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Medical management of bronchial endometriosis: Case report and literature review

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Abstract
We are presenting 29 year old female complaining of cataminal haemoptysis 3 months post-delivery of her second child. She had no previous history of endometriosis or chronic pelvic pain. She was investigated thoroughly by respiratory and immunology teams for all possible diagnosis and clinically the final diagnosis was bronchial endometriosis. This case showed that combined oral contraceptive pills has successful role of management of lung endometriosis.

Keywords: bronchial endometriosis, post-delivery, respiratory, immunology

Introduction

Endometriosis is the presence of functional endometrial-like tissue outside of the uterus It is most commonly in the ovaries, uterosacral ligaments, and pelvic peritoneum the condition was first reported in 1860 [1]. Endometriosis is reported in 5% to 15% of females worldwide during their reproductive life [2]. In some rare cases ectopic endometrial tissue has been found in distant sites, such as the umbilicus, abdominal scars, breasts, the extremities, pleural cavity and lung parenchyma [3, 4]. The symptoms of extra-pelvic endometriosis are not always synchronous with the menstrual cycle, and diagnosis can be particularly difficult.

Bronchial endometriosis is an uncommon condition, and usually associated with cyclical haemoptysis and chest pain. Diagnosis mainly depends on clinical suspicion based on presenting symptoms and history, with confirmation by histopathological assessment [5]. Clinical examination often reveals only occult signs and symptoms. Reported cases that had cyclical pain and haemoptysis often have severe decidual adhesions and distortion of tissue around the decidua [6]. Additionally, the pathologist's role is made difficult by the atypical histopathological features, from typical endometrial glands to an abundance of fibrous tissue, on a background of lunch parenchyma and bronchial tissue.

The major modality of treatment for bronchial endometriosis is surgical excision through bronchoscopy, which usually provides a positive outcome [7]. Medical management such as use of contraceptive progestogens, gonadotropin-releasing hormone agonists, androgens, and non-steroidal anti-inflammatory drugs have been used with mixed effect, and often only have benefits for limited time periods, and the long term side effects of chronic use [8].

Case Report

A 27 year old female, Para 2+0 presented to a tertiary university hospital complaining of several episodes of cyclical cataminal haemoptysis (coughing blood during menstruation). The patient had a prior uncomplicated spontaneous normal vaginal delivery of her second child 3 months previously, with a routine postnatal course. Her menses resumed eight weeks post-delivery. Despite having no pre-existing respiratory problems, she developed severe new onset pulmonary bleeding post-partum, coinciding with menstruation. Each episode of haemoptysis was estimated at 15ml volume with each period, and she presented to acutely to the hospital emergency department and required inpatient admission each time. The patient never reported any cyclical pelvic pain, or abnormal gynaecological symptoms. She was initially assessed by the respiratory physicians, as each episode accompanied with significant dyspnoea. On physical examination, right-sided coarse crepitations with decreased air entry was noted on auscultation. PA Chest radiography demonstrated an opacity and regular markings in the right lower lung lobe. All haematological and biochemical investigations, including full blood count, c-reactive protein, liver function tests and renal function tests, were normal. To further assess, a CT pulmonary angiogram (CTPA) was arranged. This identified a focal area of "ground glass" changes within the superior segment of the right lower lobe, and was deemed to likely be either infectious or inflammatory by the reporting radiologist. No pulmonary embolus, plural effusion, thoracic lymphadenopathy or osseous abnormality were seen. She was started on broad spectrum antibiotics (pending microbiological analysis of sputum culture) with the recommendation to repeat the CT thorax in 4-6 weeks, and discharged home for outpatient follow up with the report.

A month after the initial presentation she re-attended the emergency room with heavy respiratory bleeding and shortness of breath, again with menstruation. A repeat CTPA was performed, and interestingly this showed an increase in the previously identified "ground-glass" changes with new extension into the anterior aspect of the apical segment of the right lower lobe. No evidence of pulmonary haemorrhage or nodules were seen. During this repeat CTPA the patient had active haemoptysis. Following review and discussion at the regional respiratory multi-disciplinary meeting a bronchoscopy was recommend, and subsequently completed during this emergency admission. Bronchoalviolar lavage (BAL) (it is a diagnostic method of the lower respiratory system in which a bronchoscope is passed through the mouth or nose into an appropriate airway in the lungs, with a measured amount of fluid introduced and then collected for examination) was performed, and histology revealed an abundant alveolar presence.

Eosinophils were not identified on May Grünewald-Giemsa (MGG) Stain (MGG is stain used for staining of blood, bone marrow smears and clinical cytological specimens), and no malignant cells were seen. A full immunology screen was then completed including: Anti-neutrophil cytoplasm antibodies (ANCA) and DNA by crithidia luciliae (it is a flagellate parasite that uses the housefly, *Musca domestica*, as a host) [20]. As part of the family of Trypanosomatidae, it is characterised by the presence of a kinetoplast, a complex network of interlocking circular double-stranded DNA (ds DNA) molecules, which were negative. Complement C3 and C4 levels were within normal range.

