Case Report

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Is this an adenocarcinoma? A case report of extragenital endometriosis with hydroureteronephrosis

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Introduction and importance: Endometriosis is a prevalent condition within the female reproductive age group, but its presentation and diagnosis still pose a challenge as it mimics other diseases and affects multiple organ systems.
Case presentation: A 24-year-old nulliparous female presented with complaints of menorrhagia, lower abdominal pain, post-coital bleeding, and significant weight loss for 7 months.
Clinical discussion: The case highlights the challenge of diagnosing endometriosis due to its ability to mimic other conditions, such as carcinoma cervix and rectum. The presence of elevated Cancer Antigen-125 (CA-125), typically associated with malignancy, underscores the need for a comprehensive evaluation to differentiate between endometriosis and other pathologies. Furthermore, the involvement of multiple organ systems emphasizes the systemic nature of endometriosis and the importance of considering it in the differential diagnosis of pelvic masses and symptoms in young women.
Conclusion: Endometriosis, although a common disease among females of reproductive age its variation in presentation causes significant misdiagnosis and undertreatment. Multi-modal diagnosis and early treatment are necessary for proper outcomes.

Keywords: adenocarcinoma, case report, endometriosis, hydroureteronephrosis

Introduction

Endometriosis is characterized by the presence of endometrial glands and stroma outside the uterine cavity and walls and is usually a benign condition[1]. Upon penetration of the intestinal walls by endometrial-like-glands and stroma reaching at least the subserous fat tissue or adjacent sub-serosal plexus, the condition is appropriately referred to as "bowel endometriosis"[2]. An estimated 3.8% and 37% of the patients with a diagnosis of endometriosis have bowel involvement with frequently affected regions being the sigmoid colon (over 65% of the cases), followed by the rectum, the ileum, the appendix, and the cecum[2]. Despite being a commonly prevalent condition, endometriosis has an extremely variable presentation, which is particularly true for extragenital endometriosis, making it difficult to recognize, diagnose, and treat[3]. Here, we present a case of a 24-year-old female with abdominal endometriosis affecting multi organs and mimicking infiltrating cancer invading the cervix, vagina, and rectum on Magnetic Resonance Imaging (MRI) and colonoscopy and causing moderate hydroureteronephrosis.

HIGHLIGHTS

- Despite its prevalence, endometriosis presents with highly variable symptoms, posing challenges in recognition and diagnosis.
- Symptoms of the condition can be non-specific despite the involvement of multiple organ systems, complicating early identification.
- Ureteric endometriosis, though rare, represents a serious complication with the potential for irreversible renal impairment.
- Effective treatment of endometriosis requires a multidisciplinary approach tailored to the extent of the disease.
- Timely diagnosis and intervention are crucial, necessitating thorough clinical, radiological, and pathological assessments in reproductive-age females presenting with severe abdominal pain and genitourinary symptoms.
- This case emphasizes the need for comprehensive diagnostic approaches and increased awareness among healthcare professionals to recognize and manage endometriosis effectively.

Case presentation

The patient is a 24-year-old nulliparous female, married for 3 years, who presented with 7 months of heavy menstrual bleeding, lower abdominal pain, post-coital bleeding, and significant weight loss (12% in the last 6 months). On per vaginal examination, multiple

