CP/TE after adjustment for confounding factors by logistic regression analysis.

	OR (95% CI)	
	Non-adjusted	Adjusted[a]
Age (≥28 years)	4.8 (2.2–10.6)	2.6 (1.0–6.7)
Current smoking	0.4 (0.2–0.8)	0.4 (0.2–0.9)
Infertility	5.2 (1.9–14.0)	4.2 (1.3–13.9)
History of pelvic surgery or uterine scraping	3.8 (1.8–8.2)	2.9 (1.1–7.3)

[a]Adjusted for BMI, age at first pneumothorax, current smoking, infertility and history of pelvic surgery and/or uterine scraping.

Discussion

TE is still an under diagnosed disease: we found a mean interval between the first pneumothorax episode and the diagnosis of TE as high as 19 months in our population of women undergoing surgery for spontaneous pneumothorax. Thanks to the size of this series, we were able to analyze various clinical aspects of CP/TE, some of which are described for the first time. Our main findings were the prevalence of CP/TE among women with spontaneous recurrent pneumothorax, a previous history of thoracic or scapular catamenial pain in the year preceding the first episode of TE-related or CP, the high specificity of CP and symptoms suggesting pelvic endometriosis for the diagnosis of TE, and the link between CP/TE and previous pelvic surgery with uterine procedure and/or uterine scraping.

We confirmed our previous findings of CP/TE prevalence in this larger series (Alifano *et al.*, 2007). Although there has been an improvement in detecting CP/TE, the prevalence remained unclear and underestimated until recently. In the present study, CP accounted for 23.7% of women with spontaneous pneumothorax referred for a surgical treatment. In contrast, Nakamura *et al.* (1986) found that <1% of cases were considered catamenial in a study of a large series of women with spontaneous pneumothorax published in 1986. More recently, two studies found a much higher prevalence of CP (33 and 24.5%, respectively) among women with spontaneous pneumothorax referred for surgery (Alifano *et al.*, 2003; Marshall *et al.*, 2005).

The major aim of our study was to compare clinical presentation and risk factors of CP/TE with idiopathic pneumothorax. An important finding was a previous history of thoracic or scapular pain in the year preceding the first episode of CP. Shoulder pain occurring before the first pneumothorax episode can be due to the irradiation mediated by phrenic nerve of symptomatic diaphragmatic implants, or as a consequence of undiagnosed minimal pneumothoraxic episodes.

The right-sided predominance of catamenial or TE-related pneumothorax in our series is obvious (92% of the cases) as previously described (Joseph and Sahn, 1996; Alifano *et al.*, 2003; Alifano, 2010; Channabasavaiah and Joseph, 2010). This reinforces the theory of the transdiaphragmatic passage of air from the genital tract through diaphragmatic perforations caused by endometrial implants (Crutcher *et al.*, 1967; Kirschner, 1998; Alifano *et al.*, 2006). Diaphragmatic endometrial implants can be explained by the Sampson's theory of retrograde menstruation and by an understanding of peritoneal circulation. This circulation from pelvis and through the right paracolic gutter lead endometrial cells to reach the right sub-diaphragmatic area. Hepatic ligaments represent barriers for further spreading and are responsible for a distribution in favor of the right leaf (Vercellini *et al.*, 2007). Cells would then implant in the diaphragm. These implants undergo cyclical necrosis, and subsequent cycles lead

to holes perforating the diaphragm that eventually allow a transdiaphragmatic passage of air and further spreading inside the chest (Alifano et al., 2006). The catamenial character of the pneumothorax is explained by the liquefaction of the cervical mucus plug during the menstrual period, which allows the retrograde movement of air (Maurer et al., 1958). In favor of the transdiaphragmatic passage of air is also the occurrence of concomitant pneumoperitoneum and CP (Jablonski et al., 2009).