The respiratory team at this stage noted that the haemoptysis was cataminal – as it occurred recurrently during the days when the patient was actively menstruating. A gynaecology consultation was then sought. She had no previous gynaecological history of note. A recommendation to commence the patient on a course of triptorelin pamoate (Decapeptyl) 11.25mg injection for 3 months to achieve ovarian and menstrual suppression was made by the attending consultant. Due to the possibility of hypoestrogenic vasomotor symptoms, and potential osteopenia if longer term use was needed, the ability to incorporate add-back hormone replacement therapy (HRT) for symptom control and bone-protection was discussed. An ultrasound pelvis was performed to assess for structural abnormalities, but was unremarkable. A provisional working diagnosis of pulmonary endometriosis was now made, and an MRI Thorax was arranged to further investigate. Unusually this showed no T1 hyper-intensities within the lung parenchyma or plural or diaphragm to suggest endometriosis deposits (figure 3, 4).

No episodes of cataminal haemoptysis occurred in the 3 months followed the triptorelin pamoate injection. A subsequent CT thorax showed complete resolution of “ground-glass” opacification, with appearance suspicious for prior pulmonary haemorrhage in the anterior basal lower lobe, but with no acute findings suspicious for current pulmonary haemorrhage (figure 1, 2). She had minimal hormonal side effects from the ovarian suppression, with only mild but tolerable episodes of warm flushing and minor mood change.

For long term management a patient centred discussion was had outlining risks and benefits of the treatment options. With no personal contraindications for combined oral contraceptive pill use, the patient was commenced on Yasmin (0.03mg Ethinylestradiol/3mg Drospirenone) for three months continuously, avoiding the pill-free weeks. She again suffered no episodes of cataminal haemoptysis throughout, however did reported ongoing light headaches. She was switched Microlite (100mcg Levonorgestrel/20mcg Ethinylestradiol) for 6 months, again “back-to-back”, with no break. She reported no adverse side effects for three months, but at this point had a small break-through bleed, together with some mild chest pain, but no haemoptysis. Since then for the last she has been maintained on the Levonorgestrel/Ethinylestradiol combination, taking pill free period every six months for one week, without complaints or further episodes.

