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Case Report

'Fish in a Net' and 'Swiss cheese' pattern 2 cases of unusual adenomyosis with different β–HCG level ☆

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ABSTRACT

Myometrial hypertrophy and hyperplasia, which usually on magnetic resonance imaging (MRI) typically reveal an enlarged uterus with ill-defined areas of low signal intensity and a diminished junctional zone, along with small foci of hyperintensity due to ectopic endometrium, are found in uterine adenomyosis. Those are caused by the presence of ectopic endometrial glands and stroma within the uterine myometrium. However, our case reports highlight the importance of recognizing atypical presentations, such as extensive mass-like hyperintense signals resembling a "Fish in a Net" and Swiss cheese pattern on T2-weighted imaging. Recognizing this pattern could aid in preventing misdiagnosis and guiding appropriate management strategies. Furthermore, there is a possibility that the same diagnosis (adenomyosis) could present a different β-human choriogonadotropin hormone (β-HCG) serum level.

Introduction

Adenomyosis is an abnormal condition when the endometrial gland and stromal area are located at the uterine myometrium; those could also be called uterine adenomyosis [1]. Its prevalence is 1.03%, or 28.9 per 10,000 women annually [2]. It has been stipulated that endometrial invagination into the myometrium and the metaplasia of Mullerian duct remnants are the leading theories in adenomyosis pathophysiology [3,4]. In 1960, it was first introduced as 'adenomyoma,' but in modern terminology, it is called adenomyosis [5]. As Bird said in 1972, "Adenomyosis may be defined as the benign invasion of the endometrium into the myometrium, producing a diffusely enlarged uterus that microscopically exhibits ectopic non-neoplastic endometrial glands and stroma surrounded by the hypertrophic and hyperplastic myometrium" [6].

The clinical symptoms of adenomyosis include abnormal uterus bleeding, dysmenorrhea, and pelvic pain. Adenomyosis at magnetic resonance imaging (MRI) shows a hypointense signal at T1 and T2, diminishing at the junctional zone. Furthermore, there is also a tiny foci-intensity signal, which shows an endometrial gland containing blood inside it. Regardless, imaging could distinguish adenomyosis from other

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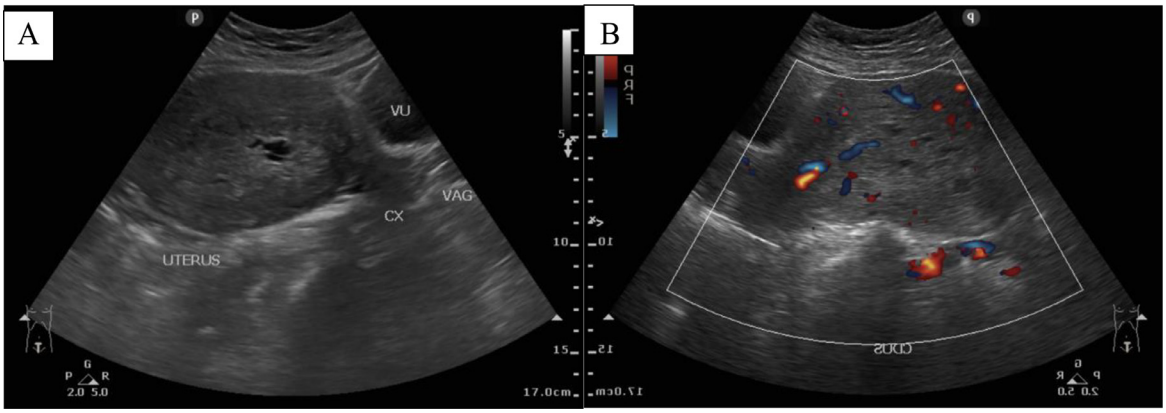

Fig. 1 – Ultrasonography examination: (A) B-mode ultrasonography of the uterus reveals enlargement of the uterus containing multiple hypo-echogenicity. (B) Hypervascularisation of the uterus is observed in a color Doppler ultrasound.