We worked closely with the Thoracic Surgery department to investigate the association between TE and pelvic endometriosis. Pelvic endometriosis was confirmed in 60% of the patients with TE who underwent pelvic MRI and/or laparoscopy, and in 80% of those when a laparoscopy was performed. The high proportion of pelvic endometriosis found among asymptomatic (from a gynecologic point of view) women with CP/TE underlines the importance of a systematic gynecologic work-up for these women. Furthermore, almost all women with CP and no TE who were investigated by laparoscopy and/or MRI actually had pelvic endometriosis, suggesting that endometriosis may be associated with CP even in the absence of diagnosed TE. The diagnostic of TE relies on the histological evidence of endometriotic tissue, which can be lacking due to paucity of material collected from diaphragmatic resection or absence of diaphragmatic resection. This further argues for a systematic gynecological consultation for these women. We also found a more frequent (but not reaching significance) familial history of endometriosis among women with CP/TE in accordance with the well-known familial aggregation of endometriosis (Matalliotakis et al., 2008a). These findings are in agreement with the proportion of women with pelvic endometriosis (84%) reported in the collective review by Joseph and Sahn (1996). Although Channabasavaiah et al. found no significant association between TE and pelvic endometriosis in general [the study compiled data from women with pneumothorax in 79 (72%) patients, hemoptysis in 16 (14%), hemothorax in 13 (12%) and lung mass in 2 (2%)], he found a similar proportion of documented pelvic endometriosis (65–70%) among women presenting with pneumothorax ($n = 33$) (Channabasavaiah and Joseph, 2010).

Thus, we think that women presenting with either spontaneous pneumothorax (especially if right-sided) or pelvic endometriosis should systematically undergo a careful medical interview. This could lead to a faster diagnosis of TE. The presence of pelvic endometriosis, when associated with recurrent catamenial thoracic pain and possibly pneumothorax, should suggest TE. In our series, 26% of patients with CP/TE had a history of recurrent thoracic catamenial pain before the first pneumothorax episode. Furthermore, in this population of women with recurrent pneumothorax, the positive predictive value of dysmenorrhea or dyspareunia for the diagnosis of TE was 51.3%. Women with pelvic endometriosis should be systematically asked about pleuritic, shoulder or upper abdominal pain occurring with menses. Identification of these women at a higher risk for TE and CP could contribute to an early diagnosis of TE. Furthermore, if these women undergo a laparoscopy for treatment of pelvic endometriosis, a careful visual inspection of the diaphragm should be performed routinely (Nezhat et al., 1998; Nisolle et al., 2007). Laparoscopy may identify small 'sentinel' lesions of endometriosis on the anterior and mid-diaphragm. However, the most significant lesions are located on the posterior diaphragm, and cannot be seen easily with a laparoscope from an umbilical port. Thus, in case of sentinel

lesion on the anterior diaphragm or in symptomatic patients, exploration of the posterior diaphragm could be recommended (Redwine, 2002; Nisolle et al., 2007). The early diagnosis of diaphragmatic lesions could lead to a better management of these women, despite the lack of a validated preventive method for the first pneumothorax occurrence. The occurrence of one pneumothorax in these women could be an indication for an appropriate thoracic surgical treatment, regarding the high risk of recurrence.

We found the history of pelvic surgery with uterine procedure or uterine scraping as strong predictors for CP/TE. Such conditions could also be taken into account during the initial consultation for pneumothorax. Trauma or manipulation of uterine tissue has been suggested to be a predisposing factor to microembolization of endometriotic cells and thus associated with a higher frequency of pulmonary localizations (Joseph and Sahn, 1996; Hope-Gill and Prathibha, 2003). However, we did not find such an association with endometriosis pulmonary nodules.

The higher proportion of infertile women among patients with CP/TE might be explained by the frequent association between TE and pelvic endometriosis.

Our study confirms, with an increased power, other clinical aspects previously described in the literature. As expected, pneumothorax triggering factors were much more frequent in non-CP, non-TE-related pneumothorax. However, among women who experienced a TE-related pneumothorax episode in the intermenstrual period, a triggering factor was also found in 18.8% of cases, which confirms a previous observation that a triggering factor could be involved in this kind of pneumothorax (Alifano et al., 2004). The mechanism of endometriosis-related pneumothorax in the intermenstrual period is unclear. Air might be forced from the outside to the peritoneum through physical activity or sexual intercourse (Muller and Nelems, 1986; Kirschner, 1998) and then enter the thorax through diaphragmatic defects with a subsequent pneumothorax.

