

Endometriosis presenting as relapsing haemorrhagic ascites in a South Asian woman: A case report

Ushna Ahmed, Novera Chughtai

Abstract

Endometriosis presenting as ascites is a rare entity, and is more so in women of Asian ethnicity. Less than a hundred cases have been reported worldwide. Majority of patients present with abdominal distension and pain, draining massive blood stained serosanguineous fluid. This hinders future fertility prospects of these women. Ovarian suppression has been employed as a successful treatment, followed by definitive surgical treatment, such as bilateral salpingo-oophorectomy, to end the possibility of recurrences, which are otherwise always possible. We present the case of a woman of reproductive age, seeking fertility treatment, who had a more subtle presentation of moderate, but relapsing ascites of unknown origin in the past two years. Diagnostic laparoscopy and histopathology of the peritoneal deposits suggested endometriosis. Her ovarian function was suppressed, and she is currently underway of assisted reproduction for achieving a pregnancy.

Keywords: Endometriosis, ascites, ovarian suppression.

DOI: <https://doi.org/10.47391/JPMA.9438>

Introduction

Endometriosis, a benign, oestrogen dependent disease, with ectopic implantations of endometrial glands and stroma, affecting 6-10% of all women of reproductive age, and can present as chronic pelvic pain, dysmenorrhoea, dyspareunia, and infertility.^{1,2}

Endometriosis-related ascites was initially described by Brews in 1954, and further explored by Charles in 1957.³ It is a rare occurrence, more so in women of Asian ethnicity. Fewer than a 100 reported cases recognised nulligravid, African women as the primary victims. Recognising this diagnosis is further complicated by the presence of ascites in other circumstances like malignancy, infection, cirrhosis, or trauma.⁴ Thus the purpose of this case report is to propose as to how a diagnosis of ascites secondary to

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Submission complete: 04-04-2023

Review began: 19-05-2023

Acceptance: 06-09-2023

Review end: 06-09-2023

endometriosis could be suspected in Asian women and what would be its predictable course and management options.

Case Report

A nulliparous 24-year-old south Asian woman, married for 3 years and desiring fertility, presented on 7th December, 2021 at Aga Khan University Hospital, Karachi with the complaint of abdominal distension and abdominal pain for 2 years, denying any experience of weight loss, weakness or fatigue. Menstrual cycles were complicated by moderate to severe dysmenorrhoea and deep dyspareunia. No contraceptives were ever used. No other associated medical or surgical problems were reported and her family history was unremarkable.

She was being investigated for primary subfertility for two years at Civil Hospital Sukkur, when ascites was first discovered on imaging. Since then, it had been drained percutaneously thrice, once about 900 ml, and further investigated using cytology and imaging. It re-accumulated at all times. The cytology tested negative for malignancy and Acid-Fast Bacilli for tuberculosis. Her liver function tests, complete blood picture, renal function tests, acute inflammatory markers and ovarian tumour markers (both surface epithelial and germ cell) were within normal range.

Empirical treatment for suspected tuberculosis was administered for 3 months in 2019, which was in vain. She underwent diagnostic laparoscopy in 2019 for ascites of unknown aetiology at Liaquat National Hospital, which demonstrated moderate haemorrhagic ascites, and peritoneal deposits, with extensive inter bowel, uterine, ovarian and fallopian tube adhesion. These deposits were biopsied, and histopathology suspected endometriosis. However, no intervention.

Computerized tomography (CT) conducted in 2019 suggested moderate pelvic ascites and a left sided 2.9 x 2.3 cm haemorrhagic ovarian cyst. In 2020, she undertook one ineffective cycle of clomiphene citrate for ovulation induction.

On presentation to our clinic, she was in good health, vitally stable, and had a BMI of 27.9 kg/m². Her abdomen was



Figure-1: CT scan of abdomen and pelvis suggesting moderate ascites.

