

CASE REPORT

CONCOMITANT INGUINAL ENDOMETRIOSIS AND GROIN HERNIA – CASE REPORT

Daniel Ion^{1,2}, Alexandra Bolocan^{1,2}, Silviu M. Pițuru¹, Petronela E. Mateoiu¹,
Florentina Mușat¹, Octavian Andronic¹, Dan N. Păduraru^{1,2}

¹The University of Medicine and Pharmacy „Carol Davila“, Bucharest, Romania

²The University Emergency Hospital of Bucharest, Romania

ABSTRACT

Endometriosis is a condition which affects women during their reproductive age. We present the case of a 42 years old Caucasian nulliparous woman accusing in the last three months a painful bulging mass in the right groin. The patient reported that the mass increases in size during prolonged standing and lifting of heavy weights. In addition, the inguinal pain was exacerbated during ovulation. The patient underwent surgery, during which wide excision of the nodule was performed. Furthermore, both direct and indirect hernia sacs were found and dissected, the content was reduced and the sacs were excised. Four months after the surgery, the patient was free of symptoms and had no signs of recurrence.

Key words: inguinal endometriosis, groin hernia, surgery.

INTRODUCTION

Endometriosis is a condition which affects women during their reproductive age. It is characterized by the presence and proliferation of endometrial stroma and glands outside the uterine cavity. Its occurrence

RÉSUMÉ

Endométriose inguinale associée à la hernie inguinale – rapport de cas

L'endométriose est une maladie qui touche les femmes pendant leur âge de procréation. On présente le cas d'une femme nullipare, caucasienne âgée de 42 ans qui accuse, depuis trois mois, une masse douloureuse située au niveau de l'aîne droite. La patiente a rapporté que la masse augmente en taille pendant la station debout prolongée et en levant de poids lourds. De plus, la douleur inguinale était exacerbée pendant l'ovulation. On a réalisé une large excision du nodule. Plus, des sacs de hernie directe et indirecte ont été trouvés et disséqués, le contenu a été réduit et les sacs ont été excisés. Quatre mois après la chirurgie, la patiente était exempte de symptômes et elle ne présentait aucun signe de récurrence.

Mots clés: endométriose inguinale, hernie inguinale, chirurgie.

is mainly in the pelvic organs, particularly the ovaries, pouch of Douglas, pelvic peritoneum and only occasionally in extrapelvic localization, especially the skin (surgical scars) and the viscera (small intestines, descending colon, rectum, appendix), cases which represent a specific interest for the general surgeon.¹

Inguinal endometriosis (IE) has been rarely reported so far, and accounts for less than 1% of patients affected by endometriosis.² It is relevant that 90% of cases with endometriosis in the extraperitoneal part of the round ligament were right-sided and between 30% and 40% of them were concomitant with an inguinal hernia.^{3,4}

CASE PRESENTATION

A 42 year-old Caucasian nulliparous woman presented for a painful bulging mass in the right groin, which appeared in the last three months. The patient reported that the mass increases in size during prolonged standing and lifting of heavy weights. In addition, the inguinal pain was exacerbated during ovulation. The patient received hormonal therapy for infertility, followed by in vitro fertilization.

Physical examination confirmed the presence of the protrusive painful mass, with cough impulse and reducible by maneuver of taxis. In addition, it revealed a 2x2 cm lump proximal to the external inguinal orifice, presenting an irregular surface and fibrous consistency. The lump was mobile on the surface plane, but fixed in depth. It was irreducible and did not transmit cough impulse.

The patient was submitted to surgery under general anesthesia for exploration of the right groin, with a preoperative diagnosis of mixed right inguinal hernia and either an incarcerated femoral hernia or an enlarged lymph node. During surgery, after opening the aponeurosis of the large oblique muscle, a 2 cm sized fibrous nodule was found (Fig. 1), close to the superficial inguinal orifice, and wide excision was performed. Furthermore, both direct and indirect hernia sacs were found and dissected, the content was reduced and the sacs were excised. The excision of the round ligament was also performed. Using

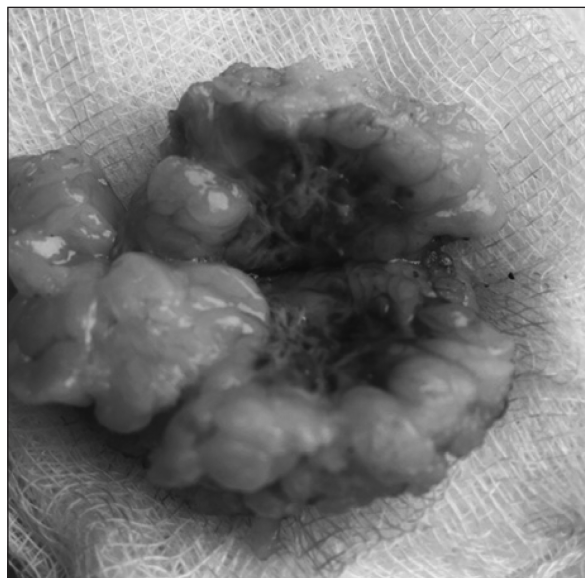


Fig. 1. The excised mass measuring approximately 2x2 cm.

