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Case Report

Adenomyosis-a rare cause of chronic lumbago in adolescents

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ABSTRACT

Adenomyosis is a benign condition of the uterus that was previously diagnosed through histology after hysterectomy. Specifically, ectopic endometrial glands and stroma were seen at a minimum depth of 2.5 mm below the Endo myometrial junction, along with hypertrophic and hyperplastic surrounding myometrium. Adolescent adenomyosis is not known to occur frequently. Furthermore, in extremely young individuals, the classic diagnostic indicators of adenomyosis may not be present during clinical, sonographic, or radiologic testing. Although cystic form of adenomyosis is more common in the younger age group, diffuse adenomyosis should always be considered in refractory pelvic pain. Ultrasound and MRI done together has improved sensitivity for the diagnosis of adenomyosis. Dienogest, the millennial molecule, not only serves its purpose in endometriosis but also one of the first line drugs in the management of adenomyosis, particularly the diffuse forms.

Keywords: Adenomyosis, Millennial molecule, Endo myometrial junction

INTRODUCTION

Adenomyosis is a benign condition of the uterus that was previously diagnosed through histology after hysterectomy. Specifically, ectopic endometrial glands and stroma were seen at a minimum depth of 2.5 mm below endo myometrial junction, along with hypertrophic and hyperplastic surrounding myometrium.[1] Non-invasive methods for detecting adenomyosis has been made possible with advancements in imaging techniques, enabling early diagnosis of the condition. Both TVUS and MRI are increasingly used to detect the presence of adenomyotic lesions and to determine the best course of treatment. It has been reported that both technologies have similar sensitivities and specificities in the detection of adenomyosis.

Women with adenomyosis account for 20% of all cases, however perimenopausal and multiparous women are most likely to have it.[3] Adolescent adenomyosis is not known to occur frequently. Furthermore, in extremely young individuals, the classic diagnostic indicators of adenomyosis may not be present during clinical, sonographic/radiologic testing. For clinicians, this renders the diagnosis essentially invisible. Clinical characteristics, blood investigations, ultrasound/MRI findings and therapy choices for a young girl with diffuse adenomyosis, will all be covered in this article.

CASE REPORT

A 20 year old single woman presented to us with complaints of severe low back pain and right leg pain from day 6 to day 29 of her cycle for the past 5 years, along with dysuria and dyschezia. Her pain disappears just before the start of menstruation and is completely pain free from day 1 to day 5 of her cycle, while she is menstruating. Her cycles are otherwise regular, with moderate flow, and she has no dysmenorrhea. She underwent multiple evaluations at various hospitals, where she was reassured and managed conservatively with analgesics. Due to the patient's ongoing discomfort, an outside hospital performed an MRI pelvis (Figure 1), which revealed diffuse adenomyosis of the uterus. At the same hospital, she received treatment with six doses of GnRH analogues. She was asymptomatic while receiving GnRH medication, but she relapsed after stopping the last dose. Thus, she sought additional

evaluation at our facility. A positive family history of endometriosis in her mother was elicited. Per rectal examination, revealed a retroverted, bulky uterus with cervical motion tenderness.

Her blood tests showed that her liver function and renal function tests were within normal limit. She had a hemoglobin level of 12 g/dl, a total count of 6680 cells/mm3, CA 125-6.1 U/ml, and CA19.9-30.5 U/ml. Trans-abdominal 2D USG (Figure 2) performed by an experienced radiologist at our hospital showed a bulky uterus measuring 9.6×5.7×6.1 cm with ill-defined heterogenous echotexture of anterior and posterior myometrium of uterus. Endometrial thickness was 5.5 mm. The right ovary was normal and a small functional cyst 3x2cm was noted in the left ovary. No mass or fluid collection in the adnexa.

