



Case Report

Polypoid endometriosis – A rare entity of endometriosis mimicking ovarian cancer

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ABSTRACT

Objective: To report a rare case of polypoid endometriosis with initial impression of ovarian cancer and review the published literature about this disease.**Case report:** A 23-year-old female presented with sudden onset of acute lower abdominal pain. Image studies revealed an irregular shaped, heterogeneous mass at the cul-de-sac, but without ascites or enlargement of pelvic or paraaortic lymph nodes. Blood tests showed an elevated CA-125 value (1317 U/ml). Resection of the mass was performed by laparotomy and the frozen section and final pathology both revealed polypoid endometriosis. Post-operative gonadotropin-releasing hormone agonist was given for 6 months followed by oral contraceptives. She remained disease free 3 years after operation.**Conclusion:** Polypoid endometriosis is an uncommon and distinctive variant of endometriosis. Gynecologists should be aware of this rare form of a commonly benign disease to avoid excessive resection in younger patients of childbearing age.© 2019 Taiwan Association of Obstetrics & Gynecology. Publishing services by Elsevier B.V. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

Introduction

Endometriosis is defined as presence of viable, estrogen-sensitive endometrial-like glands and stroma outside the uterus [1]. This disorder may cause scar tissue and adhesion formation, and result in severe dysmenorrhea and infertility among women of childbearing age. Endometriosis affects about 6–10% of women of reproductive age. About 50–60% of women and teenage girls with pelvic pain and up to 50% of women with infertility were related to this disease [2].

Polypoid endometriosis is an uncommon and distinctive variant of endometriosis with histological features simulating an endometrial polyp. It was first introduced by Mostoufizadeh and Scully in 1980 [3]. This type of endometriosis may form large, often multiple, polypoid masses that “not only simulate malignant tumors at operation but may also recur after operative removal” [3].

In this paper, we report a young female who presented with a pelvic tumor mimicking ovarian cancer and was later diagnosed to be polypoid endometriosis.

Case report

A 23 year-old nullipara female suffered from progressive dysmenorrhea for 2 years. Gynecologic ultrasounds performed 6 months ago did not show any lesions. This time, she experienced sudden onset of acute lower abdominal pain without vaginal bleeding. Transvaginal ultrasonography (TVS) revealed an irregular-shaped, heterogeneous mass measured 10 cm at the cul-de-sac (CDS) (Fig. 1A) with clear visualization of the uterus and bilateral adnexa. Lab tests showed an elevated CA-125 (1317 U/ml, normal range: < 35 U/ml). Other tumor markers were all within normal limits (CEA: 1.2 ng/ml, AFP: 2.0 ng/ml, β -hCG: <0.1 mIU/ml, LDH: 167 IU/L). Abdominal and pelvic computed tomography scan showed an irregular-shaped pelvic tumor (Fig. 1B) with no enlargement of pelvic and para-aortic lymph nodes.

She received exploratory laparotomy under the impression of pelvic tumor. During operation, grayish tan colored soft tissue resembling a polypoid, cauliflower-like structure occupying the