pathologies, although there are some unclassical types of adenomyosis that might be imaging pitfalls for radiologists. In this article, we illustrate 2 cases of unclassical types of adenomyosis; the first one is polypoid adenomyoma (adenomyomatous polypoid), which is classified as a particular form of adenomyosis, with a prevalence of 1.3 % among all the endometrial polyps [7]. Meanwhile, the second case was adenomyosis with a hyper-extensive pattern in T2 weighted imaging, which is known as the 'Swiss cheese pattern' [1].

Case description

Case 1

A female, 36 years old, visited our oncology outpatient clinic due to abdominal pain and abnormal vaginal bleeding. The patient also reported a problem of infertility. The tumor marker investigation revealed negative beta-human chorionic gonadotropin (β-hCG) ($<$ 2.3 mIU/mL). The patient had a history of curettage in 2017 due to a hydatidiform mole. At clinical examination, a uterine enlargement was found that was similar to a 24-week pregnancy, with a solid mass measuring 4 $\times$ 6 cm palpated at the cervix, which seems to originate from the endocervix. Ultrasound examination revealed multiple hypoechoic striations and multiple cystic lesions at the myometrium with hypervascularization inside the myometrium (Fig. 1).

A pelvic MRI showed uterine enlargement measuring roughly 9.6 $\times$ 13.2 $\times$ 15.4 cm, and the junctional zone was diminishing. Unlike typical adenomyosis, there was a diffuse extensive hyperintensity signal in the uterine cavity at the T2-weighted imaging (T2WI) sequence, giving the appearance of a 'fish in net' appearance (Fig. 2). Nonetheless, the right and left ovaries were normal. Contrast agents were also administered at T1WI, showing heterogenous contrast enhancement (Fig. 3).

The patient underwent surgery 3 months after the MRI examination. An enlarged uterus with massive bleeding was revealed during the procedure, prompting a total abdominal hysterectomy and a bilateral salphyngo-oophorectomy. The macroscopic tissue obtained after the surgery was an enlarged uterus measuring 15.5 $\times$ 14 $\times$ 9 cm, with slices of the uterus showing hollow myomatic areas containing 2 pedunculated polyps of the endometrium (Fig. 4). Microscopic sampling of endometrial polyps was performed. A polypoid mass lining with endometrial glands in the proliferation phase was found, with some of the tissue arranged densely and the other having cystic dilatation, giving a Swiss cheese pattern appearance. Among the tissue, endometrial stroma and myometrium were visible (Fig. 5).

Case 2

A 12-year-old girl reported extensive menstrual bleeding with no history of prior pregnancy. The patient was initially suspected of having a trophoblastic tumor due to a high level of β-HCG (serum: 724,153 mIU/mL). A pelvic MRI examination was requested, demonstrating an enlargement of the uterus, measuring approximately 6.1 $\times$ 12.8 $\times$ 17.8 cm, with endometrial thickening and a diminishing junctional zone. A simple cyst was also noted in both ovaries. Multiple hemorrhagic foci were detected inside the uterus, forming a mess-like appearance. Extensive hyperintense T2 signals were also apparent in the myometrium, giving a "Swiss cheese pattern". At diffusion-weighted imaging, there was no restricted diffusion (Fig. 6).

After 5 months of Levonorgestrel therapy, the patient no longer had abnormal menstrual bleeding. An evaluation MRI revealed normal uterine size and shape, along with a simple cyst in both ovaries, which had already decreased in size compared to the previous MRI (Fig. 7).

