Original Study

Regression of Adenomyosis on Magnetic Resonance Imaging after a Course of Hormonal Suppression in Adolescents: A Case Series

Roshanak Mansouri MD *, Xiomara M. Santos MD, Jennifer L. Bercaw-Pratt MD, Jennifer E. Dietrich MD, MSc

Division of Pediatric and Adolescent Gynecology, Department of Obstetrics and Gynecology, Baylor College of Medicine, Houston, Texas

A B S T R A C T

Study Objective: To demonstrate that adenomyosis is a rare cause of dysmenorrhea or chronic pelvic pain (CPP) in the adolescent population that can be identified with magnetic resonance imaging (MRI) and to report resolution of adenomyosis by MRI after a course of hormonal suppression in 4 adolescents.
Design, Setting, and Participants: Retrospective case series of 4 adolescents with adenomyosis on pelvic MRI at Texas Children's Hospital.
Interventions: Continuous oral contraceptive (COC) therapy or leuprolide acetate.
Main Outcome Measures: Lesions on pelvic MRI after treatment.
Methods: We reviewed medical records of 4 adolescents with CPP and adenomyosis on T2-weighted pelvic MRI. All patients had initial diagnostic pelvic MRI and then definitive hormonal intervention. Repeat imaging was obtained after a symptom-free interval.
Results: Patient ages ranged from 12 to 16 years. One patient had resolution of symptoms with COC therapy. MRI performed 3 years later showed no adenomyosis. Three patients failed COC therapy. All were symptomatically improved after therapy with a gonadotropin-releasing hormone agonist. Follow-up MRI performed at intervals between 6 months and 3 years showed resolution of adenomyosis.
Conclusion: MRI can raise suspicion for the diagnosis of adenomyosis in adolescents with refractory CPP. Subsequent MRI can show regression of lesions after symptom resolution with hormonal therapy.
Key Words: Adenomyosis, Adolescent, Chronic pelvic pain, Dysmenorrhea, Pelvic MRI

Introduction

While adenomyosis is commonly reported in perimenopausal or multiparous women, the diagnosis is rare in the adolescent population. Available literature is generally in the form of case reports and small series.[1–9] In this population, adenomyosis or adenomyotic cysts have been discovered when patients have chronic pelvic pain or severe dysmenorrhea that is refractory to traditional first-line treatments with nonsteroidal anti-inflammatory drugs (NSAIDs) or continuous oral contraceptive (COC) therapy.[1,10]

Although historically adenomyosis has been a surgical diagnosis, transvaginal ultrasound (TVUS) and magnetic resonance imaging (MRI) allow for noninvasive diagnosis of these lesions.[11–15] Both are considered to be useful, and while there are diagnostic challenges with both methods, diagnosis with TVUS is more dependent on the skill of the radiologist and may be difficult to reproduce across institutions.[11] Practically, in the adolescent population, TVUS may also be more difficult to attain, as transvaginal examinations are generally avoided prior to the onset of sexual activity. MRI has the benefit of being less invasive, may be superior in accuracy in the presence of other lesions such as uterine fibroids, and in prospective studies has been shown to have a sensitivity of 46% to 88% and specificity of 67% to 99% for the diagnosis of adenomyosis. Positive predictive value is reported between 58% and 94%.[15] The defined criteria for the diagnosis of adenomyosis by MRI include uniformity of the junctional zone, thickness of the junctional zone greater than 12 mm, and focal areas in the myometrium that are not well demarcated on T2-weighted images.

In older populations, especially those who have completed childbearing, therapy commonly includes surgery. Definitive surgery by hysterectomy is common, but in recent years, fertility-sparing procedures such as local excision, hysteroscopic ablation and excision, laparoscopic electrocoagulation, and minimally invasive approaches such as uterine artery embolization and endometrial ablation have been attempted and reported with some success.[16]

In adult women with adenomyosis, medical therapy has been extrapolated from the treatment of endometriosis, and some medical therapies have reported success. The levonorgestrel-releasing intrauterine system has been shown to improve dysmenorrhea and heavy menstrual bleeding associated with adenomyosis.[17,18] In one study, patients with adenomyosis by MRI were given leuprolide acetate for 6 months and found to have an improvement in adenomyotic lesions, with focal lesions responding better than diffuse adenomyosis.[14] Endometriosis and adenomyosis lesions have also shown microscopic regression in the form of apoptosis and decreased tissue inflammation

The authors indicate no conflicts of interest.

* *Address correspondence to:* Roshanak Mansouri, MD, 6651 Main St, Suite 1020, Houston, TX 77030; Phone: +1 (832) 826-7464; fax: +1 (832) 825-9349
E-mail address: roshi.mansouri@gmail.com (R. Mansouri).

