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IMAGE | INFLAMMATORY BOWEL DISEASE

Simultaneous Presentation of Idiopathic Duct-Centric Pancreatitis and Ulcerative Colitis

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CASE REPORT

A 22-year-old female with no significant past medical history was admitted with chronic bloody diarrhea and persistent epigastric pain radiating to the back for the previous 2 months. There was no history of nonsteroidal anti-inflammatory drugs or alcohol consumption. Physical examination revealed tenderness in the epigastrum, without peritoneal signs. Laboratory results showed leukocytosis (15,400/μL), elevated C-reactive protein (26.4 mg/dL), elevated lipase (8,158 UI/L), and positive anti-saccharomyces cerevisiae antibodies (ASCA) and anti-neutrophil cytoplasmatic antibodies (ANCA). Neither abdominal ultrasound nor computed tomography showed any abnormality. A colonoscopy and ileoscopy were performed and revealed granular, swollen, and friable mucosa with continuous involvement of the entire colon and rectum, with terminal ileum spared (Figure 1). Biopsies from the colon and rectum showed distortion of crypt architecture with branching crypts, globet cells depletion, increased inflammatory cells in the lamina propria, and crypt abscesses (Figure 2). An endoscopic ultrasound revealed a dif-

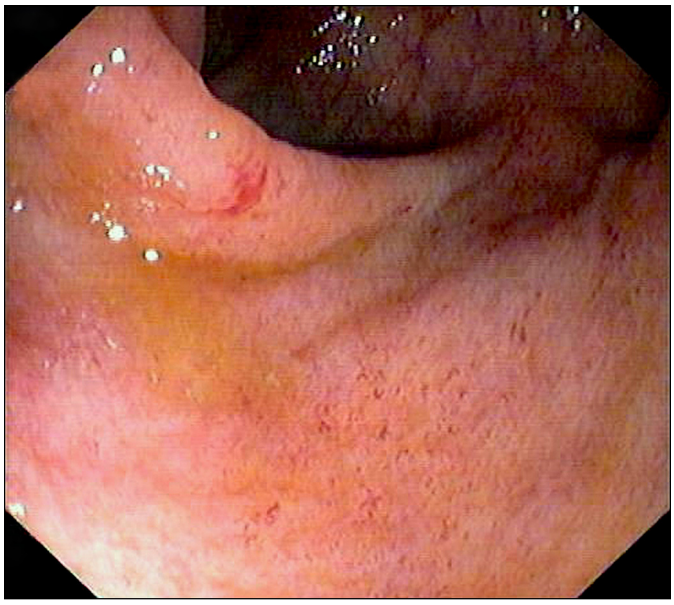

Figure 1. Colonoscopy showing granular, swollen, and friable mucosa with continuous involvement of the entire colon (Mayo score 1).

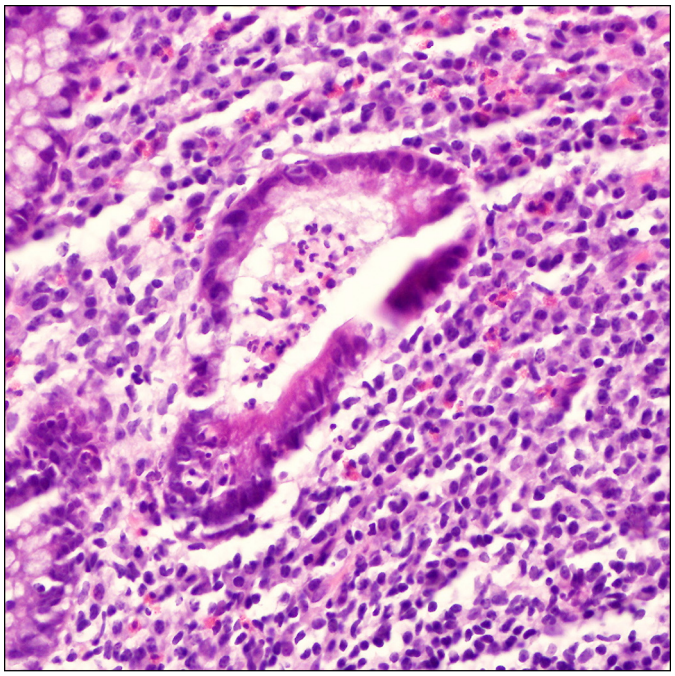

Figure 2. Colon biopsy (hematoxylin and eosin, 100x) showing distortion of crypt architecture with branching crypts, globet cells depletion, increased inflammatory cells in the lamina propria and crypt abscesses.

