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Case report

Primary bilateral inguinal endometriosis: A case report

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ABSTRACT

Introduction: Endometriosis is a chronic benign recurrent gynecologic disease commonly affecting 10% of women worldwide wherein endometrial glands implant and mature outside the uterine cavity causing symptoms such as dysmenorrhea, dyspareunia, or abdominal pain.
Case presentation: Herein we describe a case of a 40-year-old female with primary bilateral inguinal endometriosis presenting with catamenial pain for which surgical excision was performed providing definitive treatment. The patient has been asymptomatic with no recurrence at 6 months of follow-up.
Discussion: Most cases of endometriosis occur within the pelvis however, extra-pelvic sites have been reported which include previous surgical scars, bladder, diaphragm, or inguinal area. It is usually classified as primary or secondary but can also be based on location. Oftentimes, these patients can present as a diagnostic dilemma for clinicians and treatment requires surgery and/or medications such as oral contraceptives or hormonal agents. Common diagnoses include hernia, lipoma, lymphadenopathy, or even malignancy.
Conclusion: We would like to highlight the atypical presentation, pathogenesis, and management of endometriosis in this rare site.

1. Introduction

Endometriosis is a chronic benign painful disease occurring frequently in women of reproductive age. It can be subdivided into primary or spontaneous and secondary endometriosis. Primary or spontaneous endometriosis is characterized by the absence of any preceding surgical procedure on the patient and its pathogenesis is not well understood. Secondary endometriosis on the other hand usually occurs in surgical scars after a surgical or gynecologic procedure which is the result of iatrogenic implantation of endometrial tissue. Although commonly found within the pelvis, lesions outside the pelvic area have been documented such as the inguinal area [1]. Because of its rarity and sometimes atypical presentation, inguinal endometriosis is usually misdiagnosed as hernias, lipoma, or lymphadenopathy A hallmark feature is the presence of inguinal pain associated with the menstrual cycle. Herein we describe a case of bilateral inguinal endometriosis for which surgical excision was performed. The case presents an opportunity to highlight its incidence, presentation and discuss its contemporary management. This report has been written in line with the recent SCARE criteria for case reports [2].

2. Case summary

A 40-year-old nulliparous female came in with a 1-year history of gradually enlarging bilateral inguinal masses associated with pain during menstruation. Ten years prior to consult, the patient noted a 1.5 × 1.5 cm right inguinal mass with no associated symptoms which was diagnosed as lymphadenopathy and advised observation. The patient remained asymptomatic until a year prior to admission, when the patient noted the appearance of a 1 × 1 cm left inguinal mass with pain upon palpation. Ultrasound done revealed bilateral lobulated hypoechoic masses with irregular margins (Fig. 1A, B). She was advised further work-up however, the patient decided to delay her management. She had no dysmenorrhea or pelvic pain, as well as no previous abdominal or gynecology surgery. At the time the patient presented, the right inguinal mass was already 3.5 × 3.5 cm, and the left was 2 × 1.5

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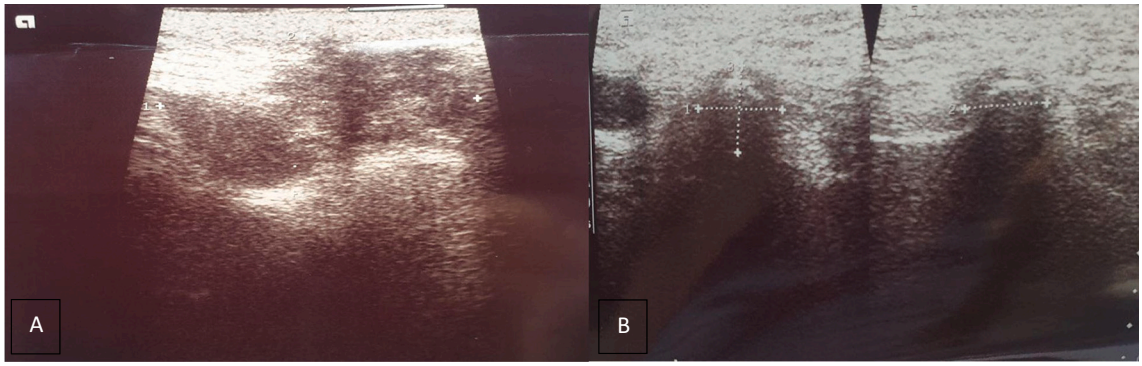

Fig. 1. Ultrasound images of the inguinal region. A. Right inguinal lobulated mass measuring 4.6 × 3.1 × 2.4 cm B. Left inguinal masses revealing two ovoid nodularities measuring 2.2 × 1.3 × 1.0 cm and 2.0 × 1.2 × 1.0, respectively with central calcification and posterior shadowing.

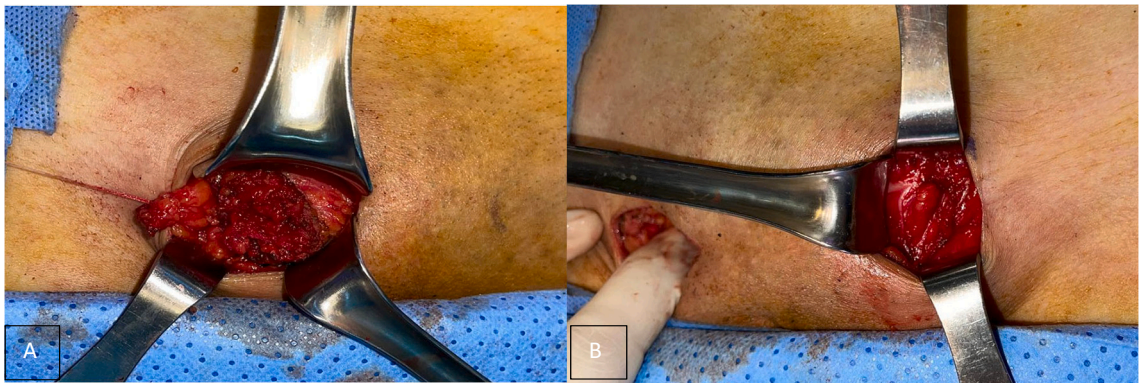

Fig. 2. Intraoperative images. A. The right inguinal mass was adherent to the right round ligament (with tag). B. Two left inguinal masses were excised.

