ACG CASE REPORTS JOURNAL

CASE REPORT | INFLAMMATORY BOWEL DISEASE

Granulomatous Salpingo-oophoritis Secondary to Crohn's Disease

Ianina Belén Capaldi, MD[1], Florencia Giraudo, MD[1], María Laura Garbi, MD[1], Martín Yantorno, MD[1], Nicolás Capurro, MD[2], Fernanda García Vaz, MD[3], and Gustavo Javier Correa, MD[1]

[1]*Gastroenterology Department, Hospital Interzonal General de Agudos San Martí n de La Plata, La Plata, Argentina*
[2]*Pathology Department, Hospital Interzonal General de Agudos San Martí n de La Plata, La Plata, Argentina*
[3]*Surgery Department, Hospital Interzonal General de Agudos San Martí n de La Plata, La Plata, Argentina*

ABSTRACT

Crohn's disease (CD) represents a subtype of inflammatory bowel disease and can affect any portion of the gastrointestinal tract, from the mouth to the anus, with the capacity to affect extraintestinal organs. Salpingo-oophoritis is an uncommon manifestation of CD. There is only a limited number of documented case reports. We present the case of a patient with ileocolonic CD and secondary granulomatous salpingo-oophoritis. We emphasize the significance of clinical suspicion and an interdisciplinary approach as crucial factors in ensuring the effective management of the case.

KEYWORDS: case report; inflammatory bowel disease; Crohn's disease; salpingo-oophoritis; genitourinary involvement

INTRODUCTION

Crohn's disease (CD) represents a subtype of inflammatory bowel disease (IBD) characterized by the presence of chronic transmural inflammation, which can subsequently lead to abscesses, fistulae, and stenosis.[1] This condition can affect any portion of the gastrointestinal tract, and the development of extraintestinal manifestations is also possible.[2] Genitourinary involvement is considered a rare extraintestinal manifestation and may manifest either before or after intestinal activity, occasionally resembling pelvic disorders.[3] Moreover, the patient with IBD complains of urinary tract symptoms frequently.[7]

There is limited literature with respect to ovarian involvement secondary to CD.[2,3] Granulomatous ovarian manifestation is generally uncommon, except in instances involving an entero-ovarian fistula or contiguous extension.[6] Diagnosing this condition can be challenging, and histopathological assessment is essential. It is characterized by the presence of noncaseating granulomatous inflammation, and there are documented case reports of a related suppurative inflammatory reaction.[1] In this study, we present the clinical case of a patient with right salpingo-oophoritis and ileocolonic CD.

CASE REPORT

A 31-year-old woman was referred to our Gastroenterology Department because of a perianal abscess, which had been unsuccessfully treated with antibiotics and surgical drainage. She presented with chronic diarrhea and a history of an appendectomy performed 4 years ago, as well as a right salpingo-oophorectomy because of an adnexal inflammation and a right hemicolectomy with ileotransverse anastomosis as postoperative complications. At that moment, the anatomopathological evaluation of the surgical specimen was reported as nonspecific inflammation. Given the patient's medical history and clinical presentation, IBD was suspected. An ileocolonoscopy under conditions of optimal bowel preparation according to good standards was performed, with no signs of lesions.[4] This was followed by a contrast-enhanced pelvic magnetic resonance imaging, revealing a complex transsphincteric perianal fistula involving the rectovaginal septum and extending to the ischiorectal fossa, along with 2 collections (35 × 17 mm and 55 × 26 mm), one of them extending to the skin. There was also a 13-mm thickening of the rectum and sigmoid wall. An enterography was performed, revealing no pathological findings. The ileum and colon biopsies showed active chronic inflammation

ACG Case Rep J 2024;11:e01388. doi:10.14309/crj.0000000000001388. Published online: June 21, 2024

Correspondence: Ianina Belén Capaldi, MD (ianinacapaldi@gmail.com).

