CASE REPORT:

Adolescence endometriosis with abnormal uterine bleeding: A challenging case report

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ABSTRACT

Endometriosis is the presence of endometrial tissue outside the Uterus. The true incidence of endometriosis is not really known, but it is believed that 10-15% of all women in their reproductive age will develop endometriosis and 25-35% of all women who are infertile have endometriosis. The true incidence of endometriosis in adolescents is difficult to quantify and estimates vary among different studies. Although in the past it was assumed that endometriosis presented only after many years of menstruation, studies have shown endometriosis to occur prior to menarche and between 1-6 months after the onset of menarche. A 66% of adult women with endometriosis report the onset of pelvic symptoms prior to age 20. Although etiology and pathophysiology of endometriosis is not well-understood, but increased rate of retrograde menstruation in patients with heavier menses as with bleeding disorder, is a possible mechanism of disease. Here, we report an early adolescent admitted with dysmenorrhea with bleeding disorder, suspected Von Willebrand disease, presenting abnormal uterine bleeding.

Keywords: endometriosis, adolescence, bleeding disorder, blood coagulation disease, abnormal uterine bleeding

ABSTRAK

Endometriosis adalah adanya jaringan endometrium di luar rahim. Insiden endometriosis sebenarnya tidak benar-benar diketahui, tetapi diyakini bahwa 10-15% dari semua wanita di usia reproduksi mereka akan mengembangkan endometriosis dan 25-35% dari semua wanita yang mandul memiliki endometriosis. Insiden endometriosis yang sebenarnya pada remaja sulit untuk dikuantifikasi dan perkiraan bervariasi di antara berbagai penelitian. Meskipun di masa lalu diasumsikan bahwa endometriosis hanya muncul setelah bertahun-tahun menstruasi, penelitian telah menunjukkan endometriosis terjadi sebelum menarke dan antara 1-6 bulan setelah onset menarke. Sebanyak 66% wanita dewasa dengan endometriosis melaporkan timbulnya gejala panggul sebelum usia 20 tahun. Meskipun etiologi dan patofisiologi endometriosis tidak dipahami dengan baik, tetapi peningkatan tingkat menstruasi retrograde pada pasien dengan menstruasi yang lebih berat seperti dengan gangguan perdarahan, adalah mungkin. mekanisme penyakit. Di sini, kami melaporkan seorang remaja awal yang dirawat dengan dismenore dengan gangguan pendarahan, diduga penyakit Von Willebrand, yang menunjukkan pendarahan rahim yang abnormal.

Kata kunci: endometriosis, remaja, gangguan perdarahan, penyakit pembekuan darah, perdarahan uterus abnormal

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INTRODUCTION

Endometriosis is defined as a growth of ectopic endometrial tissue outside the uterine cavity that responds to hormonal stimulation, causing dysmenorrhea, dyspareunia, menstrual irregularities and infertility.[1] It occurs most commonly in the pelvic sites such as the ovaries, pouch of douglas, ligaments of the uterus, pelvic peritoneum and rectovaginal septum.[2] The true incidence of endometriosis in adolescents is difficult to quantify and estimates vary among different studies with a literature reported a 47% incidence of endometriosis in adolescent undergoing laparoscopy for chronic pelvic pain.[3,4] Another literature reported a 67% incidence of endometriosis in adolescents who have pain refractory to common medical treatments like nonsteroidal anti-inflammatory agents (NSAIDs) or oral contraceptive pills (OCPs).[5] Although etiology and pathophysiology of endometriosis is not well-understood, but increased rate of retrograde menstruation in patients with heavier menses, is a possible mechanism of disease. Unfortunately, there is a weak evidence that female with bleeding disorder are likely to develop endometriosis.[6]

CASE REPORT

A 13 year old adolescent had complained a dysmenorrhea and heavy menstrual bleeding since 1 year before admission. These complains started 2 months after her menarche and occurred each time her period every month. She complains about 7-10 pads a day for 10 consecutive days. She was previously a healthy woman with no significant medical history, especially blood coagulation disease. Blood sample was taken and revealed that there was severe microcytic hypochromic anemia with hemoglobin level was 5.8 g/d, prolonged prothrombin time (PT) 4.7x control, and prolonged activated partial thromboplastin time (aPTT) 4.7x control, suspected blood coagulation disease such as Von Willebrand disease. Unfortunately, there was a normal VIII and IX factor, 122.9% (normal range 40-170%) and 112.6% (normal range 51-137%) with normal antigen VWF 94% (normal range 54-148% for O blood group). Blood smear revealed a normal count and morphology of platelet, but severe microcytic hypochromic anemia with anisopoikilocytosis, existence of oval, pencil, target cells.