Discussion

Adenomyosis is a subtype of endometriosis where endometrial tissue proliferates within the myometrium, causing diffuse enlargement of the uterus. Carl von Rokitansky, in 1860,

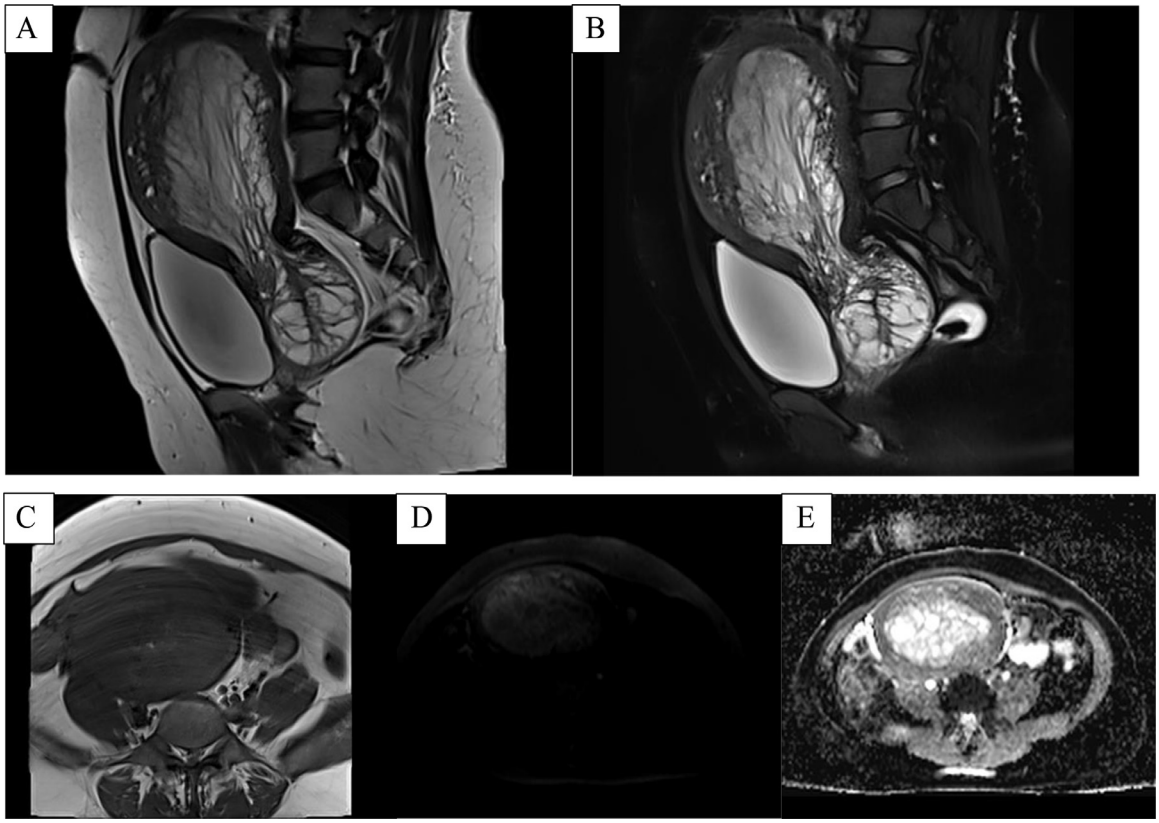

Fig. 2 – T2 weighted sagittal plane (A) and T2 weighted sagittal plane + fat saturation (FS) (B) show the enlargement of the uterine cavity with an extensive hyperintense signal resembling a 'fish in net' appearance. T1 weighted axial plane (C) shows a diffuse low-intensity signal with focal hyperintense inside, suggesting an endometrium gland. Diffusion-weighted imaging (DWI) (D) and apparent diffusion coefficient (ADC) (E) demonstrate a T2 sign with no sign of high-cellular mass.