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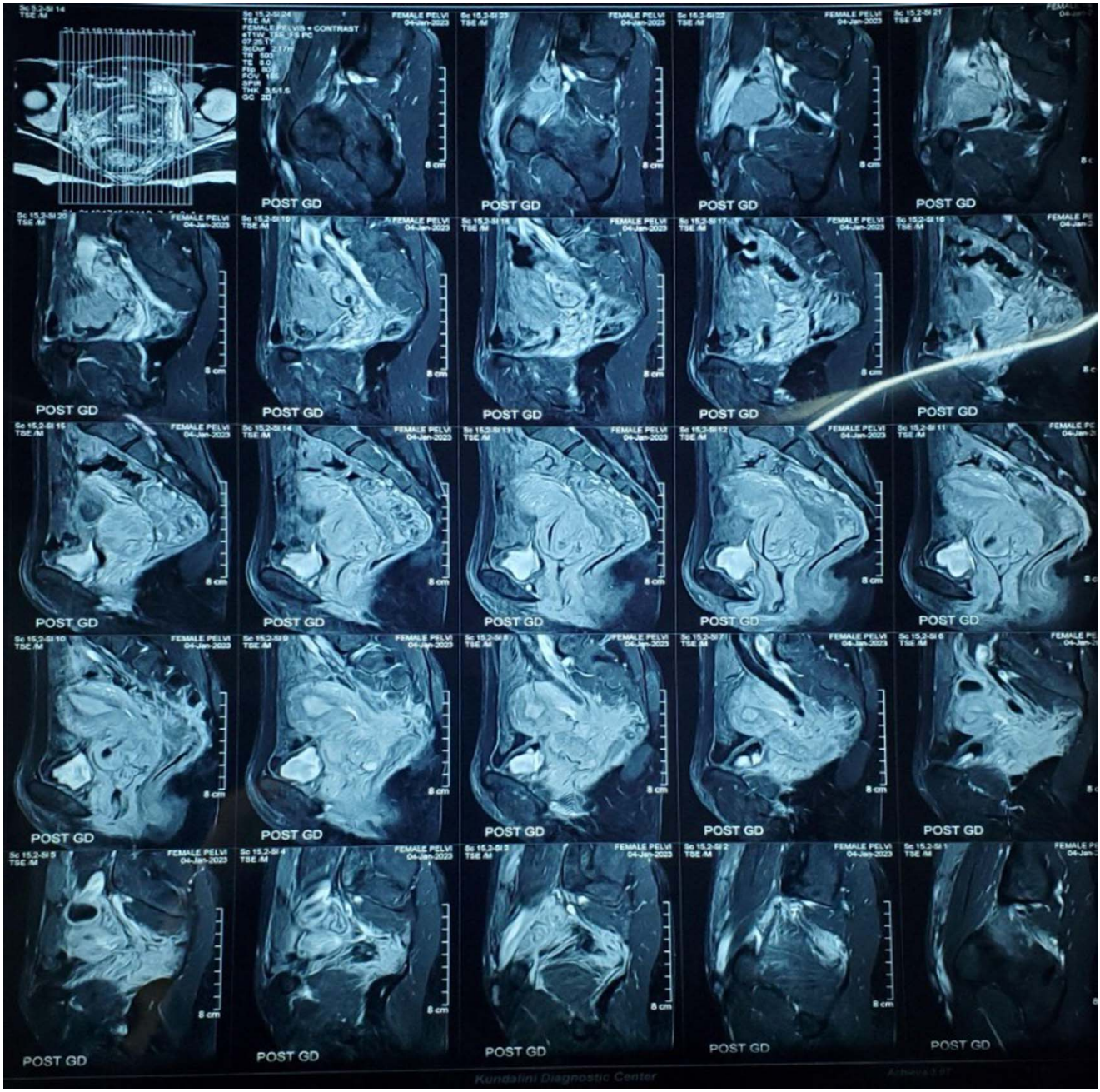

Figure 1. MRI of abdomen and pelvis saggital view. MRI indicate Magnetic Resonance Imaging.

fungating fingerlike projections were arising from the posterior fornix and posterior upper rugae. She underwent an Ultrasonography of the abdomen and pelvis, which showed a bulky cervix with irregularly thickened endometrium with polypoid lesion and fluid-filled endometrial cavity at the lower uterine segment that was suggestive of cervical carcinoma. MRI of the pelvis plus screening of the upper abdomen showed a large heterogeneously enhancing ill-defined mass in the left side of the pelvis infiltrating into the cervix, vagina, rectum and left distal ureter with moderate proximal hydroureteronephrosis (Fig. 1, Fig. 2).

Colonoscopy was done, which showed an ulcer-proliferative growth in the rectum ~8 cm from the anal verge, and an impression of rectal carcinoma was made (Fig. 3), multiple biopsies were taken from the site, which showed chronic active colitis with reparative changes with no evidence of malignancy seen in the submitted biopsy. Tumor markers were also sent where CA 19-9(14.29 U/ml), CEA (1.14 ng/ml) levels were within normal ranges while CA-125 (109.50 U/ml) was elevated (reference range- < 35 U/ml).