Ultrasound findings showed multilocular left cystic ovarian neoplasm sized 7 cm in diameter with inhomogenous echogenicity and irregular border suspected cyst ovarian neoplasm with hemorrhage (Figure 1). We performed CT Scan with contrast and found a mixed encapsulated solid and liquid mass attached to left lateral uterus sized 8 cm in diameter suspected left ovarian neoplasm (Figure 2 & 3). On laparoscopic view, there was 8 cm left adnexa cyst with chocolate fluid inside corresponds to suspected endometriosis cyst. We performed laparoscopic cystectomy and tissue was sent to histopathology examination which revealed an endometriosis cyst of the ovary.

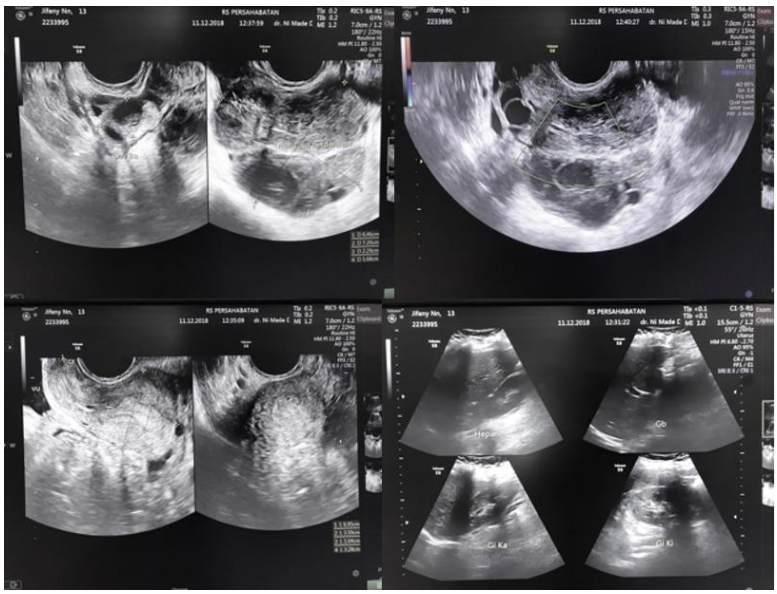

Figure 1. Ultrasound findings (transrectal)