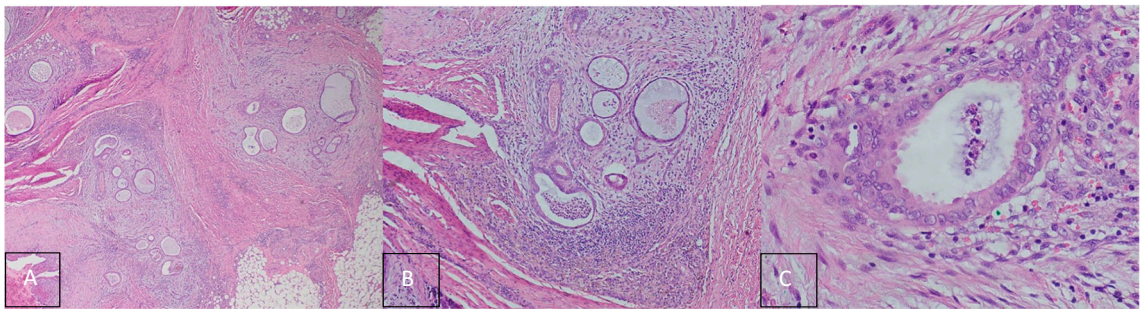

Fig. 3. Histopathology. A. Scanning view of the specimen shows clusters of ectopic benign glands with surrounding myxoid stroma (40×). B. On low power view, the glands are lined with single layer cuboidal cells. The surrounding stroma is composed of round basophilic cells in a myxoid stroma (100×). C. High power view showing an endometrial typed gland line with single layer of cuboidal cells with basal located nucleus. No atypia was noted (400×).

cm for which a needle biopsy showed endometriosis. The patient underwent excision of the inguinal masses with note of the right mass being adherent to the round ligament which was removed en-bloc (Fig. 2A, B). No hernial sac was noted intraoperatively. The patient was discharged a day after surgery. Final histopathology of both masses was consistent with endometriosis (Fig. 3A, B, C). No recurrence was noted on recent follow-up after 6 months.

3. Discussion

Endometriosis is defined as the growth of endometrial glands and stroma outside the uterine cavity occurring in 10% of women worldwide. In many cases, it presents with catamenial pain and/or infertility with frequent sites being within the pelvis. Extra-pelvic locations have been reported which include bladder, intestine, surgical scars, diaphragm, umbilicus, and inguinal region [3]. Inguinal endometriosis occurs in <1% of patients and commonly is unilateral involving the extraperitoneal portion of the round ligament [4]. It is often confused with other inguinal pathologies such as lymphadenopathy, hernia, malignancy or lipoma with needle biopsy providing a definitive diagnosis. Ultrasonography is the initial diagnostic test of choice which can also identify hernial sacs although findings can be variable. The pathogenesis of inguinal endometriosis remains unclear although theories proposed are that of retrograde menstruation, wherein endometrial cells move through the fallopian tube and into the peritoneal cavity; coelomic metaplasia, which designates normal peritoneal tissue transformation into ectopic endometrial tissue; and vascular or lymphatic spread [5]. There is no known cure and treatment is usually aimed at controlling symptoms.

Our patient presented with bilateral inguinal endometriosis which is

exceedingly rare with less than 5 cases reported in the literature [6]. It has been recommended to rule out pelvic endometriosis thru a diagnostic laparoscopy as these two conditions often coexist, however, with no history of dysmenorrhea or pelvic pain, as well as a normal gynecologic exam and transvaginal ultrasound, it was deemed not necessary. Niitsu et al. classified inguinal endometrosis into three types based on location: type I, hernial sac or hydrocele in the Canal of Nuck; type II, round ligament; and type III, under the skin, hence, the patient had a type II on the right and a type III on the left [7]. Secondary causes such as previous gynecologic procedures or surgeries were not present in our patient which makes her case likely primary. The right is predominantly involved in unilateral occurrence the reason for which also remains unclear although some have hypothesized that the sigmoid colon protects the left inguinal canal and endometrial cells remain on the right for a longer period due to the clockwise flow of intraperitoneal fluid [8]. Surgical excision is the mainstay of treatment and preventing recurrence as incidence of malignant transformation is low. Medical treatment such as the use of oral contraceptives, gonadotropin-releasing hormones (GnRH) agonists, or pain killers have been used but with limited symptomatic relief.

In conclusion, inguinal endometriosis is a rare disease that can present with or without menstrual cycle pain. A high index of suspicion is essential for inguinal masses particularly for women in their reproductive age. Needle biopsy is relatively quick and easy to perform yielding histopathologic diagnosis with excision rendering adequate treatment.

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Consent

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Author contribution

Eduardo Ayuste Jr. MD: writing of paper, draft preparation.
Emmanuel Limpin MD: data collection, draft preparation.
Jemuel Laygo MD: study concept, writing of paper.
Orlando Ocampo, MD: draft preparation, data collection.
Siegfredo Paloyo, MD, MPH: final editing, supervision, reviewing.

Research registration

N/A.

Guarantor

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Declaration of competing interest

None.

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