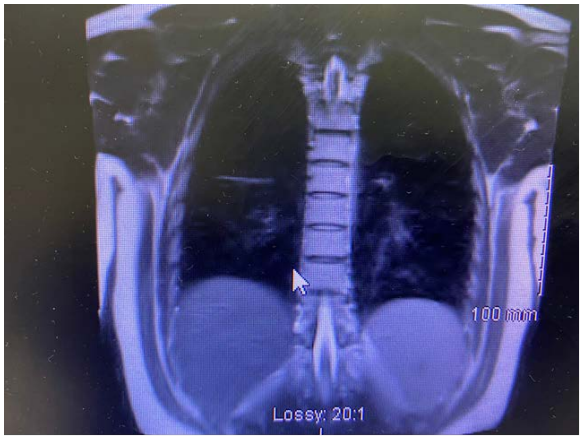

Fig 1

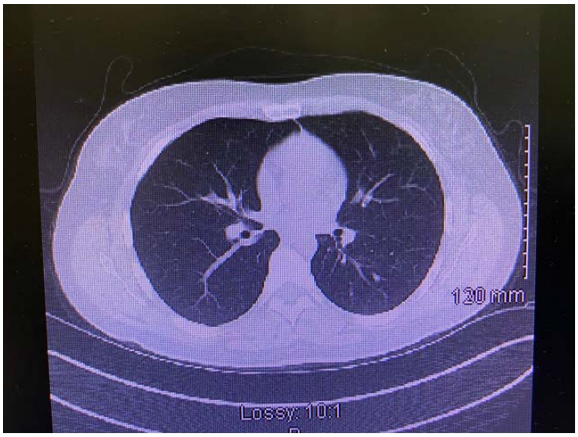

Fig 2

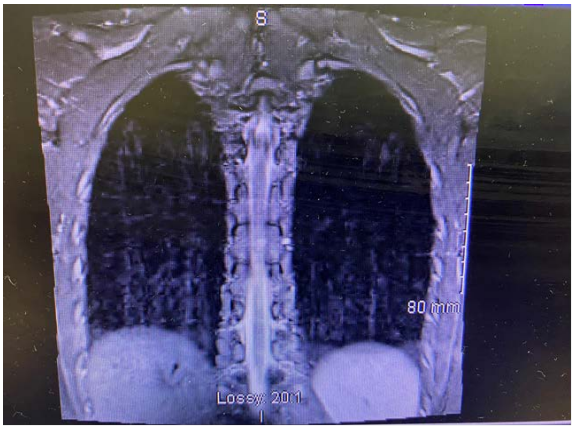

Fig 3

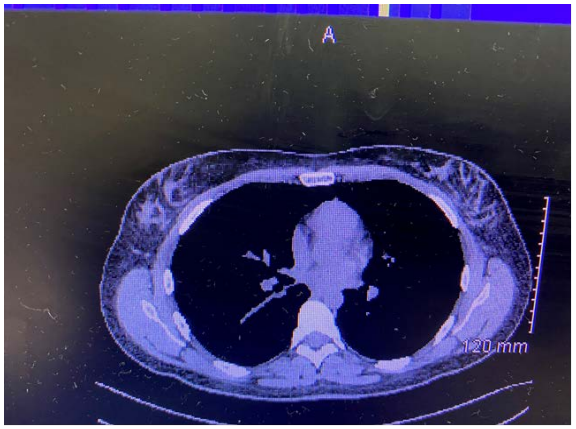

Fig 4

Discussion

Bronchial endometriosis is a rare condition of non-pelvic endometriosis [9]. Approximately 60% of cases also have true co-existing pelvic endometriosis [9, 10, 11]. Simultaneous extra-pelvis endometriosis of the diaphragm and the visceral pleura are found in 38.8% and 29.6% of cases respectively [10]. Endometriosis of the lung parenchyma is much less common [9, 11]. The pathophysiology of cyclical haemoptysis remains unclear, though three existing predominant theories dominate the literature [12]. The leading hypothesis, proposed by Schron and Ruysh [13], is a variant of Sampson's theory of retrograde menstruation. This model suggests that endometrial-like cells enter the peritoneal cavity then pass to pleural space by lymphatic channels. An alternative second hypothesis considers that high levels of prostaglandin F2 at the time of ovulation may result in vasospasm and associated ischemia in the lung parenchyma. A combination of prostaglandin-induced bronchospasm, may cause the alveoli to rupture resulting in haemoptysis, and also explain the associated risk of pneumothorax with thoracic endometriosis [13]. Thirdly, an anatomical theory is that the loss of the cervical mucus plug during menses results in communication between the environment, peritoneal cavity, and subsequently the pleural space [14]. As many women will have retrograde menstruation with their monthly periods, yet thoracic endometriosis is rare, none of these proposed models can completely explain this phenomenon. More recent theories in molecular and cellular pathophysiology have looked at a spread of endometrial micro RNAs through exosomal trafficking [15], though this is a novel theory for extra-pelvic endometriosis spread and as of yet has not been demonstrated in bronchial endometriosis.

Diagnosis of bronchial endometriosis is almost always grounded initially on clinical suspicion [11]. The majority of patients present to hospital with a combination of cataminal haemoptysis, shortness of breath, cough, and pleurisy. Investigations including chest radiography, CT thorax, MRI thorax and bronchoscopy can also give help to support the clinical suspicion for a diagnosis of bronchial endometriosis. However, in the majority of cases reported, diagnosis is usually made, and treatment performed, with the use of video-assisted thoracoscopic surgery (VATS) [14]. This allows direct visualisation of any pathology, and targeted biopsy of any suspected endometriotic lesion [14]. The initial management is often symptom control through medical treatment; which brings side-effect profiles, recurrence risks and long-term high cost to the patients without definite treatment of the lesion. Surgical interventions such as chemical pleurodesis, pleurectomy, resection and lobectomy have proven successful management options [14].

While in this case no evidence of pelvic endometriosis or gynaecological disease was found on radiological images; the CT findings for pulmonary endometriosis may include well-defined opacities, nodular lesions, thin-wall cavities, or bullous formations, but in cases with haemoptysis they have transient radiologic densities in the part of the lung [16]. In this case, the CT revealed an opacity at the anterior part of the lower right lobe, histopathology of bronchial lavage samples did not indicate ectopic endometrial pathology. Generally, diagnostic requirements for bronchial endometriosis is the presentation of periodic haemoptysis that is synchronous with menstruation. Most previously reported cases were diagnosed based on the clinical history of the patient; and a histological confirmation of ectopic endometriosis is not always completed and reported. Unfortunately, though bronchoscopic attempts at tissue sampling were made, we were unable to get positive histological samples. However, the presence of cataminal haemoptysis, synchronous to menses combined with a response to ovulation suppression confirmed the diagnosis.

Recently, VATS for surgical resection to treat cyclical haemoptysis was reported to be safer and less invasive than lobectomy [17, 18, 19]. Most of the time management of cases of endometriosis treated with hormonal therapy, typically gonadotrophic-releasing hormone analogs, or prostogenic drugs, but these remain controversial. We report here that a light dose of combined oral contraceptive pill continuous use for one year gave excellent symptomatic relief for our patient with this benign thoracic lesion.

Conclusion

We present the use of the combined oral contraceptive pill for symptomatic treatment of bronchial endometriosis, as an effective option for medical management.

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