Berliner technique, the strength of the posterior wall of the inguinal canal was restored.

Histopathological study of the nodule revealed fibrous tissue with various foci of endometrial type glands and stroma (Fig. 2).

Post-operative evolution was good, and the patient was discharged on post-operative day 2.

Four months after the surgery, the patient was free of symptoms and had no signs of recurrence or other complications.

DISCUSSION

Endometriosis represents a gynecological, benign, estrogen-dependent disease, with a prevalence of 8-15% and peak incidence between 40 and 50 years old.⁵

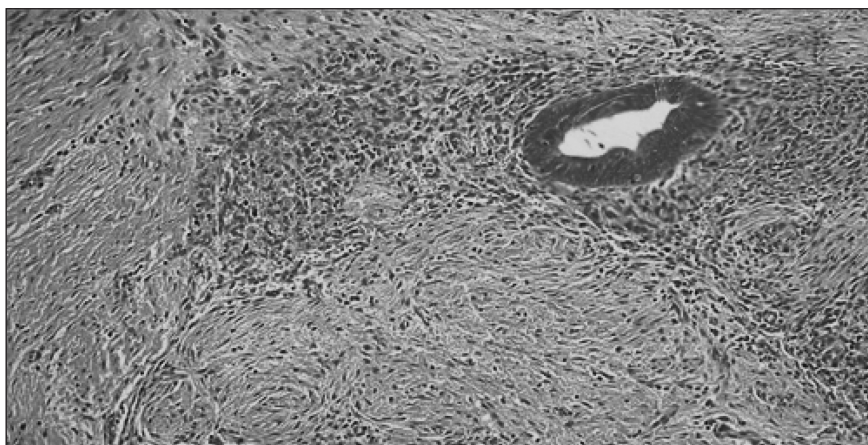


Fig. 2. Histopathological aspect of the specimen (endometrial glands and stroma).

Most frequent location of endometriosis is in the pelvis, unusual sites including intestine, surgical scars, diaphragm, umbilicus and groin.^{6,7} IE was first described by Allen in 1896 and is extremely rare, with around 60 cases reported in the literature.⁹ One study revealed that out of 958 patients who underwent clinical evaluation for endometriosis, 6 of them (0.6%) met the criteria of IE.¹⁰ More precisely, the foci of endometriosis can be found in the extraperitoneal portion of the round ligament, in the inguinal lymph node, in the fat subcutaneous tissue, and in the wall of sacs of inguinal or femoral hernias.^{1,11,12}

Age correlations

One study, addressing the age distribution among women suffering from endometriosis, showed that this type of pathology is not characteristic only to patients of reproductive age, but can also be encountered in many perimenopausal and postmenopausal women. Thus, out of 42,079 cases of endometriosis, 80.36% were in the premenopausal group (age 0-45 years), 17.09% in the perimenopausal group (age 45-55 years), and 2.55% (1,074 patients) in the postmenopausal group (age 55-95 years).¹³ The average age for abdominal wall endometriosis (defined as endometrial tissue superficial to the peritoneum) was found to be 31 years.¹⁴ In our case, the patient was 42 years old and also received hormonal therapy for infertility, which can be suggestive for unbalanced hormonal levels.

Diagnosis

Clinical features of IE usually result from functioning endometrial tissue and manifest as symptoms synchronized with menstrual cycle: pain, groin lump, catamenial symptoms.^{15,16} One review study showed that the pain is not necessarily cyclic, as more than half of the 445 patients group presented with cyclic symptoms¹⁴; this characteristic can be a useful information of the history. Endometrial tissue situated further away from the uterus can lose its hormonal receptors and response, therefore lack of manifestations or noncyclic symptoms may occur.¹⁷

Preoperative differential diagnosis of an inguinal mass is complex and includes: strangulated femoral or inguinal hernia, lymphadenopathy, hematoma, inguinal canal hydroceles, abscess, neuroma, lipoma, sarcoma, subcutaneous cysts and endometriosis.¹ An inguinal endometriotic mass is usually clinically misdiagnosed to be an incarcerated groin hernia and represents a difficult diagnosis for the physicians. Histopathological examination after surgery plays an important role in establishing the final diagnosis. The primary diagnostic imaging tool is ultrasonography, which allows identification of cystic, solid or

mixed hypoechoic masses, with absence of vascular flow around the lesion.¹⁸ CT is reserved to rule out the rare cases of malignancy. MRI is more accurate, but not widely implemented.