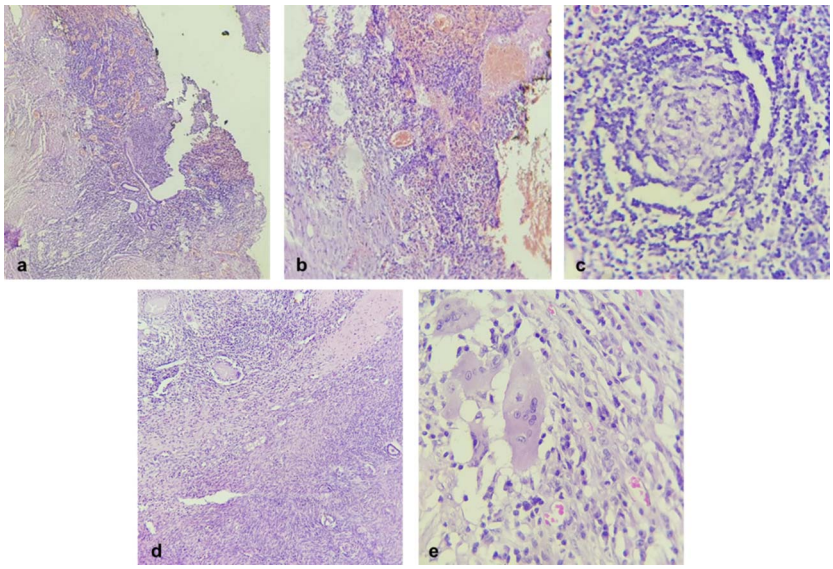

Figure 1. Hematoxylin-and-eosin–stained histological sections from colonic mucosa, at 4×, 10×, and 40×, respectively. Colonic mucosa with signs of acute abscessed inflammation (A and B) and pseudogranulomatous reaction (C). (D and E) Hematoxylin-and-eosin–stained sections from ovarian stroma, at 4× and 10×, respectively. Ovarian stroma with inflammatory process and gigantocellular reaction.

with architectural distortion and isolated crypt micro-abscesses. Subsequently, a re-evaluation of the surgical specimens belonging to the right salpingo-oophorectomy and right hemicolectomy was undertaken, revealing acute abscessed salpingo-oophoritis with a microfocus of non-caseating granulomatous inflammation and ileocolonic inflammation with signs of acute abscessed inflammation and pseudogranulomatous reaction, along with the presence of an enterotubal fistula (Figure 1). The clinical presentation was reinterpreted, leading to a diagnosis of CD with ileocolonic involvement, characterized by a penetrating behavior. This condition affected the ovary and Fallopian tube, manifesting as a severe onset that required emergency surgery. Therefore, the manifestations displayed by the patient are considered as a new flare of the disease, characterized by complex perianal involvement but no endoscopic evidence of luminal activity. Antibiotic treatment and surgical drainage of the collections with the placement of a seton were chosen as the course of action. Biologic therapy was initiated with anti-TNF, infliximab at a dose of 5 mg/kg with an induction regimen at weeks 0, 2, and 6, in combination with azathioprine at daily dose of 50 mg. Maintenance treatment was administered every 8 weeks. The patient progressed favorably, experiencing clinical remission of the perianal disease, endoscopic evaluation revealed mucosal healing, and pelvic magnetic resonance imaging, indicating transmural healing with resolution of the fistula and normalization of thickening in the intestinal wall.

DISCUSSION

CD is a subtype of IBD, which is characterized by the presence of chronic transmural inflammation, with the consequent potential formation of fistulas or stenosis.[1] The skin, small intestine, colon, and bladder are the organs commonly involved in fistula formation, and less frequently, transmural involvement of the ileum, proximal colon, or rectum can penetrate into the uterus, ovaries, and Fallopian tubes. Right adnexal involvement is the most common form, and it is typically the result of penetrating terminal ileitis.[6]

CD with genitourinary involvement can either precede or occur independently of intestinal involvement, and it can resemble various pelvic diseases, making diagnosis challenging. There are limited records of ileotubal fistulae related to CD in the literature. In 1975, Wlodarski et al reported the first case of CD with ovarian involvement, describing granulomatous inflammation of 1 ovary because of an ileocecal-tubo-ovarian mass.[10] Likewise, in 1978, Donaldson et al described the development of abscesses, fistulae, fissures, ulcers, and infections involving both internal pelvic structures and the perineum, labia, rectogenital septum, anus, rectum, vulva, and vagina in a group of 103 women with CD. In addition, they observed 12 patients with internal fistulae, 5 of which were located between the affected intestinal segment and the bladder, uterus, vagina, or pelvic adnexa. In 3 cases, the primary surgical findings were ileitis and appendicitis secondary to their IBD, with involvement of the right ovary and Fallopian tube.[3]

In our case, when considering the patient's medical history, her previous episode of surgical acute abdominal pain, initially interpreted as secondary to appendicitis, and its complicated progression because of an adnexal inflammation, which ultimately led to surgical resolution involving right hemicolectomy and salpingo-oophorectomy, turned out to be the onset of her IBD. This IBD affected the terminal ileum and appendix, with a penetrating nature extending to her female genital organs. Her initial symptoms and the low frequency of annexal involvement in CD led to a delayed diagnosis of IBD.