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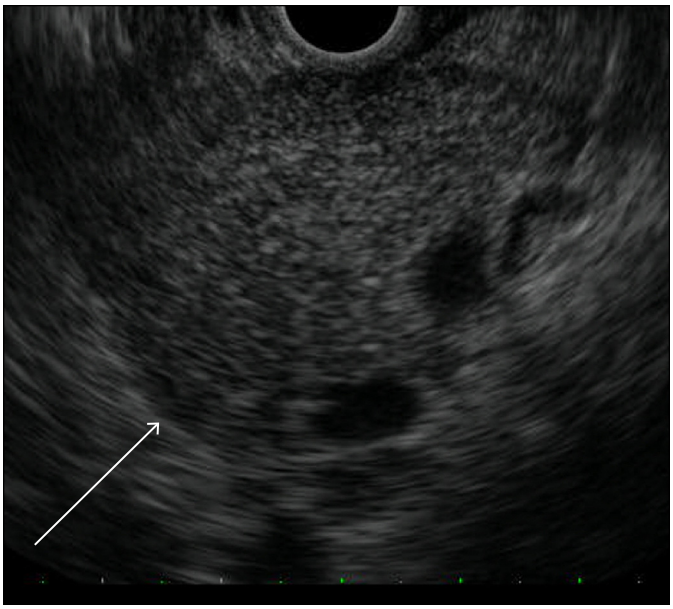

Figure 3. Endoscopic ultrasound showing diffusely enlarged pancreas with hypoechoic parenchyma and narrowed main pancreatic and common bile ducts.

fusely enlarged pancreas with hypoechoic parenchyma and narrowed main pancreatic and common bile ducts (Figure 3). Pancreatic fine needle biopsies (19-G ProCore [Cook Medical, Bloomington, IN] needle) showed lymphocytic and granulocytic cell infiltration (Figure 4). Serum immunoglobulin G4 (IgG4) levels were within normal range, and pancreatic IgG4 stain was negative.

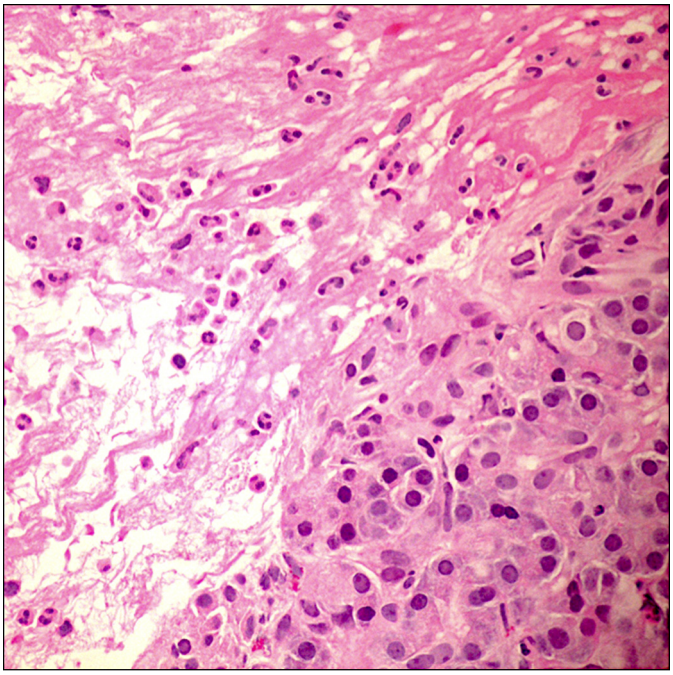

Figure 4. Pancreatic fine needle biopsy (19-G ProCore needle, hematoxylin and eosin, 100x) showing lymphocytic and granulocytic cell infiltration.

This is a rare case of simultaneous presentation of ulcerative colitis and idiopathic duct centric pancreatitis (IDCP). The patient was started on oral (4.5 g/day) and topic mesalamine and oral prednisolone (40 mg/day). One week later, complete resolution of symptoms was achieved and all laboratory findings returned to normal. During a 3-year follow up, ulcerative colitis remission was achieved using combination therapy with infliximab and azatioprine and no IDCP relapse was observed.

IDCP, previously known as type 2 autoimmune pancreatitis (AIP), is a rare and newly recognized pancreatic inflammatory disorder with a presumed autoimmune etiology. It is much less common than type 1 AIP and has a distinct histologic and clinical profile. It is characterized by neutrophilic infiltration within the lumen and epithelium of the pancreatic duct (granulocyte epithelial lesion). It is usually diagnosed at a younger age, does not have a gender predilection, and does not show serum IgG4 elevation. Definite diagnosis requires demonstration of granulocyte epithelial lesion on histology. It responds rapidly to steroid therapy, and relapses are rare.[1-3]

Recently, it has been established that inflammatory bowel disease (IBD) is more prevalent in AIP patients than in general population (6% of patients with proven AIP had a diagnosis of IBD).[2] This is particularly evident in IDCP, since it is associated with IBD in 25% of patients, predominantly with ulcerative colitis.[1] Therefore, in IBD patients with pancreatitis, it is mandatory to rule out IDCP.

DISCLOSURES

Authors contributions: S. Marques and J. Carmo wrote the manuscript. M. Bispo and P. Pinto-Marques revised the manuscript. S. Marques is the article guarantor.

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