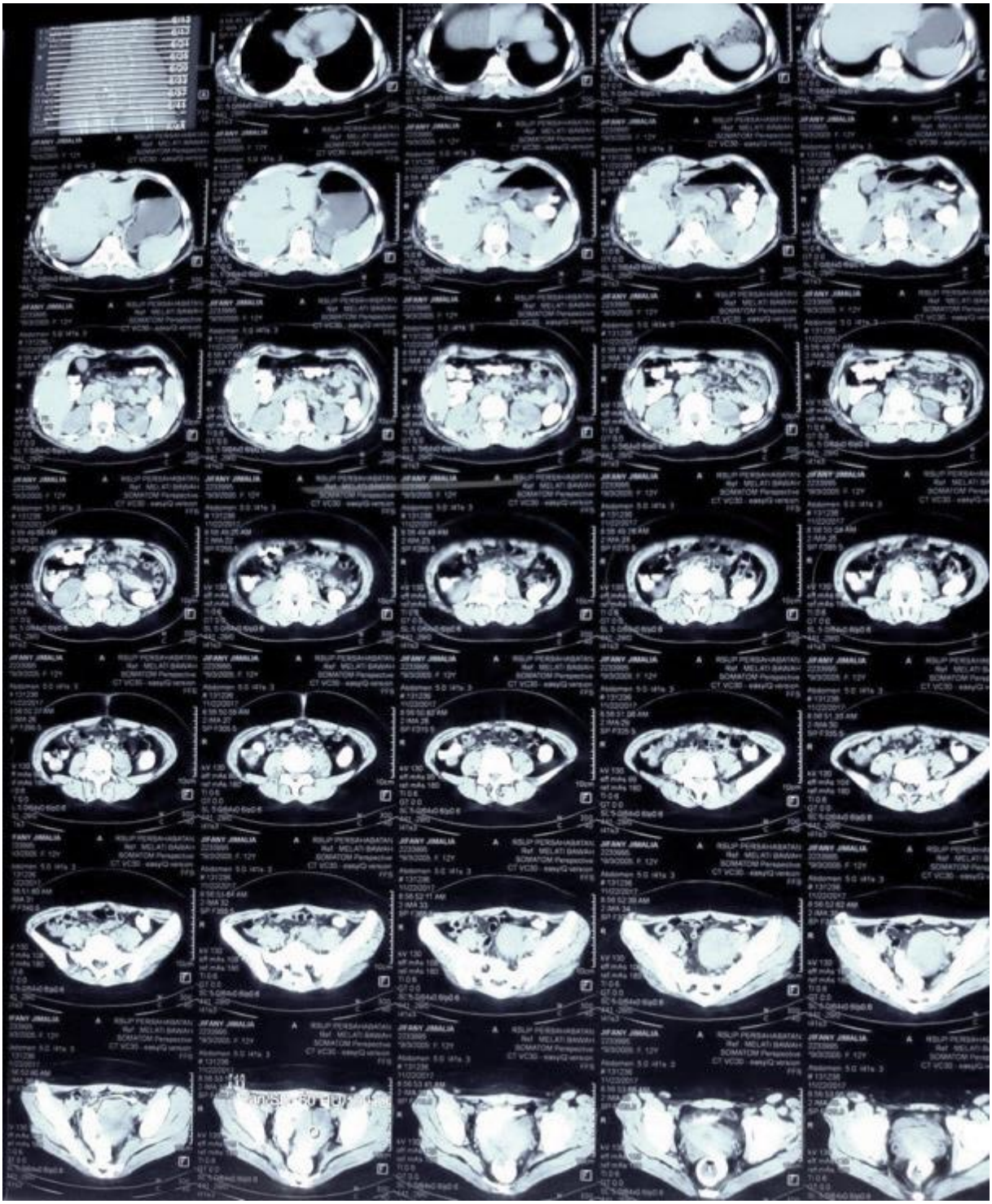

Figure 2. Axial sections of contrast CT scan.

