CASE REPORT

Rare Case of Perianal Endometriosis Complicated with Perianal Fistula: Case Report

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Abstract

Perianal endometriosis is a rare pathology which occurs mainly in patients with history of vaginal birth associated with an episiotomy scar. We present the case of a 42 year old patient with history of multiple interventions for right perianal fistula, accusing pain, purulent secretions and bleeding at the site of the interventions. Biopsies collected were specific for endometriosis. In the context of the sphincter involvement, which brings a high risk of incontinence, and the hormone-dependent evolution of this pathology, which tends to regress once menopause is reached, a fistulotomy was made, with subsequent remission of the purulent secretions, but with the persistence of the bleeding from the region during the menstrual cycle.

Keywords: endometriosis perianal, fistula, fistulotomy.

Rezumat

Endometrioza perianală este o patologie rară, ce survine mai ales la pacienți cu istoric de nașteri vaginale asociate cu epiziotomie. Prezentăm cazul unei paciente de 42 de ani, în antecedente cu multiple intervenții pentru fistulă perianală dreaptă, acuzând dureri și exteriorizare de secreții purulente și sangvinolente de la acel nivel. Biopsiile prelevate intraoperator susțin diagnosticul de endometrioză. În contextul afectării sfincteriene ce aduce cu sine un risc mare de incontinență, și a evoluției hormo-dependente a acestei patologii, ce tinde să regreseze odată cu apariția menopauzei, s-a optat pentru practicarea unei fistulotomii, cu remiterea secrețiilor purulente și menținerea unei secreții sangvinolente concomitent cu ciclurile menstruale.

Cuvinte cheie: endometrioză, perianal, fistulă, fistulotomie.

INTRODUCTION

Endometriosis is a pathology defined by the presence of functional endometrial tissue outside of the uterus. The most frequent locations are the ovaries, uterosacral ligaments, and the rectum. Perianal region is a rarely affected^{1,2}. It is a condition that affects between 20% and 50% of women that are infertile³. In one study in a specialised centre in China, between 1992 and 2011,

out of a total of 17263 patients operated for endometriosis, 64 patients (3.7% out of the total) presented endometriosis in the perianal region, and 31 (1.8%) had sphincter involvement⁴.

CASE PRESENTATION

We present the case of a 42 year old patient, accusing perianal pain associated with purulent secretions and

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Figure 1. Focal tubular glands with a single epithelial layer, embedded in a dense connective tissue, HE stain, x200.

Figure 2. Positive immunohistochemistry reaction for estrogen receptors in glandular epithelium, x200.

bleeding. The patient has history of multiple interventions for perianal fistula between 2015-2018, the last operation consisting of the insertion of a drainage seton. The patient has history of vaginal birth associated with right lateral episiotomy at the age of 21.

At the clinical examination we detect low transphincteric perianal fistula, with an applied drainage seton and multiple perianal and right ischio-rectal fluid collections, suggestive for „hourglass” shape abscesses. Anuscopy revealed the internal orifice of the fistula at 1cm in the anal canal, at 1 o'clock in lateral left decubitus and turgescient haemorrhoids with signs of recent bleeding.

The MRI and endoanal ultrasound have detected a perianal abscess of about 3 cm, with multiple fistulous ramifications that go deep into the adipose tissue, with

surrounding fibrous scar tissue in the context of previous surgeries, which made the evaluation of sphincter involvement difficult.

As for blood tests, the patient had no inflammatory syndrome, and the tumour marker CA 125 was within normal range. There are studies that have demonstrated the superiority of this marker in detecting pelvic endometriosis⁵, although it is not specific for other sites of endometriosis, perianal included⁶.

The surgery consisted of incision, evacuation of the abscesses, fistulotomy, multiple sphincter and ischio-rectal biopsies and wound dressing. Postoperative evolution is initially favourable, but in the 22nd day, the surgical wound gained a swollen aspect and presented bleeding at the time of the menstrual cycle. Bleeding was not associated with pain. The biopsy results (hematoxylin and eosin colouring) are positive for perianal fistula, immunohistochemistry confirms the endometriosis (stroma and glands strongly express estrogen, glandular epithelium expresses CK7 and CD10 with minimal reaction). Thus the final diagnosis is perianal endometriosis complicated with low transphincteric fistula and perianal and right ischio-rectal abscesses. Endocrinological and gynaecological assessment recommends hormonal inhibitor treatment with diphereline. The patient refuses both hormonal treatment and surgical castration, choosing therapeutic abstinence in the context of approaching menopause and the possibility of the regression of endometriosis.

DISCUSSION

Perianal endometriosis is a rare pathology, and in most of the cases presented in literature, the patient presented episiotomy scar from vaginal birth, incision that is the primary implantation site for endometrial tissue⁷. The multitude of previous surgeries that led to the persistence of local suppuration made it impossible to determine the cyclic character of the bleeding. In a study of endometriosis of the abdominal wall, abdominal pain was not associated with the menstrual cycle in 75% of cases⁸. At the operating moment, in lack of the suspicion of perianal endometriosis, the choice was made to cure the fistula and to evacuate the perianal and ischio-rectal abscesses, the wide curettage of the tissues that were considered fibrous at the time of the intervention, avoiding the areas where the apparent fibrous tissue involved the anal sphincter. The final diagnosis in this case came after the operation, in the results of the biopsy collected during surgery. There are studies that have demonstrated the superiority of the fistulotomy

regarding continence impairment and healing time⁹. In this case the fistulotomy was completed with wide curettage of the apparent fibrous tissues, avoiding the anal sphincter. Studies have constantly shown a correlation between an episiotomy scar in patient history and perianal endometriosis. The time between the episiotomy and the appearance of the first symptoms was 18 years. The patient did not acknowledge the presence of perianal nodules before the first perianal abscess occurred, and none of the previous surgeries had collected a biopsy. The patient has chosen conservative treatment in the context of age approaching menopause, which favours natural regression of endometriosis. A case of perineal endometriosis that had spontaneously regressed after a pregnancy was reported, sustaining that fact that it is a hormone-dependent pathology¹⁰. The malignancy rate of endometriosis is cited to be low, between 0.7%-1%, of which 80% occurs in the ovaries¹¹. Postoperative follow up, for a period of 18 months, consisted of local bandaging of the wound during menstrual cycles, without local suppuration.

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CONCLUSION

There are 3 criteria on which the diagnosis of perianal endometriosis can be based: history of vaginal birth, palpable nodules of moderate consistency in the perianal region, and pain worsened during the menstrual cycle, followed by perianal bleeding¹². One role in adopting an adequate therapeutic conduct is the hormone-dependent evolution of this pathology, especially when the anal sphincter is involved and there is a high risk of incontinence in the case of wide excision, which are to be avoided.

Compliance with ethics requirements:

The authors declare no conflict of interest regarding this article.

The authors declare that all the procedures and experiments of this study respect the ethical standards in the Helsinki Declaration of 1975, as revised in 2008(5), as well as the national law. Informed consent was obtained from all the patients included in the study.