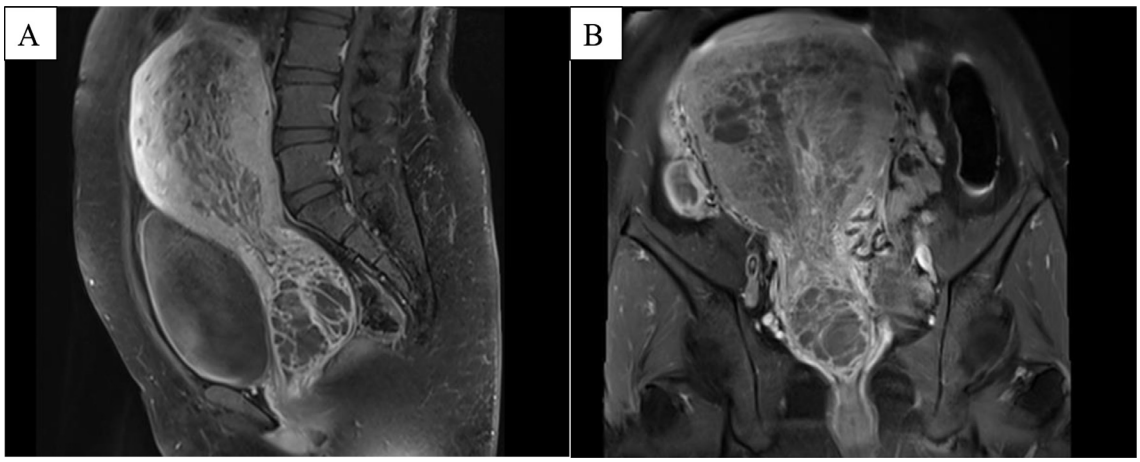

Fig. 3 – T1 weighted with contrast administration at the sagittal plane (A) and T1 weighted with contrast administration at the coronal plane (B) show heterogeneous contrast enhancement at the uterine cavity.

first described adenomyosis as myometrium containing endometrial gland and called it *'cystosarcoma adenoid uterinum'* [5,6].

The accurate demographics of patients with adenomyosis were still unclear due to underreporting. It was described that adenomyosis occurs in 5%-70% of cases, with recent data narrowing it down to 20%-35% [8]. In the American population, the incidence was 1.03%, or 28.9 per 10,000 women per year, with the highest incidence among females aged 41-45 years [2]. A study conducted by Krentel et al. in a German hospital showed that the final diagnosis of adenomyosis ranks first (40%) in patients with hysterectomy [9].

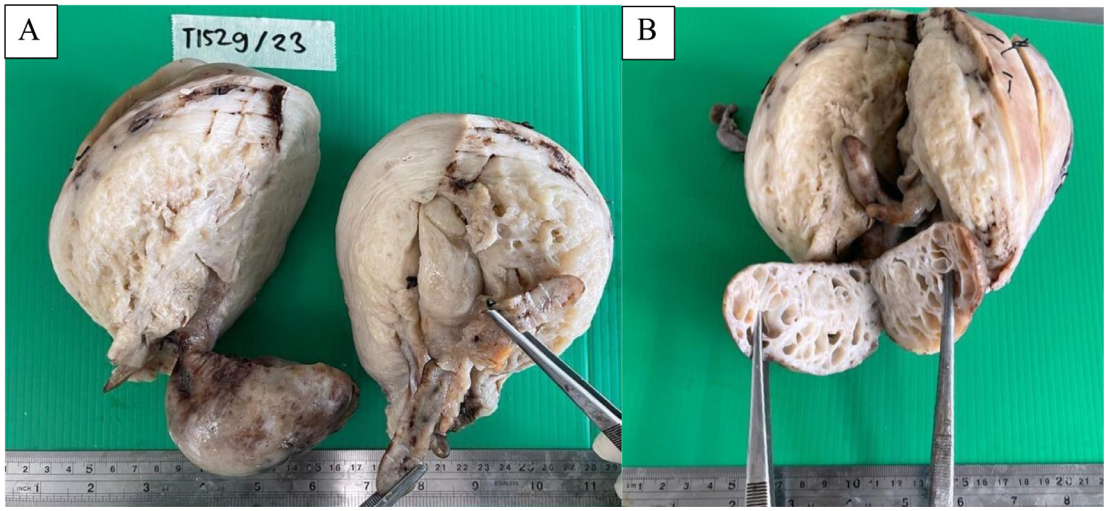

Fig. 4 – Macroscopic appearance of the uterus (A) shows uterus enlargement with 2 pedunculated polyps. The polyp contains a hollow myomatic area (B).

