

Thoracic Endometriosis Syndrome–Uncommon Entity of Pelvic Endometriosis

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Submitted: 21 July 2017; **Accepted:** 08 Aug 2017; **Published:** 05 Sep 2017**Abstract**

Thoracic Endometriosis syndrome (TES) is a complex condition consisting of four distinct clinical entities: catamenial pneumothorax, catamenial hemothorax, hemoptysis, and pulmonary nodules. TES poses a clinical dilemma when presented with chest symptoms rather than predictable symptoms of pelvic endometriosis. It is a complex condition often diagnosed late. The treatment includes hormonal management and surgical treatment if needed.

We report the case of an 18-year-old girl who was initially diagnosed with recurrent spontaneous pneumothorax and subsequently proved to have catamenial pneumothorax secondary to pelvic endometriosis.

Introduction

The presence of endometrial tissue outside the uterine cavity in pelvic structures like ovary, uterine ligaments, pelvic peritoneum, cervix and vagina is called pelvic endometriosis. It can affect the bowels in abdomen and pelvis either on surface (superficial) or penetrate into bowel walls (deep infiltrative endometriosis). Rarely endometriosis can present outside abdominal peritoneal cavity in or around lungs referred to as Thoracic endometriosis. Thoracic endometriosis is a rare entity.

Case Report

An 18 year old girl was seen by cardiothoracic surgeon with the view to treat her recurrent right sided spontaneous pneumothorax, which she had twice within a year. She underwent VATS bullectomy and pleurectomy with operative findings of bloody pleural effusion, not normally associated with primary spontaneous pneumothorax. She was also noted to have obvious diaphragmatic fenestrations with haemosiderin deposits around them. Histological report of pleural lesion showed aggregates of CD10 positive stromal cells supporting diagnosis of endometriosis and concluded with identification of features consistent with a diagnosis of catamenial pneumothorax.

On gynaecological review she revealed presence of pelvic pain and previous treatment for menorrhagia. A transvaginal scan confirmed bulky heterogeneous adenomyotic uterus and the presence of likely left ovarian endometrioma measuring 42x37x25mm. She was started on conservative management with COCP. The mapping MRI ruled out involvement of bowels and confirmed presence of endometriotic cyst. Her pelvic pain did not improve reasonably well. She had another recurrence of pneumothorax and had repeat VATS with mechanical open pleurodesis and talc pleurodesis. Her

repeat MRI revealed resolution of endometrioma but significant adhesions between anterior rectum and posterior surface of uterus. She underwent laparoscopy which revealed a frozen pelvis with significant adhesions in pelvis and none of the ovaries could be visualised. The dye test was negative on either side. A reasonable adhesiolysis was done including bowel adhesiolysis but intraoperative decision of performing further adhesiolysis and extirpation of endometriosis after GnRH analogue was taken in view of level of difficulty and bleeding. Her chest symptoms have improved and she remains on GnRH analogues before her further surgical treatment of endometriosis.

Discussion

Endometrial tissue from pelvis migrates to right side hemidiaphragm following the physiological movement of peritoneal fluid in clock-wise direction from the pelvis along the right paracolic gutter to the subphrenic space. The endometrial tissue migrates into thorax through diaphragmatic fenestrations which may be either congenital or the result of direct erosion by endometrial implants and could be as large as four inches in diameter [1,2]. TES is almost exclusively seen on right hemithorax following the same directional flow of peritoneal fluid, landing more commonly on the right hemidiaphragm [3,4].

Catamenial pneumothorax which is seen in two-thirds of TES is believed to occur within 72 hours from the onset of menstruation [5]. It is believed that trans-diaphragmatic movement of air happens through the fenestrations and air travels from vagina through fallopian tubes and subsequently into thorax, when cervical mucus plug is absent at the time of menstrual periods [2,6]. Catmenial haemothorax, hemoptysis and pulmonary nodules are very rare entities of TES.

The symptoms of pleuritic chest pain and dyspnea with or without cough or hemoptysis at the time of menstrual periods should raise the suspicion of TES particularly in a woman with known pelvic pain or confirmed endometriosis. MRI of pelvis and chest is a useful investigation in mapping not only pelvic endometriosis but also differentiating pleural from parenchymal endometrial implants. TES requires multidisciplinary approach in its management by gynaecologists and thoracic surgeons [7].

The principle of treating TES is ovarian estrogen suppression with oral contraceptives, progesterone agents, danazol, or gonadotropin-releasing hormone (GnRH) agonists. The surgical treatment approach of thoracic endometriosis is excision of any endometrial implants, closure of diaphragmatic fenestrations and mechanical or chemical pleurodesis with talc or tetracycline [8]. Surgical excision or extirpation of pelvic or bowel endometriosis may also be required.

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