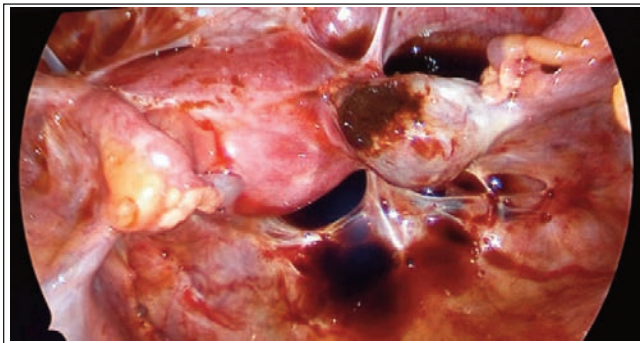


Figure-2: Laparoscopic findings showing intraperitoneal adhesions and peritoneal deposits.

mildly distended. Fluid thrill and shifting dullness were positive. There were no masses or tenderness upon palpation. Examination revealed a healthy cervix, an anteverted, mobile, non-tender uterus, 6 weeks in size, and fullness in both adnexa with no tenderness. A repeat CT scan of the abdomino-pelvic region suggested moderate ascites and a simple 2.9x2.6 cm right sided ovarian cyst, depicted in figure 1. Her CA-125 level, CA19-9, and CEA levels and germ cell tumour markers were within the normal range.

A diagnostic laparoscopy and tubal patency testing was performed and 600 ml of haemorrhagic fluid was drained. Extensive adhesions were prominent between the uterus, fallopian tubes, ovaries and the bowel (Figure 2). Abdomen and pelvic peritoneal surfaces, and the uterine surface were covered in bleb like structures. Multiple peritoneal biopsies were obtained. No adhesiolysis was attempted, and both fallopian tubes were found to be patent via dye test. Patient remained stable and pain free postoperatively.

Cytology of the ascitic fluid revealed no malignant cells, negative for tuberculosis, and did not grow any microorganism on culture. Histopathology of the

peritoneal biopsies suggested endometriosis.

Postoperatively, she received three doses of Goserelin, 3.6 mg, subcutaneously, in an effort to suppress ovarian function, with the last dose being delivered on 8th February, 2022. An evaluation regarding assisted reproduction in the upcoming months has been planned for her.

Discussion

Ascites secondary to endometriosis is a rare phenomenon, first reported in 1954.³ Cirrhosis of the liver is the most common aetiology.⁴ Conversely, 20% of cases are not linked to liver cirrhosis.⁵ In developed countries, tumours and heart diseases are the predominant causes, while developing countries are mainly concerned with infectious diseases, among which tuberculosis is the leading association.^{5,6} Theories postulated are either a shedding of endometrial cells into the peritoneal cavity so that peritoneal cells are stimulated and ascites may eventually occur,⁷ a consequence of excessive ovarian transudation similar to a Meigs syndrome⁸ or rupture of endometriotic cysts resulting in irritation of peritoneal cells and subsequent formation of a reactive exudate.⁸ None of these theories have been definitely proven yet.

Endometriosis linked ascites simulates various gynaecological malignancies⁹ such as ovarian, fallopian tube, or primary peritoneal carcinomas. Their symptoms overlap, with poor appetite and weight loss described in one-third of reported cases of endometriosis associated ascites, the fourth most common symptom.^{4,9} Thus, the diagnosis of this clinical entity is made after the exclusion of other more common causes. The cases reported had undergone abdominal paracentesis for cytological analysis, and tissue biopsy for histopathology, to exclude malignant cells and tuberculosis infection.^{6,7,9}

