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Vulvar involvement in inflammatory bowel diseases: when gynecological assessment assists gastroenterological management

Envolvimento vulvar nas doenças inflamatórias intestinais: quando a avaliação ginecológica contribui para a abordagem gastroenterológica

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Abstract

Inflammatory Bowel Disease is a complex entity which complications that can lead to long-term repercussions. It includes two major disorders: ulcerative colitis (UC) and Crohn disease. Although they have different clinicopathologic characteristics, sometimes they overlap, making the diagnosis challenging. These patients can exhibit extraintestinal manifestations, namely vulvovaginal fistulas, with increase burden on health of affected women. We report a case of 30-year-old women with a longstanding history of UC complaining of vulvar edema, perineal pain and abnormal vaginal discharge with 2 weeks of duration and whose multidisciplinary approach enabled a new management of her gastroenterological pathology.

Keywords: Inflammatory bowel diseases; Genitalia; Physical examination; Magnetic resonance imaging; Fistula.

Resumo

A doença inflamatória intestinal (DII) é uma entidade complexa cujas complicações podem ter repercussões a longo prazo nas doentes. Inclui dois grupos principais: a colite ulcerosa (CU) e a doença de Crohn. Embora tenham caraterísticas clinicopatológicas diferentes, ocasionalmente, sobrepõem-se, dificultando o seu diagnóstico. Estas doentes podem apresentar manifestações extraintestinais, nomeadamente fístulas vulvovaginais, com impacto significativo na sua saúde. Apresentamos o caso de uma mulher de 30 anos com antecedentes crónicos de CU e com queixas de edema vulvar, dor perineal e corrimento vaginal anormal com 2 semanas de evolução, cuja abordagem multidisciplinar estabeleceu uma nova orientação da sua patologia gastroenterológica.

Palavras-chave: Doenças inflamatórias intestinais; Genitais; Exame físico; Imagem por ressonância magnética; Fístula.

Inflammatory bowel disease (IBD) is a complex condition formed by two major disorders: ulcerative colitis (UC) and Crohn disease (CD)[1]. Although primarily affecting the gastrointestinal tract, up to 40% of patients develop a variety of extra-intestinal manifestations (EIM)[2]. Based on literature, "Knife-cut" ulcers, fistulas and/or abscesses and vulvar edema are presentations of vulvar CD[3]. However, vulvovaginal symptoms of IBD are a rare EIM, especially in UC[4,5].

A 30-year-old woman, G1P0, with a longstanding history of uncontrolled UC treated with mesalazine

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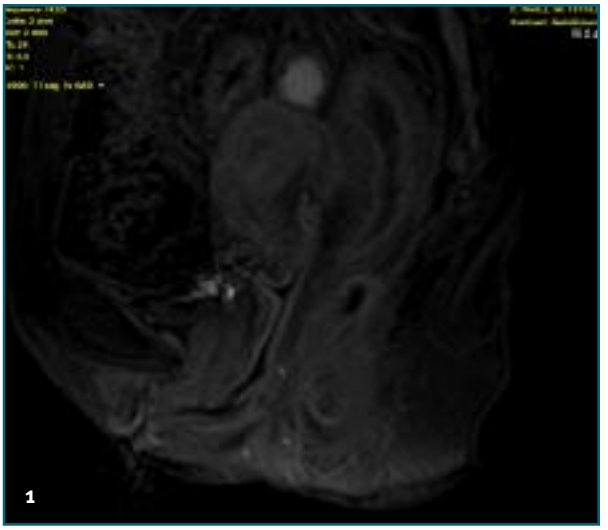