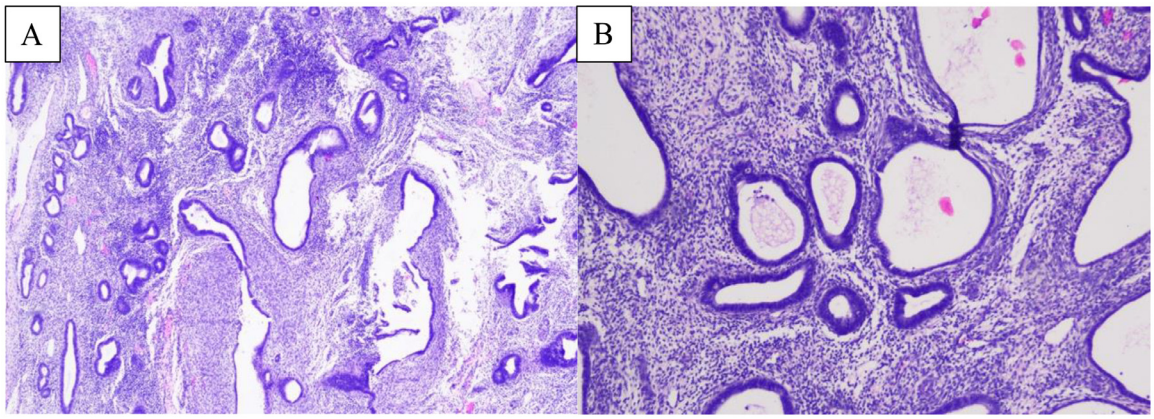

Fig. 5 – Microscopic examination of the uterus (A and B) gives an impression of a 'Swiss chess-like' pattern with a gland in the gland and a gland in the back appearance.

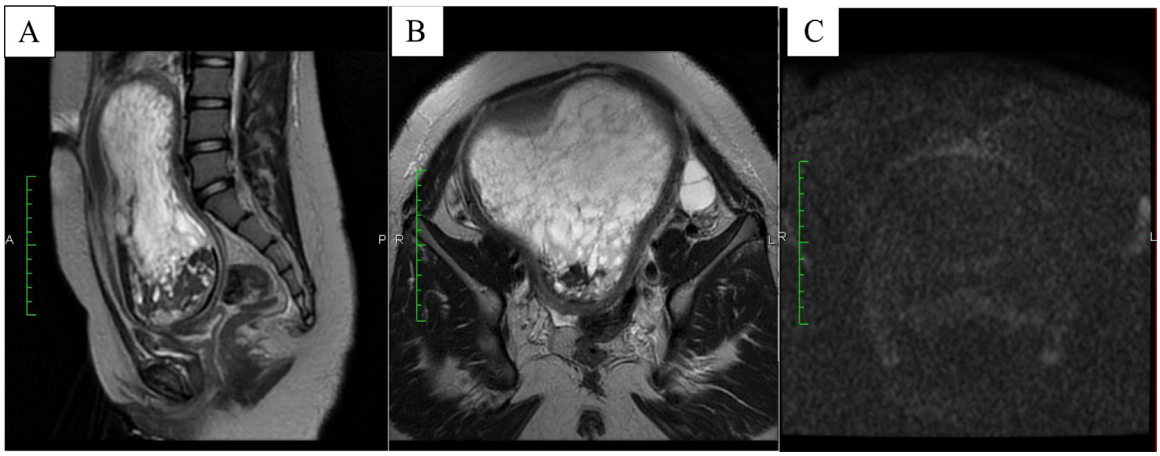

Fig. 6 – (A) Sagittal T2 weighted MRI and (B) coronal T2 weighted MRI demonstrate uterus enlargement with extensive hyperintense T2 signal, show 'Swiss cheese' pattern. Multiple hemorrhagic foci are also noted at the bottom of the uterine cavity. (C) Diffusion-weighted imaging shows no restricted diffusion area.