Examination under anesthesia and Cervical biopsy were also done showing a cervical polyp with squamous metaplasia with acute on chronic cervicitis.

Repeated colonic biopsy demonstrated focal low-grade adenoma with a focus of endometriotic changes. Further, immunohistochemical staining proved to be positive for CD10 and CK7, which solidified the diagnosis of endometriosis. The patient was treated conservatively with DJ stenting for Hydroureteronephrosis and

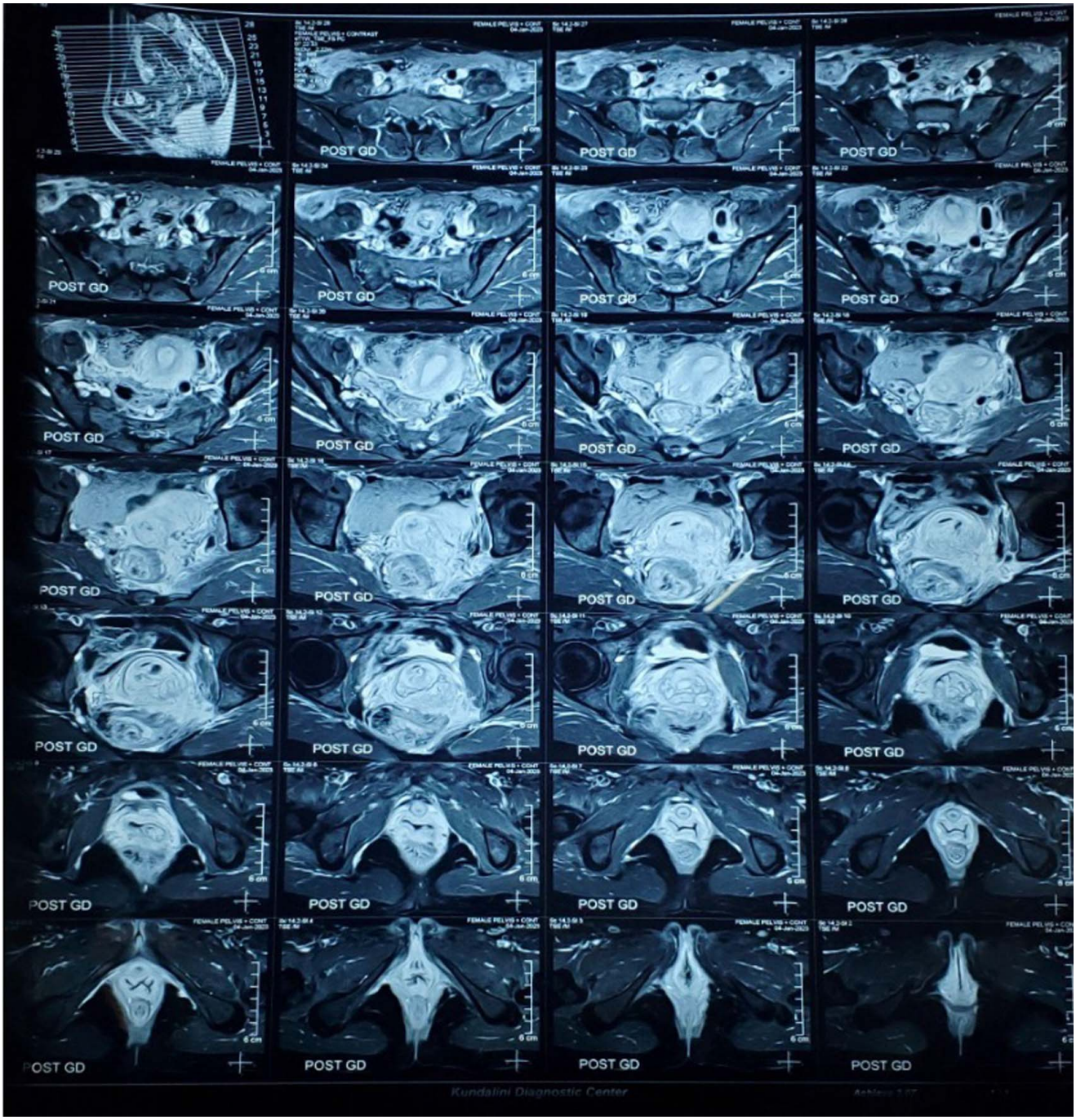

Figure 2. MRI of abdomen and pelvis coronal view. MRI indicate Magnetic Resonance Imaging.

monthly injections of 3.75 mg of Leuprolide intramuscularly with add-back therapy with low-dose combined OCPs for regression of the size of the lesion. She is under monthly follow-up and evaluation of the lesion in terms of changes in size and extent so that she can undergo surgical interventions in the future.

