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## Management of thoracic endometriosis: single institution experience

Inmaculada Duyos\*, Ana López-Carrasco, Alicia Hernández, Ignacio Zapardiel, Javier de Santiago

Gynecology Department, La Paz University Hospital, Madrid, Spain

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### ABSTRACT

**Objective:** Thoracic endometriosis is a relatively rare type of endometriosis and includes catamenial pneumothorax, hemothorax and hemoptysis, and presence of intrathoracic endometriotic nodules. We want to clarify and resume the most appropriate management of this pathology.

**Study design:** We retrospectively reviewed all the cases of thoracic endometriosis diagnosed and followed up in our Unit from 2005 to 2013. This search revealed five women, with a mean age of 34.5 (26–44). Four had previous history of endometriosis or severe dysmenorrhea. One patient complained of chronic shoulder pain with diaphragmatic implants, another one presented catamenial hemoptysis, and three women suffered from catamenial pneumothorax, with right-side preference.

**Results:** Three patients underwent surgery and all of them were treated with GnRH agonists at least during 6 month referring improvement in symptoms. After 1–8 years follow-up, all these patients remain asymptomatic.

**Conclusion:** The optimal management of thoracic endometriosis needs further evaluation but the combined approach by hormonal therapy and surgery could be the best option.

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## 1. Introduction

Endometriosis is a disorder characterized by the growth of endometrial tissue outside the uterine cavity. Usually the ectopic endometrial foci are located in the pelvis, but extra-pelvic disease can be rarely found. Endometriosis is thought to affect 5–15% of women in reproductive age and the incidence of extra-pelvic endometriosis is approximately 12% in these women [1,2].

Thoracic endometriosis is the most common extra-pelvic manifestation [3] and includes catamenial pneumothorax, hemothorax and hemoptysis, as well as the presence of intrathoracic endometriotic nodules [4]. The treatment includes hormonal therapy and surgery.

We report a case series of five women with thoracic endometriosis followed up at Endometriosis Unit at our hospital.

*Abbreviations:* MRI, magnetic resonance imaging; VATS, video assisted thoracoscopic surgery; GnRH, gonadotropin-releasing hormone.

\* Corresponding author at: Obstetrics and Gynecology Department, University Hospital, “La Paz” Paseo de la Castellana 261, 28046 Madrid, Spain. Tel.: +34 917277008; fax: +34 917277207.

E-mail address: [macuduyos@yahoo.es](mailto:macuduyos@yahoo.es) (I. Duyos).

## 2. Materials and methods

After Institutional Review Boards approval, we retrospectively reviewed all cases of thoracic endometriosis diagnosed and followed up at La Paz University Hospital from 2005 to 2013. Thoracic endometriosis was defined as an extrapelvic manifestation that includes four clinical entities: catamenial pneumothorax, catamenial hemothorax, catamenial hemoptysis and lung nodules suggestive of endometriosis [4]. We searched in our Unit of Endometriosis database and the inclusion criteria were: operations for recurrent spontaneous pneumothorax, hemothorax or chronic shoulder pain in the absence of an underlying lung disease, and presence of characteristic intraoperative findings or pathologic diagnose of endometriosis. Three of the patients were referred to our Unit for evaluation from Thoracic Surgery Department presenting pneumothorax and two of them from our own Gynecology Department. A Medline search was carried out in order to clarify and resume the most appropriate management in those cases. Follow-up was made by clinic visit in all cases, every 3 months during the first year and then twice a year. All patients were questioned about symptoms improvement with treatment. Gynecologic examination, abdominal and vaginal ultrasound were performed in every visit. Patients presenting

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pneumothorax or hemoptysis were questioned about new episodes, and chest radiography, TC or MRI scans were requested if there was worsening of the symptoms. Annual bone densitometry was performed in women receiving GnRH-agonist therapy.