In terms of the mechanism, CD can affect the adnexa not only through direct extension but also metastatically or as a true extraintestinal manifestation.[12] These cases commonly present as unilateral pelvic pain or a pelvic mass, which can resemble conditions such as pelvic inflammation, endometriosis, active intestinal inflammation, diverticulitis, appendicitis, and primary ovarian pathology, among others.[6] Hence, a correlation can be observed between the proposed pathological mechanism and histological findings, with the presence of noncaseating granulomas in cases of direct involvement by CD and suppurative granulomatous inflammation in cases of intestinal fistulization. In the case of visceral granulomatous inflammation, it is crucial to rule out other causes such as tuberculosis, actinomycosis, or fungal infection.[3,5]

Regarding the management of genital involvement in CD, anal/rectovaginal fistulas may respond to medical treatment with biologic agents, and surgical treatment is an option in refractory cases or complex perianal disease.[9] The initial steps typically involve the placement of setons and drainage of associated collections or, in the case of low fistulas, fistulotomy.[2] Moreover, internal genital fistulas are typically diagnosed during surgical procedures and often require resection of the affected intestinal segment (usually the terminal ileum) along with the adjacent compromised gynecological organ.[6] Finally, vulvar involvement, either because of the extension of the inflammatory process or, less commonly, metastatic disease, if left untreated, can lead to various complications ranging from local inflammatory processes to local lymphatic obstructions, disfiguring anatomical distortions, and squamous cell carcinoma.[8] Currently, there are no treatment guidelines concerning the vulvar compromise. However, oral antibiotics such as metronidazole combined with corticoids are commonly used.[11] In cases of refractory disease, especially if it is associated with luminal involvement, biologic agents are an excellent option.[8] In summary, salpingo-oophoritis is a rare manifestation of CD, with few case reports in the literature. Clinical suspicion is crucial for timely diagnosis, and a multidisciplinary approach is essential for its proper management.

DISCLOSURES

Author contributions: IB Capaldi and F. Giraudo wrote the manuscript; ML Garbi provided medical history information. M. Yantorno contributed to the multidisciplinary approach. N. Capurro participated as the pathologist in charge of the case. FG Vaz participated by providing surgical information. GJ Correa supervised and approved the manuscript. IB Capaldi is the article guarantor.

Financial disclosure: The authors have no conflict of interest to declare regarding the publication of this paper.

Informed consent was obtained for this case report.

Received November 23, 2023; Accepted May 7, 2024

REFERENCES

1. Allen DC, Calvert CH. Crohn's ileitis and salpingo-oophoritis. *Ulster Med J* 1995;64(1):95–7.
2. De la Poza G, López-Sanroman A, Taxonera C, et al. Genital fistulas in female Crohn's disease patients: Clinical characteristics and response to therapy. *J Crohn's Colitis* 2012;6(3):276–80.
3. Donaldson LB. Crohn's disease: Its gynecologic aspect. *Am J Obstet Gynecol* 1978;131(2):196–202.
4. Gravina AG, Pellegrino R, Romeo M, et al. Quality of bowel preparation in patients with inflammatory bowel disease undergoing colonoscopy: What factors to consider? *World J Gastrointest Endosc* 2023;15(3):133–45.
5. McCluggage WG, Allen DC. Ovarian granulomas: A report of 32 cases. *J Clin Pathol* 1997;50(4):324–7.
6. Mohammed H, Bokhary R, Nassif M, Mosli M. Ovarian Crohn's disease: A case report and review of the literature. *Case Rep Gastrointest Med* 2020; 2020(32):1–5.
7. Romano L, Pellegrino R, Arcaniolo D, et al. Lower urinary tract symptoms in patients with inflammatory bowel diseases: A cross-sectional observational study. *Dig Liver Dis* 2024;56(4):628–34.
8. Shields BE, Richardson C, Arkin L, Kornik R. Vulvar Crohn disease: Diagnostic challenges and approach to therapy. *Int J Womens Dermatol* 2020; 6(5):390–4.
9. Sambuelli AM, Negreira S, Gil A, et al. Manejo de la enfermedad inflamatoria intestinal. Revisión y algoritmos de tratamiento. *Acta Gastroenterol Latinoam* 2019;49(S2).
10. Wlodarski FM, Trainer TD. Granulomatous oophoritis and salpingitis associated with Crohn's disease of the appendix. *Am J Obstet Gynecol* 1975; 122(4):527–8.
11. Sides C, Trinidad MC, Heitlinger L, Anasti J. Crohn disease and the gynecologic patient. *Obstetrical Gynecol Surv* 2013;68(1):51–61.
12. Tonolini M, Villa C, Campari A, Ravelli A, Bianco R, Cornalba G. Common and unusual urogenital Crohn's disease complications: Spectrum of cross-sectional imaging findings. *Abdom Imaging* 2013;38(1):32–41.