Anal Structure in Crohn’s Disease: Real Challenge Experience of a Medicine Department
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ABSTRACT
Crohn's disease is a chronic inflammatory disorder of the gastrointestinal tract. Perineal manifestations are a marker for disease severity. Anal strictures result from fibrotic scarring after several inflammatory attacks and are responsible for impaired quality of life. The aim of our work is to study the epidemiological profile and the management of these lesions. This is a descriptive retrospective study over a 17-year period from January 2002 to January 2019, concerning a monocentric cohort of 1053 cases of Crohn's disease. 22 patients had anal stricture, a prevalence rate of 2.08%. The main symptoms were painful passing of stools in all patients, emission of pus in 12 patients (54.5%) cases, imperiosity in 5 patients (22.7%), abdominal pain in 10 patients (45.4%) and an Obstructed defecation syndrome in 3 patients (13.6%). Penetrating phenotype B3 and the colonic localization L2 were the most frequent. Pelvic MRI confirmed anal stenosis in all patients, associated with complex fistulas, PARKS grade 4 in 18% (n = 4) cases. The treatment of the stenosis consisted of finger dilatation in 21 patients (90%), associated with dilation by Hegar dilators in 63.6% (n = 14) cases and by balloons in 31.8% (n = 7) cases; the stenosis biopsy revealed squamous cell carcinoma in only one case, 4.5%. The medical treatment for CD was based on immunosuppressants in 72.7% (n = 16) cases, anti TNF in 18.18% (n = 4) cases, and Combination therapy in 9% (n = 2) cases. The prevalence of anal strictures in our study remains low compared to literature series. These are young women with LAP, a Penetrating phenotype and pancolitis.

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Keywords
Crohn’s Disease, Gastrointestinal Tract.
Table 1. Characteristics of the study population

<table>
<thead>
<tr>
<th>Characteristic</th>
<th>n(%)</th>
</tr>
</thead>
<tbody>
<tr>
<td>Gender (M/F)</td>
<td>4 /18</td>
</tr>
<tr>
<td>Middle (age)</td>
<td>37 years +/- 9.3</td>
</tr>
<tr>
<td>B2- stricturing</td>
<td>10 (45.45%)</td>
</tr>
<tr>
<td>B3- penetrating</td>
<td>12 (54.5%)</td>
</tr>
<tr>
<td>L1-ileal</td>
<td>3 (13.6%)</td>
</tr>
<tr>
<td>L2-colonic</td>
<td>10 (45%)</td>
</tr>
<tr>
<td>L3-ileocolonic</td>
<td>9 (41%)</td>
</tr>
<tr>
<td>stricture</td>
<td>22(2.08%)</td>
</tr>
<tr>
<td>S1</td>
<td>7 (31.8%)</td>
</tr>
<tr>
<td>S2</td>
<td>15(68.2%)</td>
</tr>
<tr>
<td>Ulcers</td>
<td></td>
</tr>
<tr>
<td>U1</td>
<td>5 (83.3%)</td>
</tr>
<tr>
<td>U2</td>
<td>1 (16.6%)</td>
</tr>
<tr>
<td>Fistula</td>
<td></td>
</tr>
<tr>
<td>F1</td>
<td>4 (30.7%)</td>
</tr>
<tr>
<td>F2</td>
<td>9 (69.2%)</td>
</tr>
<tr>
<td>Medical treatment before dilatation</td>
<td></td>
</tr>
<tr>
<td>Anti TNF</td>
<td>4 (18.8%)</td>
</tr>
<tr>
<td>Immunosuppressant (AZA or 6MP)</td>
<td>16 (72.72%)</td>
</tr>
<tr>
<td>Combination therapy</td>
<td>2 (9%)</td>
</tr>
<tr>
<td>Colectomy (colonic stricture)</td>
<td>3 (13.63%)</td>
</tr>
<tr>
<td>Anal dilation (finger, bougies, balloon)</td>
<td>21 (95.4%)</td>
</tr>
<tr>
<td>Second dilation</td>
<td>4 (18%)</td>
</tr>
<tr>
<td>improved medical treatment.</td>
<td>16 (72.7%)</td>
</tr>
<tr>
<td>Definitive stoma</td>
<td>1 (4.54%)</td>
</tr>
</tbody>
</table>

Fig 1. Pseudocondylomatous tumors, with 2 major ulcers and fibrous stricture admitting the fingertip (S1).

Fig 2. Anal fistulas with pseudocondylomatous tumors and anal stricture (S2).

The penetrating phenotype B3 and the colonic localization L2 were the most frequent with, respectively, 12 cases (54.5%) and 10 cases (45.4%). All our patients benefited from pelvic MRI, which showed fibrous anal stenosis in 95.45% (n = 21) cases, complex fistulas in 41% (n = 9) cases, and locally advanced rectal cancer in a single case, in 4.5% of patients. None of our patients have had endoscopic ultrasound because of anal stenosis. The stricture biopsy made systematically revealed a case of squamous cell carcinoma. The medical treatment of CD was immunosuppressants in 72.72% (n = 16) cases, anti TNF in 18.18% (n = 4) cases, and combination therapy in 2 (9%) patients. Specific stenosis treatment was dilatation with Hegar dilator in 66.6% (n = 14) cases, and balloon in 33.3% (n = 7) cases (Fig 3) associated in both cases with maintenance finger dilatation in 21 patients (95.45%). All patients with associated active fistula benefited from drainage with antibiotic therapy based on metronidazole and ciprofloxacin. 3 patients (13.63%) had an associated colonic stenosis, they underwent colectomy with restoration of continuity, the medical treatment consisted for the patient who had an inflammatory stenosis, in addition of Anti TNF, after a few months from the start of treatment, clinical improvement was noted. 16 patients received optimized medical treatment. 16 patients improved after the first dilation session, one patient bled after balloon dilation. 4 patients had a recurrence of stenosis, a rate of 18%. For the latter benefit from a second dilation session, three patients responded. Definitive stoma was performed in one patient.