Due to the ease of performing and accuracy in results, Perez-Seoane et al consider that fine needle aspiration biopsy should probably be the first step in diagnosis of IE.¹² Immunohistochemical staining with progesterone receptor antibodies is the strongest positive staining in the nucleus of the stromal cells, in comparison with estrogen receptor antibodies, COX-2 and CD10 antibodies. However, the CD10 antibodies have the highest specificity in the cytoplasm of the stromal cells.¹⁵ Therefore, CD10 antibody may be useful in confirming a diagnosis of endometriosis when there is morphological doubt.

Surgical treatment

According to Romanian Law 95/2006, art. 660, the patient was informed about the possibility of carrying out imaging investigations that would provide additional information, but would not influence the surgical management. After all these aspects have been explained by the surgeon, according to paragraphs (2) and (3), the patient's written consent was requested. In order to obtain the patient's written consent, the attending physician provided the patient with the necessary information (diagnosis, nature and purpose, risks and consequences of the proposed treatment, viable alternatives, risks, consequences and prognosis of the disease without treatment) at a scientifically reasonable level of understanding.

Another very important aspect is represented by the professional component, namely the competence of the medical staff, who informed the patient and applied the treatment plan.

In cases of concomitant groin hernia and IE, the treatment of choice is primarily surgical. In bloc excision of the lesion and the extraperitoneal portion of the round ligament is required in order to prevent recurrence and allow histological diagnosis. Considering the high correlation with pelvic endometriosis, a gynecological referral⁹ is recommended for assessing the need of associated hormonal therapy.

Another aspect which should be considered in the management of IE is the association with pelvic endometriosis. One review showed that a history or subsequent diagnosis of pelvic endometriosis is associated only in 13% of the patients with endometrial tissue located superficial to the peritoneum.¹⁴ There have been reported cases where, when performed, laparoscopic exploration did not reveal the involvement of pelvic endometriosis in other sites.¹⁹⁻²¹

On the other hand, many studies and case reports^{10,22} demonstrated, by open or laparoscopic

exploration of the abdomen, that these concomitant localizations occur frequently. This can justify performing laparoscopic exploration of the peritoneal cavity in cases of IE. It also supports the idea that IE develops after the endometrial cells extended directly from the peritoneal fluid, through the deep inguinal ring, into the lymphatic or venous circulation of the round ligament. Our patient did not show any signs or symptoms of pelvic endometriosis and it was decided that she does not have surgical indication for a laparoscopic exploration.

Recurrence

The recurrence rate is 4.3% in cases of endometrial tissue superficial to the peritoneum.¹⁴ Although IE is a rare disease, recurrence of this lesion is frequent after inappropriate surgical management.

One study revealed that recurrence of IE appears in all cases where surgical treatment consisted only in the excision of the subcutaneous tissue, without identification of the round ligament in the inguinal canal. The 5 patients in this study with recurrent IE underwent a secondary surgical intervention for removal of the extraperitoneal portion of the round ligament, procedure which is considered to be the best treatment option in this type of pathology.^{10,22} In our case, the inguinal canal was opened, the extraperitoneal portion of the round ligament was excised, along with the 2 cm nodule and 2 indirect hernia sacs.

Complications

The association between cancer and endometriosis can be classified according to the localization of the malignant tissue in relation to ectopic endometrium cells. Thus, the most relevant association occurs in the case of co-localized or adjacent malignant tissue, whereas distant tumors are not always clearly related to the endometriosis site. Taking this into consideration, malignant degeneration is more frequently encountered in endometriosis confined to ovary (5.9%), than in extraovarian endometriosis (1.5%).²³

In IE, malignant degeneration can be a rare complication, as only 7 cases of adenocarcinoma-associated IE have been reported between 1988 and 2014, 5 of which were clear cell type, 1 serous type and 1 endometrioid type.²⁴

CONCLUSION

IE is a rare entity (less than 1% of all sites), affecting not only women of reproductive age. The symptoms include a slow growing, painful inguinal swelling, aggravated during ovulation. Although rare, IE should always be taken into consideration when

women have a presumptive diagnosis of incarcerated groin hernia. The diagnosis is usually set by histology, CD10 antibody being the most reliable immunohistochemical staining. Complete excision of the lesion is the treatment of choice and further gynecological investigations are recommended.

Authors contribution

All the authors have equal contribution to this paper. All authors approved the final version of the manuscript.