Asian women developing endometriosis-linked ascites, such as in this case is very rare, with 16.13 % of the reported women being Asian, against the bulk of 69.35 % African descent.⁴ These women were in fact more frequently nulliparous. The most common presenting complaint has been abdominal distension.^{4,9} Reported ascites has predominantly been massive, averaging 4 litres, and as much as 7 litres in cases, with a high risk of recurrence.^{4,6,7,9} Acute recurrences have been as early as eight days.⁶ So severity of ascites could be proportional to severity of endometriosis.⁴ Such ascites may even be concurrent with pleural effusion, presenting in 40% of the reviewed subjects¹⁰ with thoracentesis having been employed as an efficient and convenient approach for drainage.^{7,10}

The utility of CA-125 in the diagnosis of endometriosis is

greatly diminished for diagnosis and prognosis, due to its non-specificity. In several cases it was found only to be mild to moderately elevated,^{4,6,9} while our case reported it within normal range.

The intra-operative findings in our case were similar to those reported in other studies: extensive pelvic peritoneal adhesions, extra-pelvic endometriosis, and dark brown serosanguineous fluid; all being suggestive of advanced disease.^{4,6,7,9,10}

The medical management of such ascites aims to suppress ovarian function, which is the corner stone of the treatment of endometriosis.⁴ About 97% of patients reported using hormonal therapy, such as GnRH agonist, progesterone, combination oral contraceptive (COCP) pills or a combination.^{4,9} Danazol as an anti-gonadotropic, anti-oestrogenic synthetic steroid is effective, but its various androgenic effects preclude its use.⁹ Recent studies support GnRH agonists. These are effective in achieving ovarian suppression and increasing fertility rates. But the anti-androgenic adverse effects are an impediment, limiting its use to a maximum of 6-months.^{4,6,9} New progestins are now recommended over COCP pills for endometriosis. Dienogest, the new fourth-generation progestin, offers an effective and tolerable alternative to GnRH analogues by not reducing oestradiol concentration to post-menopausal levels, and can also be used for maintenance.⁹ But with medical management; recurrence rate is unanimously high with cessation of therapy.^{4,6,9,10}

Surgically, one can start off with diagnostic laparoscopy, in an attempt to drain ascitic fluid and obtain peritoneal biopsies for tissue diagnosis, ablate endometriotic lesions, or perform resection, and adhesiolysis.⁹ However, these techniques are temporizing and the recurrence of both ascites and endometriosis is common.⁹ Fertility sparing surgery is optimal, owing to these women being nulliparous and in reproductive age.⁴ Earlier, around 36.5% of such patients underwent exploratory laparotomy, with a significant number of cases proceeding to hysterectomy combined with bilateral salpingo-oophorectomy.⁷ The most successful treatments were bilateral salpingo-oophorectomy, which reported no recurrences.^{4,7} With the advent and extensive availability of minimally invasive techniques and increasing technical confidence among surgeons, a trend could be to use laparoscopy to establish histologic diagnosis using frozen section, and after confirmation, to plan resection, such as followed for suspected malignant ovarian tumours.⁹

Conclusion

Endometriosis associated ascites is a rare occurrence in Asian women. It should be suspected in women of child

bearing age presenting with abdominal distension and subfertility, particularly for over two years duration, with massive haemorrhagic ascites. It has to be meticulously differentiated from other benign, and more importantly, malignant diseases. Its association with severe endometriosis needs to be realised, to prevent its recurrence. Minimally invasive surgical techniques can be used to establish a diagnosis, and for planning an intervention. For women desiring fertility, hormonal suppression is the way to go, using a single or combination of medications. This can then be supplemented with the in vitro fertilization technique to expedite conception. Following completion of fertility, definitive surgical procedures, like bilateral salpingo-oophorectomy, could be done to terminate the chances of recurrence. Our patient presented with a more subtle course of the disease, with only moderate ascites, and normal CA-125. Hence, multiple possible clinical scenarios can be encountered, urging clinicians to be more vigilant.

Disclaimer: None.

Conflict of interest: None.

Funding disclosure: None.

Consent for publication: Written consent was obtained from the patient to publish the case report.

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Author Contribution:

UA: Literature search, data collection and drafting.

NC: Supervision, study design and concept, drafting