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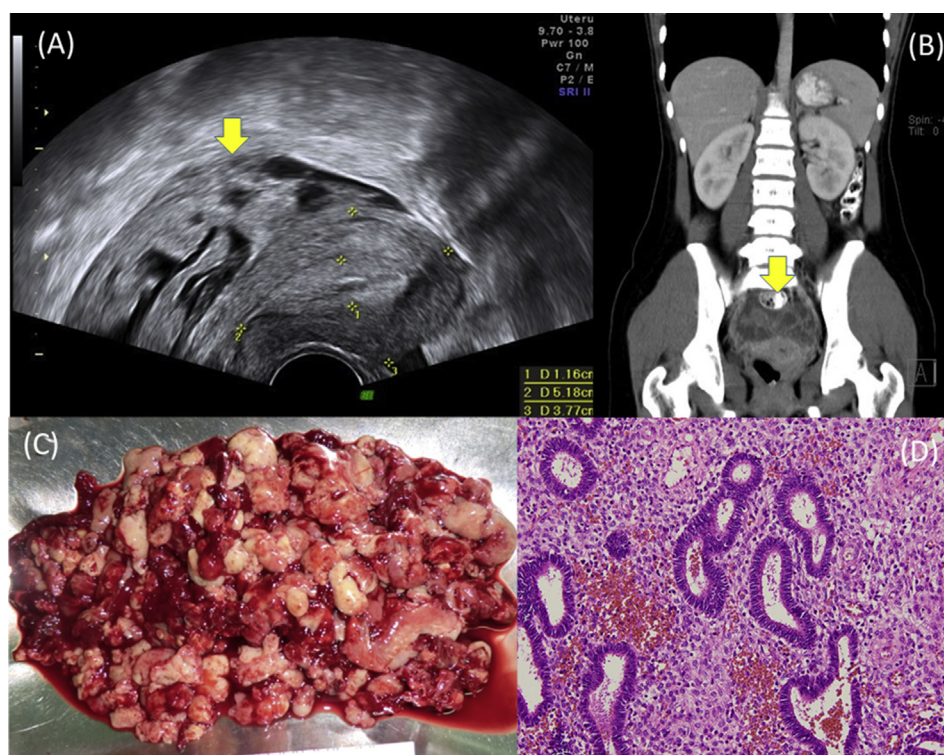


Fig. 1. **A:** The vaginal ultrasound demonstrated an irregular shaped, heterogeneous, 10 cm mass posterior to the uterus (arrow). **B:** Computed tomography (CT) imaging of the abdomen and pelvis showed an amorphous, irregular shaped mass in pelvis (arrow). **C:** Gross finding of polypoid endometriosis. Multiple fragile polypoid nodules were found in pelvic cavity. **D:** Microscopic finding showed typical endometrial glands and stroma with focal hemorrhage (H&E, $\times 200$).

CDS was found (Fig. 1C). Intraoperative frozen section and final pathologic report both confirmed the diagnosis of polypoid endometriosis (Fig. 1D). After operation, the patient recovered well and was discharged without any complications. GnRH agonist was prescribed for 6 months followed by subsequent oral contraceptives for controlling the disease. Her CA-125 level returned to normal levels and no recurrence of endometriosis was found for 3 years after operation.

Discussion

Polypoid endometriosis was first introduced by Mostoufizadeh and Scully in 1980 [3]. It is a rare variant of endometriosis that forms polypoid masses that can mimic a neoplasm on clinical, intraoperative, and gross pathological examinations. It was mainly reported in case report or small case series. The largest case series was published by Parker et al. [4] and only included 24 cases. According to previous studies, polypoid endometriosis mainly affects older women, with a mean age of 52.5 years old. The most common site of involvement were ovarian surface and colonic serosa, and size ranging from 0.5 cm to 15 cm [4].

The most common clinical presentations of polypoid endometriosis are pelvic mass, polypoid vaginal mass, and large bowel obstruction. It may cause symptoms of dysmenorrhea, dyspareunia, chronic pelvic pain, and infertility [4]. The actual pathogenesis of this disease is still not clear, but there are several hypotheses, including conventional retrograde menstruation theory with involvement of mucosal or subserosal sites, or the lining of cyst cavities, permitting polypoid growth [2]. Other theories include hormonal stimulation, including unopposed estrogen and combined estrogen-progestin therapy, tamoxifen use, or even following withdrawal of gonadotrophin-releasing hormone (GnRH) agonist usage [2,4,5].

Endometriosis is a common gynecological pathological process with a straightforward diagnosis, but there are many histological variants of this common condition, some of which can present with diagnostic difficulty [6]. Differential diagnoses include adenocarcinoma and other primary ovarian malignant tumors with hemorrhage, such as endometrioid adenocarcinoma and clear cell adenocarcinoma arising from endometriosis. In our case, an intraoperative frozen diagnosis of a benign mass prevented a more extensive surgery in this young woman. Polypoid endometriosis is a rare form of endometriosis that may present as a lesion of rapid growth mimicking ovarian malignancies. We emphasized that gynecologists and pathologists should be aware of this rare form of a commonly benign disease to avoid excessive resection in younger patients of childbearing age.

Conflict of interest

All the authors state that there are no conflicts of interest to disclose.

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