Authors' contributions

A.R.C.: patient recruitment, data collection, literature review, writing up of the first draft of the paper, revision of the manuscript; L.R.A.: data collection, literature review, revision of the manuscript; F.R.M.: literature review, revision of the manuscript; J.G.: literature review, revision of the manuscript.

Data availability statement

Data cannot be shared for ethical/privacy reasons. Data available on request. The data underlying this short report cannot be shared publicly in order to protect the privacy of the individual. The data will be shared on reasonable request to the corresponding author.

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Conflicts of interest

Alexandra Ruiz-Cerulla has served as a speaker for Takeda.

Francisco Rodríguez-Moranta has served as a speaker for Abbvie, Takeda, Pfizer, Jansen, and MSD and has served as an advisor for Abbvie, Jansen, MSD, and Pfizer.

Lorena Rodríguez-Alonso has served as a speaker for Takeda, Pfizer and MSD and has served as an advisor for Abbvie.

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Usefulness of capsule endoscopy in idiopathic complex perianal disease

Utilidad de la cápsula endoscópica en la enfermedad perianal compleja idiopática

Crohn's disease (CD) can affect any segment of the gastrointestinal tract, with ileocolonic Crohn's being the most frequent (40%), followed by ileal (30%) and Crohn's of the colon (15%–30%). Exclusive involvement of the proximal small intestine, not accessible by ileocolonoscopy, occurs in only 10% of patients.[1,2] Furthermore, about 9% of patients present with perianal disease (PAD) at the time of diagnosis of CD, with incidence varying according to the intestinal location of the disease. Thus, it is 15% when it affects the ileum, 12% with ileocolonic involvement, 40% with involvement of the colon and 92% with rectal involvement.[3] However, the association of PAD in patients with CD exclusive to the upper gastrointestinal tract has been poorly described.

We present two cases of patients with PAD and normal colonoscopy, in whom the capsule endoscopy (CE) study detected lesions suggestive of CD. The endoscopic findings and the few published studies were reviewed.

Case 1

This was a 35-year-old woman with anal fissure and fistula of two years' evolution who underwent a sphincterotomy with persistence of an intersphincteric fistula visualised by pelvic MRI. Despite this, she presented with a torpid evolution, so she was referred in order to rule out CD. Clinically, she did not report digestive symptoms, she denied NSAID intake, and laboratory tests were strictly normal (including C-reactive protein and faecal calprotectin). In the ileocolonoscopy,

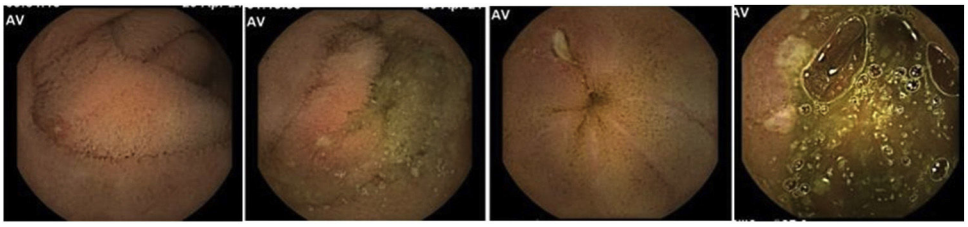

Figure 1 Capsule endoscopy frames in case 1.

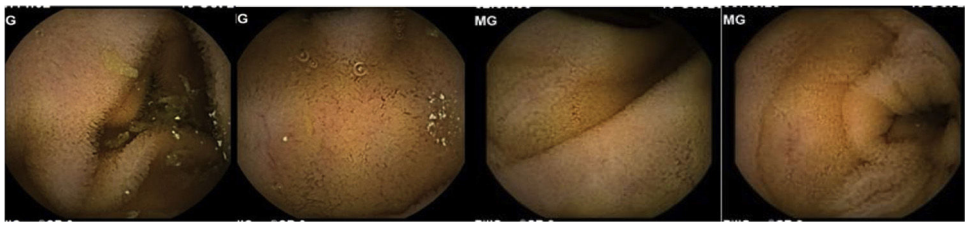

Figure 2 Capsule endoscopy frames in case 2.

four aphthous ulcers were observed in the terminal ileum, whereas the colon and rectal mucosa presented no alterations. In the ileal histology and of the different segments of the colon, a slight increase in the mixed inflammatory infiltrate in the lamina propria and foci of basal lymphoplasmacytosis were observed. The CE study identified a number of aphthous ulcers and erosions throughout the small intestine and three superficial ulcers, one in the mid jejunum and two in the distal ileum (Fig. 1). Both endoscopy and histology excluded other causes of PAD (infections, TB, neoplasia, etc.).

Case 2

This was a 38-year-old woman with a history of appendectomy of appendicular plastron with persistent abdominal pain. Seven years after surgery, she presented a perianal abscess that was drained and subsequently required a fistulotomy and the placement of setons and cavity curettage, and was referred in order to rule out CD. Clinically, there was diffuse mild abdominal pain, without other symptoms. The laboratory tests were strictly normal and treatment with NSAIDs was ruled out. The ileocolonoscopy did not identify any lesions and the magnetic resonance enterography was also normal. For this reason, the study was completed with CE, which showed multiple superficial and small-sized ulcerations throughout the small intestine; these findings, although non-specific, were suggestive of CD (Fig. 2).

In view of the findings of the CE, TNF inhibitor treatment with infliximab was proposed, since it is the biologic drug approved for PAD together with concomitant immunosuppressive agents in both patients. The clinical course with this treatment was optimal.

Limited data are available on proximal small intestine involvement in patients with CD and PAD without lesions detected on the ileocolonoscopy. Adler et al.,[4] in a prospective study involving 26 patients with PAD and a normal study with colonoscopy and imaging (CT/MRI enterography) who were examined by CE, small intestine lesions suggestive of CD were found in 24%. Subsequently, Xavier et al.,[5] in a retrospective study that included 71 CD patients who underwent CE and 17 of whom had PAD, found that patients with PAD had more lesions, particularly erosions, in the proximal intestine (94.1% vs. 66.6%; $P = .03$).

The CE also makes it possible to visualise the entire small intestine and is the exploration of choice in patients with suspected CD, normal ileocolonoscopy, and in the absence of obstructive clinical symptoms, having demonstrated greater sensitivity than radiological examinations to detect incipient and proximal lesions in the small intestine.[6] The association of PAD with CD of the colon has been described classically, and the true extent of this serious complication in patients with CD of the upper gastrointestinal tract is unknown,[3] and even less so when the intestinal involvement is exclusively in the proximal intestine. Therefore, in the presence of PAD with normal or inconclusive ileocolonoscopy and enterography, CE has a high negative predictive value for small intestine CD, making possible continuous and non-invasive study of the mucosa. In this sense, a meta-analysis has already demonstrated the significantly higher diagnostic performance of CE compared to enterography and even ileocolonoscopy.[7] Some studies have observed proximal small intestine lesions in up to 50% of CD patients when CE is utilised. The detection of lesions with CE in patients with normal ileocolonoscopy and magnetic resonance imaging may involve a change in decision-making in these patients, although the evolutionary, prognostic and management implications are yet to be determined when these lesions are so superficial that they are not detected in the enterography.[8] Despite this, and as demonstrated in the cases discussed, CE should be considered in certain clinical situations where CD is suspected but not demonstrated in conventional studies.

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