REFERENCES

1. Kim DH, Kim MJ, Kim M-L, Park JT, Lee JH. Inguinal endometriosis in a patient without a previous history of gynecologic surgery. *Obstetrics & Gynecology Science*. 2014;57(2):172-175.
2. Mourra N, Bennis M. CA. The GROIN: an unusual location of endometriosis. Groupe Hospitalier Paris Est experience. *Virchows Archiv*. 2014;465.
3. Clausen I, Nielsen KT. Endometriosis in the groin. *International Journal of Gynecology and Obstetrics*. 1987;25(6):469-471.
4. Sataloff DM, La Vorgna KA, McFarland MM. Extrapelvic endometriosis presenting as a hernia: clinical reports and review of the literature. *Surgery JT - Surgery*. 1989;105(1):109-112.
5. Nikkanen V, Punnonen R. External endometriosis in 801 operated patients. *Acta Obstet Gynecol Scand*. 1984;63(8):699-701.
6. Redwine DB. 19 Diaphragmatic endometriosis: diagnosis, surgical management, and long-term results of treatment. *Fertil Steril*. 2002;77(2):288-296.
7. Victory R, Diamond MP, Johns DA. Villar's nodule: A case report and systematic literature review of endometriosis externa of the umbilicus. *Journal of Minimally Invasive Gynecology*. 2007;14(1):23-32.
8. Seydel AS, Sickel JZ, Warner ED, Sax HC. Extrapelvic endometriosis: Diagnosis and treatment. *American Journal of Surgery*. 1996;171(2):239-241.
9. Albutt K, Glass C, Odom S, Gupta A. Endometriosis within a left-sided inguinal hernia sac. *Journal of Surgical Case Reports*. 2014;2014(5):rju046-rju046.
10. Candiani GB, Vercellini P, Fedele L, Vendola N, Carinelli S, Scaglione V. Inguinal endometriosis: pathogenetic and clinical implications. *Obstetrics and gynecology*. 1991;78(2):191-194.
11. Jenkins S, Olive DL, Haney AF. Endometriosis: pathogenetic implications of the anatomic distribution. *Obstetrics and gynecology*. 1986;67(3):335-338.
12. Perez-Seoane C, Vargas J, de Agustin P. Endometriosis in an inguinal crural hernia. Diagnosis by fine needle aspiration biopsy. *Acta Cytol*. 1991;35(3):350-352.
13. Haas D, Chvatal R, Reichert B, et al. Endometriosis: A premenopausal disease Age pattern in 42,079 patients with endometriosis. *Archives of Gynecology and Obstetrics*. 2012;286(3):667-670.
14. Horton JD, DeZee KJ, Ahnfeldt EP, Wagner M. Abdominal wall endometriosis: a surgeon's perspective and review of 445 cases. *American Journal of Surgery*. 2008;196(2):207-212.

15. Terada S, Miyata Y, Nakazawa H, et al. Immunohistochemical analysis of an ectopic endometriosis in the uterine round ligament. *Diagnostic Pathology*. 2006;1:27.
16. Li AC, Siu WT LM. Endometrioma simulating inguinal hernia: case reports. *Can J Surg*. 1999;42:387-388.
17. Markham SM, Carpenter SE RJ. Extrapelvic endometriosis. *Obstet Gynecol Clin North Am*. 1989;16:193-219.
18. Elemenoglou J, Skopelitou A, Nomikos I. Carcinoma of the inguinal region arising from endometriosis of the round ligament. Report of a case. *European Journal of Gynaecological Oncology*. 1993;14(1):28-32.
19. Kaushik R GA. Inguinal endometriosis: A case report. *J Cytol*. 2008;25:73-75.
20. Quagliarello J, Coppa G, Bigelow B. Isolated endometriosis in an inguinal hernia. *American Journal of Obstetrics and Gynecology*. 1985;152(6):688-689.
21. Wong WSF, Lim CED, Luo X. Inguinal endometriosis: an uncommon differential diagnosis as an inguinal tumour. *ISRN Obstetrics and Gynecology*. 2011;2011:1-4.
22. Fedele L, Bianchi S, Frontino G, Zanconato G RT. Radical Excision of Inguinal Endometriosis. *Obstet Gynecol*. 2007;110:530-533.
23. Stern RC, Dash R, Bentley RC, Snyder MJ, Haney AF, Robboy SJ. Malignancy in endometriosis: frequency and comparison of ovarian and extraovarian types. *International Journal of Gynecological Pathology*. 2001;20(2):133-139.
24. Bergamini A, Almirante G, Taccagni G, Mangili G, Viganò P, Candiani M. Endometriosis-associated tumor at the inguinal site: Report of a case diagnosed during pregnancy and literature review. *Journal of Obstetrics and Gynaecology Research*. 2014;40(4):1132-1136.