

## **Endometriosis Presenting as Pleural Effusion and Haemoperitoneum: Case Report and a Review of the Literature**

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### **Abstract**

A case of endometriosis presenting as haemorrhagic pleural effusion and ascites in a 40 year old multiparous woman is described. Her main clinical features were left groin swelling 12 years prior to this presentation which was confirmed at histology (after excision) to be endometriosis; secondary infertility, peri-umbilical nodule with cyclic pain especially during the menstrual flow; secondary dysmenorrhoea, dyspareunia, irregular menses, inter-menstrual bleeding, dyspnoea and abdominal swelling. Investigations revealed pleural effusion on chest X ray and a complex pelvic-abdominal mass with ascites on ultrasonography. She was managed with danazol, thoracotomy and, eventually, laparotomy. Subtotal hysterectomy with bilateral salpingoophorectomy was performed and histological examination of the tissues confirmed endometriosis and adenomyosis.

**Key Words:** Endometriosis, Pleural effusion, Ascites.

### **Introduction**

Due to paucity of diagnostic laparoscopy facilities, endometriosis seems rare in our environment. An incidence of 3-10% in women of reproductive age group and 25-35% in infertile women has been reported<sup>1,2</sup>. Less common is the occurrence of endometriosis at distant sites like skin, lungs, limbs, brain and other soft tissues or even in non-menstruating women as in Turner syndrome<sup>1</sup>. We report here a patient with haemorrhagic pleural effusion, ascites and cutaneous endometriosis.

### **Case Report:**

Mrs. A.N. was a 40 year old woman, Para 0<sup>+</sup> who presented at the Accident and Emergency Unit of Nnamdi Azikiwe University Teaching Nnewi in October 2004 with a 2-month history of shortness of breath on exertion and massive abdominal swelling. The symptoms got worse during her menstrual flow. There was no associated history of cough, haemoptysis, or loss of weight. Five years prior to presentation, she also developed a peri-umbilical induration with cyclic pain which later started discharging brownish fluid especially with onset of menstruation.

She was first seen by the physicians before referral to the gynaecology team. She had been married for 12 years with secondary infertility. She had surgical termination of an 8-week unintended pregnancy about a year before her marriage. She had a left groin lesion with cyclic pain which was excised and confirmed at histology to be endometriosis 12 years before presentation. Her last menstrual period was on 10<sup>th</sup> October 2004. She used to have regular 4 day menstrual flow in a 28-day cycle until 5 years prior to presentation when the cycle became irregular, with intermenstrual bleeding, postcoital bleeding, epimenorrhoea and menorrhagia. She also had dysmenorrhoea and deep dyspareunia. She had no further treatment for the cutaneous endometriosis

diagnosed 12 years before. However she had several courses of ovarian stimulation with clomiphene citrate prior to this presentation.

On examination, she looked depressed, well nourished, afebrile, anicteric and mildly pale. There was no pedal oedema. She had a pulse of 116 beats per minute, the blood pressure was 160/100mmHg; the apex beat was palpable on the 5th left intercostal space anterior axillary line; the 1st and 2nd heart sounds were heard with no murmurs. She had mild dyspnoea, with tracheal deviation to the left. The right side of the chest was stony dull on percussion. Tactile fremitus was absent; vocal resonance and breath sounds were diminished on the same side while the left side was normal. The breasts were normal. Abdominal examination revealed a left groin scar, peri-umbilical dark brownish firm mass attached to the skin, an ill defined 16-week size lower abdominal mass which was firm, tender and slightly mobile. There was moderate ascites. Vaginal examination revealed a fleshy mass at the external cervical os which measured about 3cm in diameter. Bimanual examination could not be performed due to pain. An impression of endometriosis with pleural effusion and ascites was made.

Chest X - ray confirmed right sided pleural effusion while abdomino-pelvic ultrasound findings suggested a complex ovarian mass with ascites. The liver enzymes, serum bilirubin, electrolytes, urea and creatinine levels were all normal. Ascitic fluid had no acid-fast bacilli and no bacterial growth on the culture. The erythrocytic sedimentation rate was 75mm per hour.

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She had thoracotomy with chest tube insertion and controlled drainage of the pleural effusion. A total of 2900ml of haemorrhagic fluid was drained over the next 3 days. She received other palliative treatment including analgesics, haematinics and antibiotics. She was started on Danazol 400mg daily. She responded very well to this treatment with resolution of symptoms and was discharged home after 6 days of admission.