The later occurrence of the first pneumothorax episode in women with TE than in women with idiopathic pneumothorax is in accordance with data previously published by Joseph and Sahn and by Channabasavaiah and Joseph, where the mean age at presentation was 35 years and 36.2 years, respectively (Joseph and Sahn, 1996; Channabasavaiah and Joseph, 2010).

We also looked at the accuracy of the classical definition of CP. The most widely accepted definition for CP is a recurrent spontaneous pneumothorax occurring within 24 h before, to 72 h after the onset of menses. Other time frames (with respect to the onset of menses) have been proposed (Alifano et al., 2006). The sensitivity of CP for the diagnosis of TE in our series is low. Nevertheless, the occurrence of at least one pneumothorax episode during the catamenial interval has a better sensitivity (80.5%), but a weaker specificity (74.2%). The enlargement of the 'catamenial window' (to 3 days before and within 5 days from the onset of menses) did not improve the sensitivity for diagnosis of TE. It would appear then that the usual definition of CP is satisfactory. Smoking is a well-known risk factor for idiopathic spontaneous pneumothorax (Bense et al., 1987; Cheng et al., 2009; Noppen, 2010). In our series, the proportion of smokers was significantly lower in women with CP/TE than in women with idiopathic spontaneous pneumothorax. Among women with CP/TE, 43.7% (95% CI = 29.7–57.7) were current smokers, which is not significantly different from the proportion of

smokers in French women of the same age in the general population (31.5–33.5%) (Ministère Français de la Santé et des Sports, 2010).

After age adjustment, mean BMI was not significantly different between women with CP/TE and women with idiopathic spontaneous pneumothorax (who are known to have a lean phenotype) (Noppen, 2010). However, the mean BMI of women with CP/TE was slightly lower than the mean BMI of French women of the same age (De Saint Pol, 2006). Women with pelvic endometriosis have also been described as having a lower BMI than control groups (Signorello *et al.*, 1997; Ferrero *et al.*, 2005; Hediger *et al.*, 2005; Matalliotakis *et al.*, 2008b).

There is no consensus on a possible relationship between the use of cyclic oral contraceptives and pelvic endometriosis (Vessey *et al.*, 1993; Sangi-Haghpeykar and Poindexter, 1995; Parazzini *et al.*, 1999). We did not find a relationship between the ever-use of oral contraception and the presence of CP/TE, although there was a higher proportion of current users in women with idiopathic spontaneous pneumothorax (not reaching significance after age adjustment).

Conclusion

Despite an increased awareness in the medical community and improved detection, CP and TE are still under diagnosed. Scapular or thoracic pain during menses, infertility and a history of pelvic surgery with uterine procedure or uterine scraping have to be looked at in women presenting with pneumothorax in order to evoke the diagnosis of CP/TE. Although there is no specific preventive management before a first CP in the context of pelvic endometriosis and systematic laparoscopy is not recommended, women presenting thoracic pain during menses should be advised to consult in case of abnormal or unusual thoracic pain and dyspnea. Better knowledge about this disease among specialists in Emergency Medicine, Pneumology, Gynecology and Internal Medicine could help in reducing the delay of diagnosis so that these young women can receive timely treatment to prevent recurrence.

Authors' roles

C.R.J. contributed to the conception of the study, acquired and analyzed the data and drafted the paper; M.A. contributed to conception and design of the study, to acquisition of data (performed surgical procedures) and to the draft and revision of the paper; G.P.B. performed the statistical analysis and participated in revision of the paper; S.C.B. contributed to acquisition of the data (reviewed all pathologic slides); P.R. contributed to acquisition of the data (performed pelvic MRI) and participated in the writing; J.F.R. contributed to conception and design of the study and performed surgical procedures; A.G. was in charge of the study, acquired the data (clinical examination and interviews) and contributed to drafting and revising the paper.

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