### 3. Results

We found five cases of thoracic endometriosis during the study period. Patients' characteristics are showed in Table 1. The detailed clinical cases are presented next:

#### 3.1. Case 1

A 26-year-old woman, nulliparous, with a history of chronic pelvic pain since age 20 presented at our unit due to 3 episodes of catamenial hemoptysis occurring during last year. The patient had previous history of endometriosis and had undergone laparoscopic cystectomy three years before. She was taking oral contraceptive pills without pain relief. Sonographic scan revealed a 40-mm left ovarian cyst and thoracic MRI showed two 1-cm nodules within the right lung, suggestive of pulmonary endometriosis, based on clinical grounds, laterality and hyperintense appearance on T1-weighted MRI images. The patient underwent laparoscopic cystectomy and during the intervention, extensive endometriosis involving the ovaries, utero-sacral ligaments, serosa of recto-sigmoid colon, recto-vaginal septum and both ureters was noted. Exploration of the upper abdominal cavity revealed a 1-cm endometriotic lesion on the surface of the right hemidiaphragm (Fig. 1). During surgery the cyst, recto-vaginal septum and the endometriotic lesion over the diaphragm were removed. Diaphragmatic lesion was superficial and was excised by peritonectomy, without accessing the thoracic cavity. Uterus and ovaries could be preserved. The pathologic report confirmed endometriosis in every specimen removed. After surgery the patient received add-back therapy during 1 year with Triptorelin and Tibolone to control the symptoms and the adverse effects of analogs. In this period the patient did not refer more episodes of catamenial hemoptysis and symptoms during menses decreased. Then she has undergone one cycle of in vitro fertilization with success.

#### 3.2. Case 2

A nulliparous 30-year-old woman, with a history of chronic pelvic pain since she was 25 and severe dyspareunia, dysuria, dyschezia and dysmenorrhea during the last 2 years, treated with hormonal contraceptive. Moreover, she presented a chronic right shoulder pain, which was exacerbated during menstruation. She had a history of extensive endometriosis and two previous laparoscopies: ovarian cystectomy and appendectomy plus fulguration of diaphragmatic lesions with CO2 laser the second time. After the surgery she was treated with GnRH agonists during six months getting improvement just of the right shoulder pain. The upper abdominal MRI revealed 2-cm, 1.4-cm and 0.6-cm hyperintense cystic lesions, which based on clinical symptoms, were suggestive of endometriotic nodules, on the abdominal surface of the right hemidiaphragm, near the VII segment of the



Fig. 1. Right diaphragmatic endometriosis nodule.

liver, and pelvic examination showed a 3 cm left ovarian cyst and endometriosis involving the uterus, utero-sacral ligaments and recto-vaginal septum. In order to achieve pregnancy she underwent laparoscopic left cystectomy and salpingectomy as well as removal of diaphragmatic implants. These were infiltrative, so surgical full-thickness resection and laparoscopic suture of the diaphragm were performed. Endometriotic tissue was confirmed in all lesions. After two in vitro fertilization cycles without success, and two years of follow-up she continued with severe dyschezia and dysmenorrhea. Finally she underwent hysterectomy and bilateral oophorectomy. After the last surgery she is receiving hormonal replacement treatment. Symptoms have decreased, and the patient reports great improvement.

#### 3.3. Case 3

A secondiparous 44-year-old woman was referred from thoracic surgery department to our unit. She had no previous history of endometriosis and never had suffered from dysmenorrhea, but the last two years she had presented five episodes of right pneumothorax related to menstruation. The first four episodes were managed with chest tubes, and the last one with bullectomy and chemical pleurodeses. Pathologic report revealed pleural endometriosis. A pelvic MRI showed a 14 mm endometriotic cyst on the right ovary. Since the patient rejected surgical treatment she is receiving GnRH-agonists and Tibolone. The patient did not report more pneumothorax or chest pain episodes after one year follow up.

#### 3.4. Case 4

A 42-year-old woman, gravida-2, para-2 presented four right spontaneous recurrent pneumothorax during menstruation in 1-year period. Two of them were treated by port video-assisted thoracoscopic surgery (VATS) performing bullectomy and pleurodeses. The patient did not have known history of endometriosis but dysmenorrhea occasionally. Pelvic examination and sonographic scan were normal. She received "add-back" therapy with GnRH-agonist and Tibolone during nine months. She tried to leave the treatment but she had two more recurrences, so she underwent

Table 1

Summary of patient's clinical characteristics.

Case	Age (years)	Parity	Main symptoms	Side	Diagnosis	Pelvic endometriosis	GnRH agonist
#1	26	0	Hemoptysis	Right	Pathologic confirmation	Yes	Yes
#2	30	0	Chronic shoulder pain	Right	Pathologic confirmation	Yes	No
#3	44	2	Recurrent pneumothorax	Right	Pathologic confirmation	Yes	Yes
#4	42	2	Recurrent pneumothorax	Right	VATS confirmation	No	Yes
#5	35	0	Recurrent pneumothorax	Right	VATS confirmation	Yes	Yes

VATS, video assisted thoracoscopic surgery; GnRH, GnRH agonists treatment.

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