During the follow up, hysterosalpingogram was done to evaluate tubal patency. This revealed a distortion of the uterine cavity, stretching of the cervical canal, intravasation of dye into the adnexae and both fallopian tubes were not outlined. An impression of bilateral tubal occlusion with leiomyoma was made. However, patient was not seen again until after three months when she now presented with rapidly increasing abdominal swelling of one week duration. She had her last menstrual period about one week prior to the worsening of the symptoms. She had stopped her medication during her absence from follow up. General examination did not reveal any significant finding; the chest was normal on auscultation; the vital signs were normal. Abdominal examination revealed persistent brownish discolouration of peri-umbilical nodule and ascites. An impression of ruptured endometrioma was made. She was admitted and counselled for exploratory laparotomy after laboratory work up.

At laparotomy three litres of haemorrhagic peritoneal fluid was drained but there was no evidence suggestive of malignant metastatic deposits. There was a ruptured endometriotic cystic deposit at the utero-vesical junction and copious pelvic adhesions. The uterine size was equivalent to a 16-week gestation, with increased vasculature. The tubes and ovaries were wrapped up in a mesh of adhesions and could not easily be discerned. A subtotal hysterectomy with bilateral salpingo-oophorectomy and excision of the endometriotic deposit was done and specimens sent for histology. The post operative recovery was uneventful; she was later counselled together with the husband on the implication of the surgery on her reproductive potential. The histology report confirmed endometriosis and adenomyosis.

### **Discussion**

This is the first case of confirmed haemorrhagic pleural effusion and ascites due to endometriosis recorded in our hospital. Since this presentation is very uncommon, it is easy to approach the treatment as one would a malignant disorder, with all the implications attendant to that.<sup>3,4</sup> Extra-pelvic endometriosis is very rare and thus can easily be missed. However a history of cyclical exacerbation of symptoms will aid the diagnosis<sup>1,4,5</sup>. This was partly the case in this patient especially with respect to the cutaneous lesions.

The diagnosis in this patient was enhanced by the previous histologically confirmed groin lesion. Cytology is not very helpful in diagnosis hence the aspirates of both pleural and peritoneal fluid were not subjected to examination for endometriotic cells<sup>6</sup>. The occurrence of endometriosis at the ectopic sites could possibly be by coelomic metaplasia, lymphatic/haematologic migration, immunological or genetic theory as there was no previous open uterine surgery to explain the implantation of endometrial tissue on the skin.

The management include conservative, medical and surgical treatment. The method adopted depends on the stage of the disease, reproductive wish of the patient, and her age<sup>6,7</sup>. This patient had stage 4 endometriosis using the scoring system of the American Society for Reproductive Medicine. She was still in her reproductive age with associated infertility. She desired conservative treatment to preserve her fertility. Following thoracotomy and the drainage of the pleural effusion, a trial of hormonal therapy was adopted to alleviate her symptoms. However, surgery became necessary when she developed acute abdomen due to ruptured endometriotic cyst and worsening condition<sup>7,8</sup>. We could not do total hysterectomy because of the extensive adhesion that hindered access to the cervix. However leaving the cervix behind has been found useful in sexually active women for maintenance of deep or cervical orgasm and genital lubrication during coitus<sup>9</sup>. The risk of cervical cancer is however a threat despite the negative Pap smear. She would therefore still benefit from regular Pap smears.

Apart from thoracotomy and tubal drainage of pleural effusion, patients without reproductive wish can be sustained on hormonal treatment while conserving the uterus to prevent recurrence; pleurodesis is also another form of surgical treatment<sup>10,11</sup>. Of all these, the most effective treatment is hysterectomy and bilateral salpingo-oophorectomy<sup>12</sup>. However there may still be recurrence in a small percentage of cases<sup>1,6</sup>. Performing hysterectomy and bilateral salpingo-oophorectomy was a difficult decision in a patient with a background of prolonged infertility/nulliparity, but she needed to be alive and well to bear children. We however counselled her on alternatives like child adoption or surrogacy. She is currently on hormone replacement therapy to reduce the effects of surgical menopause<sup>12</sup>. The risk of exacerbation or recurrence due to the treatment is however very minimal and she is being followed up for early detection of such risk<sup>13</sup>.

Endometriosis generally presents a management dilemma ranging from diagnosis, treatment to follow-up. It seems rare in this environment because of lack of suspicion and confirmatory methods of diagnosis which

is laparoscopic tissue diagnosis. Prevention is not easy but early marriage with pregnancy is fairly protective. Nevertheless endometriosis cannot yet be predicted with certainty. There is no doubt that if our patient had

presented at the early stage she would have had a better outcome, including better chances of bearing her own children<sup>6</sup>.

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