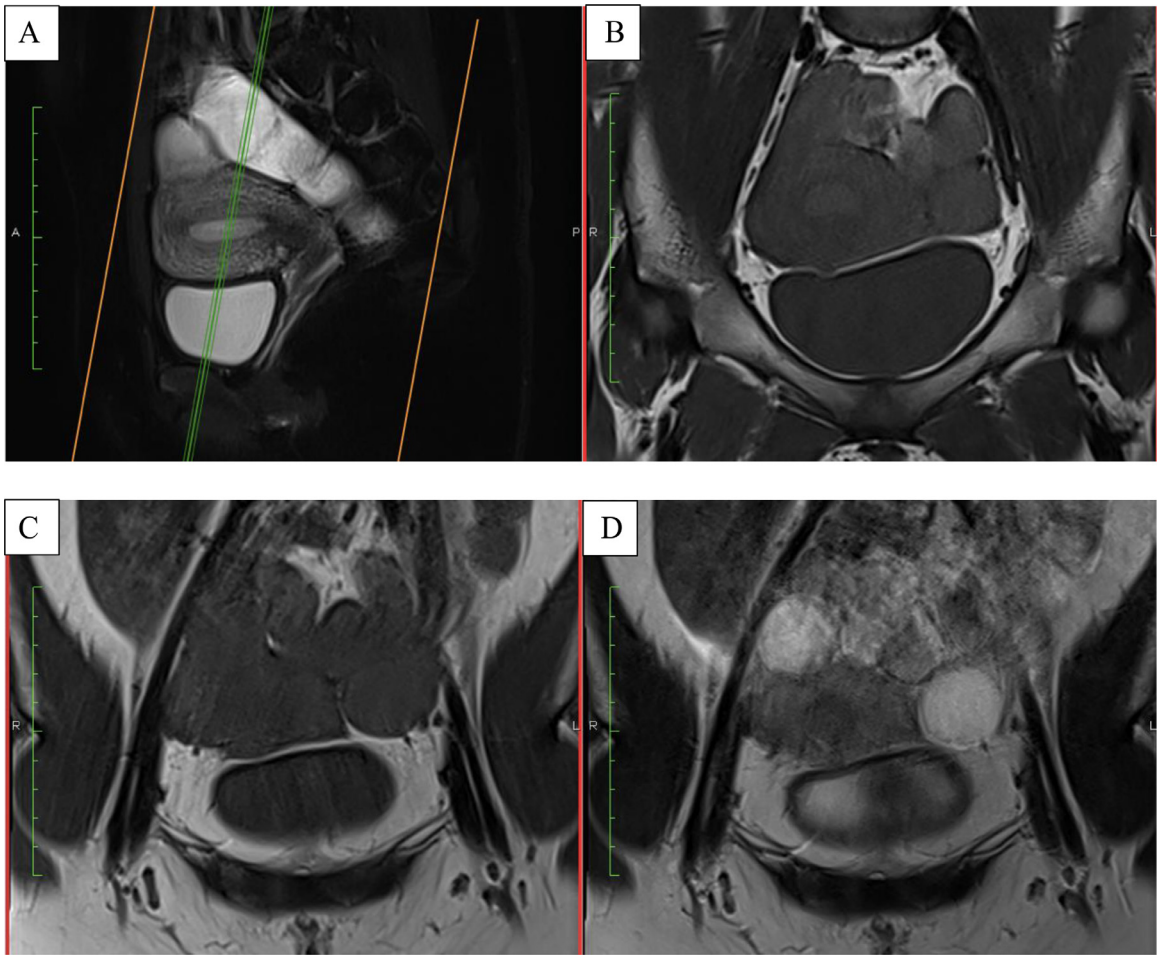

Fig. 7 – (A) Sagittal T2 weighted imaging shows normal features of the uterus, with the normal junctional zone. (B and C) Axial T1 weighted imaging shows no abnormality of the uterus. There is a hyperintense lesion at the right and left ovaries, which, on axial T2 weighted imaging (D), appears as a hyperintense signal, which is concluded to be a simple cyst on the bilateral ovarium.

The pathophysiology of adenomyosis has yet to be elucidated. In some studies, adenomyosis could occur due to 2 conditions. The first condition was the endometrium invasion into the myometrium, and the second cause was the abnormal *de novo* proliferation of endometrial glands in the Mullerian remnant. Hyperplasia and hypertrophy of smooth muscle represented a reactive process of the myometrium due to such invasion [3,4].

There are multiple risk factors for adenomyosis. Women with multiparity have a higher chance of adenomyosis, explained by the invasive nature of trophoblasts, which can invade up to myometrial muscle fibers. The high estrogen hormone during pregnancy supports the growth of endometriosis tissue, which has a high estrogen receptor ratio [5]. The history of invasive uterine procedures is also thought to implicate the elevated risk for adenomyosis due to the injury in the endometrial–myometrial lining. With the hypothesis of endometrial gland invasion to the myometrium, this surgical history increases the risk of adenomyosis [5].

Clinical symptoms in adenomyosis patients may include menstrual pain, bleeding, miscarriage, and infertility [9]. Many explanations have been studied regarding the presence of pain in adenomyosis. One of them is that adenomyosis occurs due to elevated levels of inflammatory factors, including interleukin-1β (IL-1β), corticotropin-releasing hormone (CRH), and urocortin (UCN), which initiate the formation of prostaglandins [10]. Hemorrhage in adenomyosis is due to angiogenesis, high microvascular density, and increased uterine contractility, whereas inflammation, fibrosis, and hypercontractility of myometrium contribute to miscarriage and infertility due to inadequate implantation. In addition, these symptoms vary in each individual and are influenced by genetic and epigenetic factors [10].