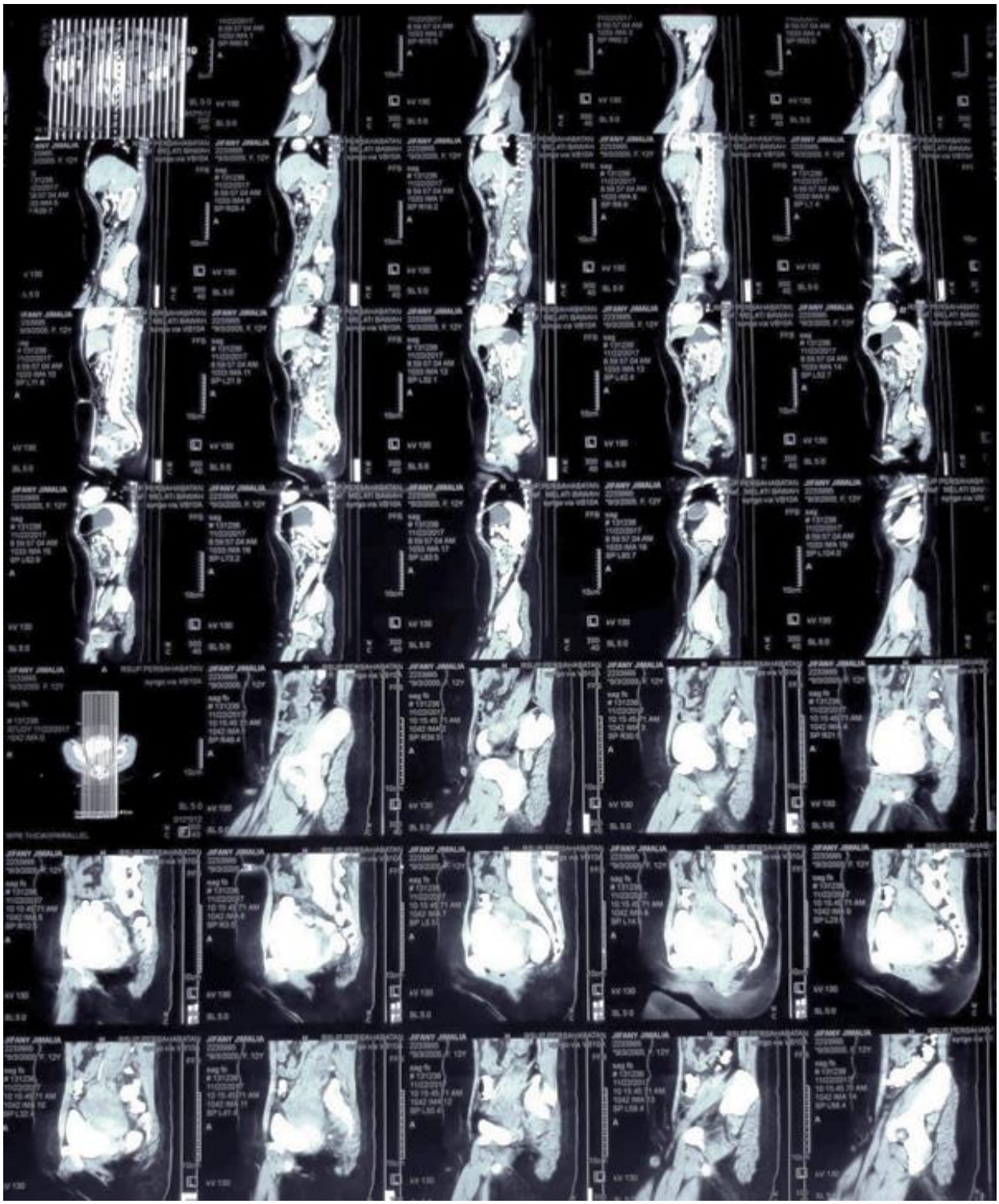

Figure 3. Sagittal sections of contrast CT scan.

DISCUSSION

Endometriosis is defined as the presence of functional endometrium tissue outside the uterine cavity in which it is normally localized. It affects 10–15% of all women of childbearing age and 6% of perimenopauses women.[7] Although in the past it was assumed that endometriosis presented only after many years of menstruation, studies have shown endometriosis can occur prior to menarche and between 1–6 months after the onset of menarche.[5] According to The Endometriosis Association, 66% of adult women with endometriosis report the onset of pelvic symptoms prior to age 20.[8,9]

Many theories have been proposed to explain the pathophysiology of endometriosis, leading the name of the "disease of theories." In 1927 Sampson suggested that retrograde menstruation through the fallopian tubes results in seeding the pelvis with endometrial tissue. These endometrial cells implant themselves on the peritoneal surface of abdominal and pelvic organs and tissues. With consecutive menstrual cycles, they undergo proliferation and bleeding, with the possibility of causing further disseminations and implants.[10] This theory does not explain the presence of disease in women with Mullerian agenesis, aplasia, or endometriosis before menarche with or without obstructive anomalies. The latter could be explained by the coelomic metaplasia theory postulated in 1929 by Meyer, which assumed that metaplasia of the multipotential coelomic epithelium is the origin of endometriosis, as peritoneal and endometrial cells are both derived from the same embryonic precursor. The presence of stress, such as inflammation or irritation (and/or abnormal estrogen stimulation) from refluxed menstrual tissues, causes the coelomic cell, previously differentiated into peritoneal cell, to transform into endometrial cells, which respond in cyclic manner. This theory not only explains the presence endometriosis in all of the possible anatomical sites, but also justifies the presence of disease in women without a uterus, in very young premenarchal girls and even in males.[4,5,9]

There is a weak evidence that female with bleeding disorder are likely to develop endometriosis. Ferrari et al., described the first association of an acquired inhibitor against factor VIII in a case of severe pelvic endometriosis.[11] Martire et al., reported catamenial hemoptysis from endobronchial endometriosis in a 12-year-old female with type 1 von Willebrand disease (VWD). In a survey by the US Center for Disease Control of 102 female withVWD, the gynecological problems including endometriosis, fibroids, uterine polyps, and endometrial hyperplasia reported more commonly in females with VWD in comparison to the controls.[12] Menorrhagia, hemorrhagic ovarian cysts, endometriosis, and postpartum hemorrhage are symptoms of VWD in women.[13,14] Although menorrhagia is the most common gynecologic manifestation of a bleeding disorder, but it is not the only manifestation and it seems that females with bleeding tendency have an increased risk of developing hemorrhagic ovarian cysts and probably endometriosis.[15,16] Since, underlying bleeding tendency may cause heavy menstrual bleeding, dysmenorrhea, intermenstrual bleeding, and endometriosis; thus coagulation study tests should be considered in women with menstrual abnormalities if they have a positive history of abnormal bleeding.[17]