Her MRI pelvis revealed a retroverted, bulky uterus measuring 98×52×65 mm with normal shape and contour. Junctional myometrium asymmetrically thickened and appears coarsely hypointense in T1W and iso-hyperintense in T2W/STIR images with multiple small hypointense foci. Serosal surface intact. Endometrial thickness 7 mm. Both ovaries normal in size and signal intensity for the age. Rest of the pelvic and abdominal organs appeared normal.

Patient and her attenders were counselled in detail regarding the pathophysiology of adenomyosis and the need for hormonal therapy over surgical modality. Diffuse adenomyosis will be associated with difficult planes of dissection and the preferred management would be ovarian suppression with medications was explained to them clearly. As patient has already taken 6 doses of GnRH analogues and progesterone with good response to the same, low dose dienogest 2 mg once daily was started and was asked to review after 3 months for reassessment of her symptoms. Dienogest tablet dosage was intended to be doubled if symptoms persisted or became severe.

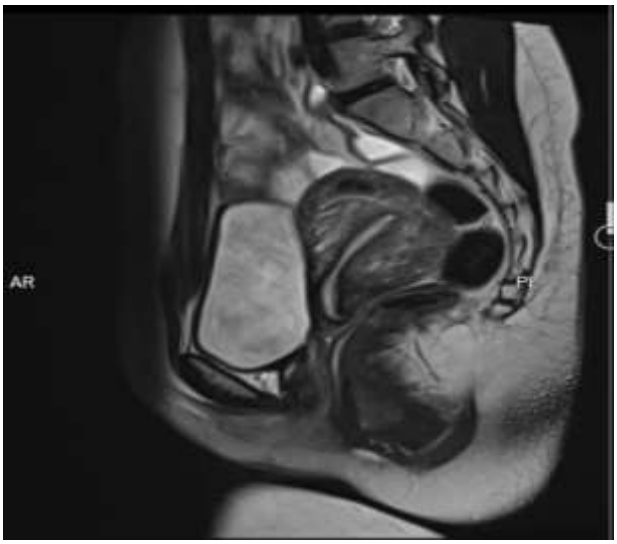

Figure 1: MRI pelvis.

Figure 2: Transabdominal ultrasound of pelvis.

DISCUSSION

Numerous theories have been put up to explain the exact pathogenesis of adenomyosis. The disease, however, is thought to occur through the invagination of the endometrium basalis into the myometrium through an altered or absent junctional zone JZ, which is the most widely accepted theory. Another idea proposes that the primary event in the process of initiation of adenomyosis is uterine auto-traumatization and the mechanism of tissue injury and repair (TIAR). Peristaltic myometrial contractions encourage recurrent bouts of auto-traumatisation, harming the JZ. As a result, basal endometrial fragments migrate into the myometrium by TIAR mechanism responding to an increase in intrauterine pressure brought on by hypercontractility. Embryonic or adult stem cells that can undergo metaplasia into the myometrium as a de-novo process is another pathogenic explanation of adenomyosis.

More research over the past ten years has shown the pathogenic mediators of adenomyosis to include sex steroid hormone receptors, inflammatory chemicals, extracellular matrix enzymes, growth factors and neuroangiogenic factors.[5]

Adenomyosis is a benign gynecological condition usually affecting the multiparous woman. The incidence of adenomyosis in adolescent population is unknown, as only few cases have been reported to date.[6] Although adenomyosis appears to be extremely uncommon before the age of 20, reports of the cystic variant primarily involve young women. Here, in our case, a 20-year-old woman who has undergone multiple evaluations for severe lumbago over the past five years was detected to have diffuse adenomyosis.

There is no pathognomonic symptomatology that can be used to diagnose adenomyosis; instead, the condition is accompanied by a wide range of symptoms that may or

may not be directly related to it. Abnormal uterine bleeding (AUB) reduced reproductive capacity and pelvic discomfort (in the forms of dysmenorrhea, dyspareunia, and persistent pelvic pain) are common symptoms. About 30% of women with adenomyosis are asymptomatic.[5] Our case is distinct from other case reports because the woman complained of significant right lumbago and right leg discomfort without experiencing dysmenorrhea or heavy menstrual bleeding. Adenomyosis may have a hereditary component because first-degree relatives had a 6.9 percent relative risk of developing endometriosis.[7] In this article, the patients aunt is a proven case of endometriosis on medical management.