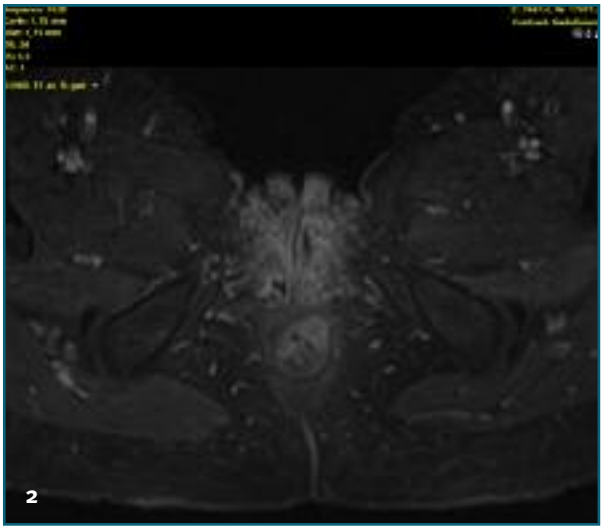

FIGURE 1 and 2. Images from sagittal (1) and axial (2) Fat Suppressed Contrast-Enhanced T1-weighted pelvic magnetic resonance imaging indicating transphincteric fistulization envolving the labia majora (arrows).

2000 mg daily presented with an initial 2-weeks history of vulvar edema, perineal pain and abnormal vaginal discharge. Following the observation of these findings on gynecological examination, a Bartholin gland abscess was considered, and empirical treatment with flucloxacillin (500 mg 8/8 h for 15 days), prescribed through the primary health care center, was initiated. Nonetheless, her complains have remained and additionally she started to exhibit fever. On physical examination, an asymmetrical vulvar swelling and an exuberant purulent discharge coating her labia were observed. A detailed inspection revealed the discharge of purulent material through small erosion areas located on the inner side of the labia minora. The perianal examination was unremarkable. Samples of vulvovaginal exudates were collected and laboratory evaluation demonstrated elevated blood inflammatory markers (neutrophilic leukocytosis and C-reactive protein level of 107 mg/L). A pelvic magnetic resonance imaging (MRI) identified two transphincteric fistulas originating from the internal anal sphincter and extending into the labia, inflammatory changes throughout the vulvar region, diffuse rectal wall thickening, and a small perirectal fluid collection communicating with the rectal wall at the four o clock position (Figures 1 and 2).

Based on the clinical presentation, imaging findings, and the absence of typical features associated with UC such as continuous superficial mucosal inflammation, the diagnosis was revised from UC to CD. The presence of complex perianal and vulvovaginal fistulas, transmural inflammation, and deep tissue involvement are characteristic of CD and exceedingly rare in UC. Accordingly, a reclassification to CD was made by consensus through a multidisciplinary evaluation with Gastroenterology and General Surgery.

The patient was admitted to inpatient management. Empiric intravenous antibiotic therapy was started with metronidazole (500 mg twice daily) and cefuroxime (500 mg twice daily). Antibiotic switch therapy for ertapenem (1 g daily) was necessary after the results of vulvar discharge cultures. Despite clinical improvement after one week, inflammatory markers remained elevated and a new MRI assessment revealed only slight radiologic improvement. Thus, surgical treatment was offered and partial fistulotomy with seton placement was performed by General Surgery team, specialized in colorectal and perineal pathology, without complications. She was discharged on postoperative day four.

At the first follow-up visit, she reported cramping abdominal pain and fecal urgency with diarrhea and a combination therapy of infliximab with azathioprine

and budesonide was started. Subsequent clinical records mention significant clinical improvement.

In summary, awareness of the gynecologic effects of chronic disease and early multidisciplinary collaboration are crucial for improving health outcomes in women with IBD, given the potential for long-term repercussions and the absence of standardized management guidelines.[3,4]

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AUTHORS' CONTRIBUTIONS

Tiago Meneses Alves was responsible for the patient's clinical care (diagnosis) and for writing the article (original draft, review and editing). Inês Falcão Reis was responsible for the patient's clinical care (diagnosis) and for writing the article (review and editing). Cátia Rasteiro was responsible for the patient's clinical care (diagnosis) and for writing the article (review and editing).

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CONFLICTS OF INTEREST

The authors report no conflict of interest.

PATIENT CONSENT

The informed consent was obtained from the patient who authorized the publication of this article.

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