Imaging methods that can help confirm adenomyosis include ultrasound and MRI. Ultrasound is the modality of first preference for adenomyosis. Evaluation on ultrasonography corresponds to 3 changes that occur in adenomyosis patients, such as [1] glands and endometrial stromal ectopic in the myometrium, [2] hyperplasia or hypertrophy of myometrial muscle tissue, and [3] hypervascularization of the myometrium [11]. Ultrasound images of endometrial glands and stroma infiltrating the myometrium will give a picture of myometrial

hypoechogenic striation, the presence of a cystic component in the myometrium, and cystic striation. Hyperplasia, or hypertrophy of muscle tissue, is a reactive process of endometrial tissue and will also appear on ultrasound with a picture of myometrial thickening, both focal and diffuse, obscuring the junctional zone, commonly referred to as an enlarged globular uterus. Uterine hypervascularization can also be evaluated with ultrasonography, especially with color Doppler ultrasonography. In adenomyosis, ultrasonography provides an image of the vasculature extending into the lesion, in contrast to leiomyoma, in which the vascular system is at the periphery of the lesion [11].

MRI is an accurate tool for diagnosing adenomyosis. The typical picture of adenomyoma on MRI is the widening of the junctional zone, which is seen with the presence of diffuse hypointense areas on the T2W1 sequence, indicating hyperplasia in the myometrium, and thickening of the uterus with thickening exceeding around 12 cm should be indicative of adenomyosis. The presence of hyperintense foci on T2WI indicates ectopic endometrial tissue inside. Whereas leiomyoma usually shows better-demarcated lesion than adenomyosis, with the presence of peripheral large vessel [12]. If the endometrial tissue is bleeding, hyperintensity will also be seen in the T1WI sequences. Although the sensitivity is only 47.5%, it has a high positive predictive value (95%) [12].