Discussion

Multiple theories regarding the pathogenesis of endometriosis have been put forward regarding the origin of the disease process, with the theory proposed by Samson[4], fitting our case the best. It describes the retrograde flow of the menstrual blood and tissue through the fallopian tube and how the rectosigmoid on the left acts as an anatomical shelter, potentially shielding endometriotic cells from being cleared by macrophages or the peritoneal circulation. This promotes endometriotic cells' peritoneal adhesion and subsequent deep penetration[5]. Endometriotic lesions usually occur in the peritoneum and pelvic organs[6] while ectopic lesions also occasionally occur in other parts of the body, such as the kidney, bladder, lungs, and even the brain[7]. Additional case

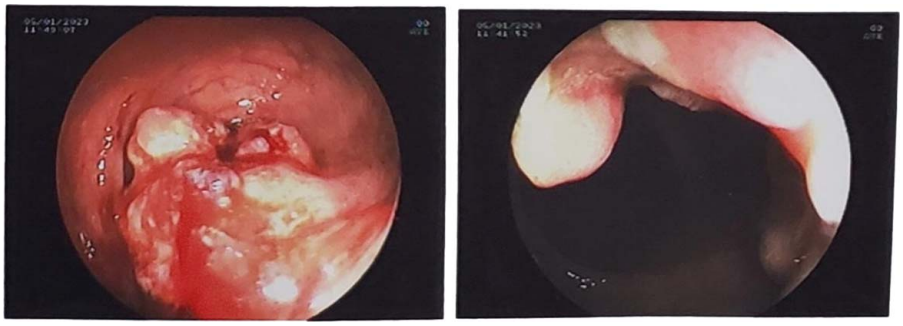

Figure 3. Colonoscopy picture showing semi circumferential ulceroproliferative growth.

studies also detail ectopic or deep endometriosis affecting a single organ that is rectum, ureter, kidneys, or ovaries. However, in our case, there is extensive involvement of various organ systems including the ureter, lower third of the vagina, cervix, and rectum. Despite the involvement of various organ systems, the presenting symptoms were non-specific and only pertinent to uterine pathology, namely menorrhagia and post-coital bleeding.

It is estimated that 5–15% of women have some degree of endometriosis with the average patient being in her 30s, nulliparous, and infertile[1]. In additional studies, the researchers evaluated 44 cases of intestinal endometriosis involving patients with ages ranging from 28 to 56 (mean age, 44 years)[5]. The patient, however, does not fall within the aforementioned demographic range as she is 24 years old.

Endometriosis of the intestinal tract can clinically and pathologically resemble several diseases[2]. 3–37% of female patients have endometriosis affecting the gastrointestinal tract, specifically in the sigmoid colon, followed by the rectum, ileum, appendix, and cecum[2]. Although a small percentage of women may not exhibit any symptoms, the majority experience various non-specific symptoms, namely abdominal pain, diarrhea, constipation, and abdominal bloating, and cause a diagnostic dilemma. The patient did not have any gastrointestinal symptoms, and only after further investigations, including a colonoscopy for the suspected assessment of carcinoma, the lesion located within the rectum. Similarly, active colitis with reparative changes was visualized in the first biopsy, which is a common finding in the case of endometriosis, which usually affects muscularis propria, while the mucosa only shows non-specific inflammatory infiltrates[8]. A rare but hazardous variant of a commonly occurring gynecological problem, Ureteric Endometriosis can cause significant and irreversible renal impairment, on occasion even being fatal[9]. Due to the early detection of the condition in this case, moderate Hydronephrosis without impairment of renal function was seen, and early placement of Double-J(DJ) stent was done to relieve the obstruction.