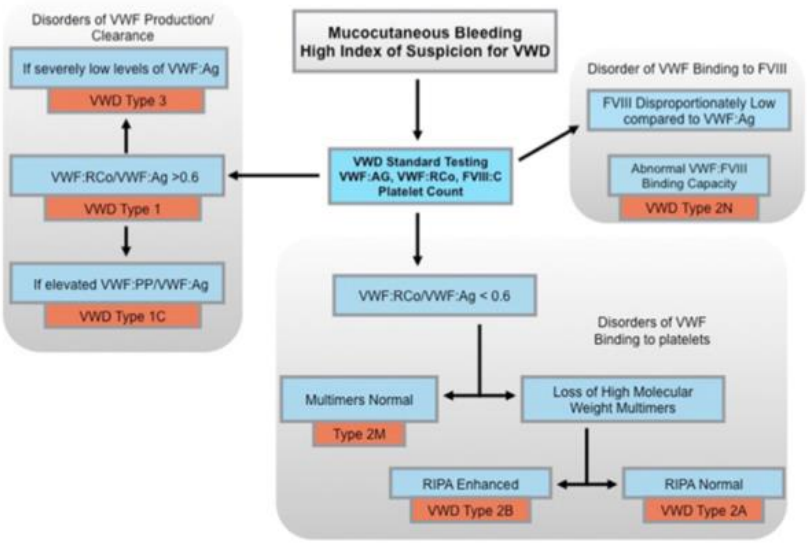

Figure 4. Algorithm of the diagnostic approach to mucocutaneous bleeding with a high clinical suspicion for VWD.[18]

Table 1. Laboratory patterns of different types of VWD.[18]

	VWF:Ag	VWF:RCo	VWF:Rco/VWF:Ag	FVIII:C	Multimer pattern	Platelet count	Specialized testing
Quantitative disorders of VWF							
Type 1	↓	↓	≥0.6	Normal to ↓	Normal	Normal	
Type 1C	↓	↓	≥0.6	Normal to ↓	Normal	Normal	Increased VWFpp/VWF:Ag ratio
Low VWF levels	↓	↓	≥0.6	Normal to ↓	Normal	Normal	
Type 3	↓↓↓	↓↓↓	≥0.6	↓↓↓	Absent	Normal	
Quantitative disorders of VWF							
Type 2A	↓	↓↓	≤0.6	Normal to ↓	Loss of HMW-VWF	Normal	
Type 2B	↓	↓↓	≤0.6	Normal to ↓	Loss of HMW-VWF	↓↓	LD-RIPA enhanced with patient-derived plasma (VWF)
Platelet-type VWD		↓↓	≤0.6	Normal	Loss of HMW-VWF	↓↓	LD-RIPA enhanced with patient-derived platelets
Type 2M	↓	↓↓	≤0.6	Normal to ↓	Normal	Normal	
Type 2N	Normal to ↓	Normal to ↓	≥0.6	↓↓	Normal	Normal	Decreased VWF:FVIII-binding capacity

HMW, high molecular weight. ↓, mild decrease: ↓↓, moderate decrease: ↓↓↓, severe decrease.

In our case, actually we could not conclude Von Willebrand disease for the patient. Christopher et al revealed some laboratory tests which can be used to diagnose Von Willebrand disease and its type. Although widely used and important in the diagnosis of VWD, the currently available clinical evaluations and laboratory tests are not infallible; often indeterminate or unreliable results can lead to confusion in the diagnosis of VWD. They also showed investigation the type of Von Willebrand disease from the laboratory finding (Figure 4 and Table 1).[18]

CONCLUSION

Endometriosis can occur in very young adolescent, premenarchal or within 1-6 months after menarche. Besides the chief complain of dysmenorrhea, heavy menstrual bleeding can be the first manifestation of confounding factor as bleeding disorder, such as Von Willebrand disease. Screening for bleeding tendency should be done in females with endometriosis corresponding to endometriosis theory. Finding an underlying bleeding tendency and appropriate management lead to improvement of symptoms in females with endometriosis.

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