In this case report, we presented the unusual features of 2 cases of adenomyosis, which can cause pitfalls in diagnosis. The first type showed 'Fish in a net pattern,' which is described as a hyperintensity signal of T2 representing the polyp (fish) in the smooth muscle tissue (net), which is mediated by the endometrial gland [1], while the second was diffuse adenomyosis with dilatation of smooth muscle and proliferation of endometrial gland that has been described by Agostinho et al. as the 'Swiss cheese-like appearance' [12]. These type2 of adenomyosis, hyperintensity signal T2 is more extensive than classical adenomyosis. It was glandular tissue exhibiting cystic dilatation, no extension along the arteries or ligaments, and progressive centripetal enhancement with contrast deficiencies in multicystic locations [1]. Those extensive T2 signals, in some cases, usually lead to malignancy. Thus, endometrial carcinoma, low-grade endometrial stromal sarcoma, adenosarcoma, and leiomyosarcoma, and invading adenomyosis are included in the radiological differential diagnosis of this proliferative uterine adenomyosis [1]. In the case of endometrial malignancy, we should find a mass with distention of the endometrial cavity, and unlike adenomyosis, there is no hypointense area at the myometrium, which indicates hyperplasia of the myometrium [13]. While cystic degeneration or necrosis can occasionally be seen, the high signal lesions on T2-weighted images in low-grade endometrial stromal sarcoma are sarcomatous components and show clear contrast enhancement, diffusion restriction, and occasionally extension of the tumor along the vessels or ligaments [1]. Adenosarcoma presents as a multiseptated cystic mass with solid regions of poor signal intensity on T2-weighted imaging, it never been reported mesh like appearance for this malignancy. And for the leomyosarcoma as a malignancy at the uterus, a heterogeneous mass with areas of necrosis and bleeding is seen [1].

In the first patient, based on clinical status and normal β-HCG level, we suggested the diagnosis of polypoid adenomyoma with a 'Fish in a net' pattern. Hysterectomy was the mainstay of treatment of this disease. The result for macroscopic appearance showed an enlargement of the uterus with a myomatic hollow area in the myometrium. We also found a polyp with multiple septa containing endometrial glands on the microscopic examination. The endometrial glands at the proliferation phase were also dilated, yielding the appearance of an adenomyosis.

A different treatment was done for the second patient. The patient was previously suspected of having a trophoblastic tumor due to a high level of β-HCG. However, based on MRI, it was suggested that the patient had adenomyosis. We opted for hormonal therapy, namely Levonorgestrel, for this patient. Levonorgestrel, a combination of progesterone and estrogen, is becoming a therapy choice for adenomyosis. Usually, Levonorgestrel, as a safe and effective contraceptive, comes in an intrauterine preparation as Levonorgestrel-releasing intrauterine system [14]. However, Levonorgestrel could not be given via the intrauterine route due to the virgin status of the patient; therefore, oral Levonorgestrel was selected in this case. After 5 months of Levonorgestrel therapy, the patient's symptoms were resolved, and the MRI examination showed a normal uterus appearance with a reduced bilateral ovarian cyst. After successful treatment with Levonorgestrel, it could be concluded that the patient's diagnosis was adenomyosis instead of a trophoblastic tumor, as suspected before.

Conclusion

Adenomyosis is a reproduction problem in women. There are usually disturbing symptoms, such as abnormal uterine bleeding and menstrual pain. Different grades of endometrial invasion give different imaging appearances, leading to a diagnostic pitfall. Diagnostic findings with precise interpretation will help clinicians determine whether the patient should have surgery on conservative therapy.

Patient consent

Written informed consent was obtained from the patient for the publication of this case report.

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