Transvaginal sonography should be the first-line modality for imaging of suspected adenomyosis. Based on ultrasound findings, adenomyosis can be classified as diffuse, focal (inner and outer myometrium) and Adenomyomas. Furthermore, diffuse adenomyosis can be classified based on the inner and outer myometrial involvement-a modification of MUSA statement. Diffuse adenomyosis of the outer myometrium can be graded based on the thickness of the uterine wall and the number of uterine walls involved (anterior, posterior, right or left lateral wall). Diffuse adenomyosis of the inner myometrium can be graded based on the thickness of the JZ and length of the infiltrated JZ tract. Focal adenomyosis of the inner and outer myometrium can be graded based the largest diameter of focal lesion and number of foci. Adenomyoma can be further graded based on size and number.[8] 2D-TVS features associated with diffuse adenomyosis were predefined as: (1) heterogeneous myometrium; (2) hypoechoic striation in the myometrium; (3) myometrial anechoic lacunae or cysts; (4) asymmetrical myometrial thickening of the uterine walls with the presence of straight vessels, extending into the hypertrophic myometrium, on power Doppler examination.[9] Hyperechoic regions, uterine wall asymmetry, and intra-myometrial cystic areas with some type of JZ changes were the most often observed ultrasonography findings. and when it comes to the layer of uterus - the posterior outer myometrium was commonly involved as per Caterina et al research article. In our patient, an ill-defined heterogenous echotexture of both the anterior and posterior myometrium was noticed on trans-abdominal ultrasound hence proceeded with MRI.

If there is an inconclusive sonographic evaluation of adenomyosis or suspicion of substantial concurrent pelvic disease, clinicians should seek magnetic resonance imaging. The changes in the junctional zone may precede or be the first signs of adenomyosis. The heterotopic endometrial tissue may be seen as small foci of increased high signal intensity in the junctional zone on MRI.[2] The specified criteria for the diagnosis of adenomyosis by MRI include focal regions in the myometrium that are poorly defined on T2-weighted images, uniformity of the junctional zone, and thickness of the junctional zone greater than 12 mm.[10] In our case, the junctional myometrium was asymmetrically thickened and coarsely hypointense in T1W and iso-hyperintense in T2W/STIR images with multiple small hypointense foci and intact serosa.

The primary therapeutic objectives in adolescents are symptom reduction and future fertility preservation. The preferred treatment is central inhibition of hormone production with GnRH agonists or continuous oral contraceptives. Those with distinct lesions may try laparoscopic removal.[10] The first line of treatment for adenomyosis related pain and heavy menstrual bleeding includes oral contraceptives, levonorgestrel releasing intra-uterine system and dienogest.[11] In this article, the girl had already received 6 doses of injection lupride 3.75 mg intramuscularly coupled with oral progesterone. Therefore, we decided to treat her with tablet Dienogest 2 mg once daily for three months with frequent liver function monitoring. The patient was asymptomatic on her third month follow up. As she was responding well to dienogest, we decided to prolong the treatment with dienogest after ensuring her LFT was within normal limits.

CONCLUSION

Adenomyosis although uncommon in adolescent population can present with vague symptoms such as chronic lumbago and right leg pain with no associated heavy menstrual bleeding or dysmenorrhea during cycle. Although cystic form of adenomyosis is more common in the younger age group, diffuse adenomyosis should always be considered in refractory pelvic pain. Ultrasound and MRI done together has improved sensitivity for the diagnosis of adenomyosis. Dienogest, the millennial molecule, not only serves its purpose in endometriosis but also one of the first line drugs in the management of adenomyosis, particularly the diffuse forms.

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