Endometriotic treatment options include medical management for the symptoms of dysmenorrhea which consists of non-steroidal anti-inflammatory drugs (NSAIDs), combined OCPs, and gonadotropin-releasing hormone (GnRH) agonists, as well as surgical procedures such as excision, fulguration, or laser ablation of endometriotic implants on the peritoneum, excision or drainage or ablation of endometriomas, resection of rectovaginal nodules, lysis of adhesions, and interruption of nerve pathways[10]. We opted for medical management with an intramuscular injection of 3.75 mg of Leuprolide, a GnRH agonist with add-back therapy of low dosage combined OCP. Add-back therapy with combined OCPs is done to prevent the loss of bone density, which is a common adverse effect of GnRH therapy.[11] Pharmacological therapy was preferred to available surgical options due to the lesion being large, ill-defined, and extending to multiple surrounding structures. She is planned for regular monthly follow-up and evaluation of the lesion until surgical interventions are amenable in the future.

Conclusion

Endometriosis, a prevalent condition among reproductive-age females, demonstrates considerable diversity in its presentation, clinical profile, and severity across patients. This instance notably exhibited an onset at 24 years of age without gastrointestinal symptoms, suggesting a potential case of Rectal endometriosis, complicating both diagnosis and treatment. Timely recognition and intervention are feasible through thorough clinical evaluation, along with comprehensive radiological and pathological assessments, particularly in reproductive-age females presenting with severe abdominal pain and associated genitourinary symptoms.

Method

We reported this case following the updated consensus-based Surgical Case Report (SCARE) Guidelines[12].

Ethical approval

Ethical approval is not required for case report in our institution (Shree Birendra Hospital).

Consent

An informed written consent was obtained from the patient for publication of this case report. A copy is available for review by the editor-in-chief of this journal on request.

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Author contribution

P.A.: history taking, examination, literature review writing the paper, editing, A.R.: literature review, writing, language edit, follow-up, editing. G.K.: editor, case selection. K.B.S.: editor, follow-up and guidance.

Conflicts of interest disclosure

The authors declare that they have no conflicts of interest.

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References

[1] Hacker NF, Moore JG, Gambone JC. Hacker and Moore's Essentials of Obstetrics and Gynecology, 6th ed. Elsevier; 2016. (Eds.).
[2] Remorgida V, Ferrero S, Fulcheri E, *et al*. Bowel endometriosis: presentation, diagnosis, and treatment. Obstet Gynecol Surv 2007;62:461–70.
[3] Douglas C, Rotimi O. Extragenital endometriosis—a clinicopathological review of a Glasgow hospital experience with case illustrations. J Obstet Gynaecol 2004;24:804–8.
[4] Sampson JA. Peritoneal endometriosis due to the menstrual dissemination of endometrial tissue into the peritoneal cavity. Am J Obstetr Gynecol 1927;14:422–69.
[5] Vercellini P, Frontino G, Pietropaolo G, *et al*. Deep endometriosis: definition, pathogensis, and clinical management. J Am Assoc Gynecol Laparosc 2004;11:127–36.
[6] Prittsand EA, Taylor RN. An evidence-based evaluation of endometriosis-associated infertility,. Endocrinol Metab Clin North Am 2003;32:653–67.
[7] Burney RO, Giudice L. Pathogenesis and pathophysiology of endometriosis. Fertil Steril 2012;98:511–9.
[8] Chen H, Luo Q, Liu S, *et al*. Rectal mucosal endometriosis primarily misinterpreted as adenocarcinoma: a case report and review of literature. Int J Clin Exp Pathol 2015;8:5902–7.
[9] Yantiss RK, Clement PB, Young RH. Endometriosis of the intestinal tract: a study of 44 cases of a disease that may cause diverse challenges in clinical and pathologic evaluation. Am J Surg Pathol 2001;25:445–54.
[10] Giudice LC, Kao LC. Endometriosis. Lancet 2004;364:1789–99.
[11] Al Kadri H, Hassan S, Al-Fozan HM, *et al*. Hormone therapy for endometriosis and surgical menopause. Cochrane Database Syst Rev 2009:CD005997.
[12] Sohrabi C, Mathew G, Maria N, *et al*. The SCARE 2023 guideline: updating consensus Surgical CAse REport (SCARE) guidelines. Int J Surg Lond Engl 2023;109:1136.