1083-3188/$ - see front matter
http://dx.doi.org/10.1016/j.jpag.2014.12.009

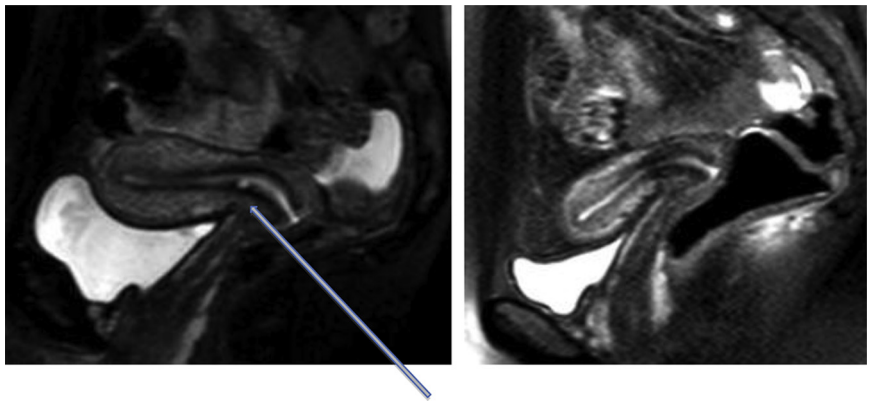

Fig. 1. Case 1 MRI before (left) and after (right) treatment, with area suspicious for adenoymosis (arrow).

and angiogenesis in the short term with gonadotropin-releasing hormone (GnRH) agonist therapy.[19] The aromatase inhibitor letrozole has been shown to have positive effects on patient symptoms and adenomyoma size.[20] There are limited data on long-term follow-up of patients who have received medical management, their symptom resolution, and the persistence or resolution of previously seen lesions.

In the adolescent population, primary goals of therapy are the relief of symptoms and preservation of future fertility. In adolescents, medical therapy has been extrapolated from treatment of adults with adenomyosis or endometriosis, with central suppression of hormone production, in the form of continuous oral contraceptives or GnRH agonists.[1,10,21] In adolescents with discrete lesions, case reports show that adenomyosis has been managed by laparoscopy for removal of adenomyotic cysts and adenomyotic uterine remnants.[2–9]

In this study, we report findings consistent with adenomyosis by MRI in 4 adolescents, with subsequent resolution of both symptoms and imaging findings after a course of hormonal suppression therapy.

Materials and Methods

Medical records of 4 patients with the diagnosis of adenomyosis were identified and reviewed. All patients were followed in the Texas Children's Hospital Pediatric and Adolescent Gynecology Clinic.

Data were collected on patients' presenting symptoms, therapies before diagnosis of adenomyotic lesions by MRI, course of therapy leading to resolution of symptoms, and findings on repeat imaging after a symptom-free interval. Where applicable, operative and pathology reports were also reviewed.

This study was reviewed by the institutional review board and deemed exempt.

Results

Case 1

A 15-year-old girl presented to clinic complaining of constant severe pelvic pain and dysmenorrhea. On initial examination, a musculoskeletal component was suspected, and there was some adnexal tenderness. The intermenstrual pain improved with COC therapy, mefenamic acid, and acetaminophen/propoxyphene, but her dysmenorrhea remained severe. Pelvic MRI showed foci of increased signal intensity in the endometrium with focal extension into the myometrium (Fig. 1). Therapy with leuprolide acetate (3.75 mg intramuscularly [IM] monthly) was initiated with normalization of physical examination findings and resolution of symptoms. Add-back therapy with norethindrone (0.35 mg daily) was started after 6 months. A repeat MRI 1 year after the initiation of leuprolide acetate showed resolution of endometrial lesions (Fig. 1). After 1 year, the patient was transitioned to combined cyclic oral contraceptives and continued to be symptom free.

Case 2

A 12-year-old girl presented complaining of acute pelvic, lower back, sacral, and hip pain that began with a menstrual cycle but persisted, causing her to miss 3 weeks of school. She had no relief with NSAIDs, acetaminophen/hydrocodone, metaxalone, or COC therapy. Menses became dysfunctional with prolonged heavy bleeding. The uterus was enlarged on ultrasound. Diagnostic laparoscopy showed an enlarged uterus with retrograde menstrual flow but no endometriotic implants. Pelvic MRI showed diffuse mild thickening of the junctional zone with focal thickening (12 mm) in the anterior corpus, suggestive of adenomyosis (Fig. 2). Therapy with leuprolide acetate (3.75 mg IM monthly) was initiated with relief of pain symptoms. Norgestrel/ethinyl estradiol (0.3 mg/30 μg daily) was used for 3 weeks during the flare. Norethindrone acetate/ethinyl estradiol (0.5 mg/2.5 μg daily) was added within 3 months for hormonal add-back due to hypoestrogenic vasomotor symptoms. Pelvic MRI was repeated after a 2-year treatment interval showing resolution of previous findings.

Case 3

A 17-year-old girl with a family history of endometriosis and adenomyosis presented with 5 months of intermittent pelvic pain not associated with menses. Pelvic ultrasound