Fig 3. Endoscopic view of anal stricture dilatation with hydrostatic balloon (Before dilatation)

Fig3. Endoscopic view of anal stricture dilatation by a hydrostatic balloon (After dilatation)

Discussion

Perineal Crohn's disease (PCD) includes non-fistulizing manifestations (cracks, ulcers, and strictures) and fistulizing lesions (fistulas, abscesses and rectovaginal fistulas). Superficial cracks represented 21 to 35% of perineal lesions, anal and rectal ulcers 5 to 10%, and anorectal strictures 9 to 22% (I).
In 1992, the Cardiff classification classified strictures on a scale from 0 to 2 (0 = no stricture, 1 = reversible stricture, 2 = irreversible stricture) and according to the location of the stenosis, rectal or anal. S1 strictures are usually inflammatory, due to spasms, so S2 results from fibrous tissue.

In Brochard’s study (2005 -2013) including 102 patients who had anorectal strictures, the male / female ratio was 37/65. 63.7% of women with an average age of 41.2 years (2), a predominance in the middle-aged adult women that we find in our study.

The main symptoms of anal stricture include anal pain, rectal bleeding, and constipation. Rarely, patients report stools or diarrhea. Bowel pain remains the main symptom in Brochard’s study, found in 51% of patients. (3) and in 69% of cases in another study (4).

Association of different anoperineal lesions is frequent, in a series including 282 patients, 94% of the strictures was associated with other lesions (n = 46/49) (5), Other studies have shown a high prevalence between anal fistula and anorectal stricture. (6)

The Penetrating phenotype and colonic location were predominant, however, the literature does not show a relationship between the luminal phenotype and perianal Crohn's disease phenotype.

Stenosis occurs after a long course of Crohn’s disease. Initially, some studies suggested that the appearance of narrowing / anal strictures was the result of the healing process of inflammatory lesions (7,8). The role of biotherapies has been suspected for a time in the appearance or worsening of anorectal strictures, however, an underanalysis based on the TREAT register (the Crohn's Therapy, Resource, Evaluation, and Assessment Tool) has shown that the risk of complications were similar in patients who received and did not receive infliximab (9).

Therapeutic management of Crohnian anal stenosis requires an endoscopic and radiological assessment beforehand, preferably by pelvic MRI. Mainly to characterize the length, the inflammatory or fibrous nature of the stenosis, and the presence of another perineal lesion (10,11). In fact, the therapeutic attitude differs, depending on the inflammatory or fibrous nature of the stenosis. Concerning inflammatory strictures, basic treatment optimization is necessary. (12)

Data from different European cohorts suggest a clinical advantage with anti TNF in Crohnian stricture (13,14,1,15,16).

In Brochard's study, the adjustment of the background therapy allowed a cure rate of anorectal strictures in more than 50% of patients after a follow-up period of 3 years. In our series, all the patients who had had an optimization of the background therapy have progressed well.

In the study by Bouguen et al. (1), out of 22 patients on infliximab with anorectal Crohn's stricture, 12 had a complete stricture regression after an average follow-up of 175 weeks: only 6 had anal dilation. It was unclear whether this was type 1 or 2 stenosis. An additional reported case described the use of vedolizumab in a patient with Crohn’s disease complicated by anorectal stenosis with good evolution (17).

For short fibrous anal or anorectal strictures, no currently approved medication is known to prevent or reverse established fibrosis. (18), dilation is the treatment of choice, it can be carried out simply with the finger, with a bougie dilatator (Hegar dilatators or metal olive dilatators) (19), or with balloons under general or locoregional anesthesia. A study on ten patients with S2 fibrous anal stenosis due to CD dilated by candles showed long-term efficacy (20). This method provides tactile feedback, which makes it possible to estimate the degree of resistance to the dilator passage, and thus avoid overdilation. Another study comparing bougies versus balloons in the treatment of benign postoperative rectal strictures, in a total of 39 patients did not objectify a statistically significant difference between the two groups concerning the success rate, major complications, number of necessary dilations, or recurrence, the only advantage was the low cost of the bougies (21). In fact, unlike balloons, bougies are reusable.

The interval between dilations depend on the clinical examination. A new dilation is proposed as soon as a recurrence is noted without waiting for symptoms appearance.

Regarding complications, the risk of anal incontinence after anal dilatation is high due to fibrosis, which is often associated with destruction of the anal sphincter. There is no hemorrhage, perforation, or serious undesirable effects described after anal dilatation in the literature, subject to very poor literature, on the other hand, it must be carried out judiciously because of a risk of sepsis. (20) Only one case of bleeding was noted in our series.

If treatment by dilation fails, and before resorting to an ano-proctectomy, other conservative surgical alternatives may be considered. (22,23,5) In the event of short anal or low rectal stenosis, two techniques of plasty can be proposed. One is to vertically incise the stenosed area and then suture the banks transversely as for intestinal stricturoplasty. The other is to excise the stenotic fibrous area over part of the circumference and to cover the defect with a rectal advancement flap. These techniques are only possible in the absence of inflammatory luminal involvement and are in fact rarely performed. They have not been evaluated to date.

![Fig 4. Management of anorectal stenosis associated with Crohn’s disease.](image-url)
The risk of anal cancer in Crohn's disease remains underestimated. In a systematic literature review, published by Slesser et al (24), the incidence of anal cancer in Crohnian patients was (2.0 / 100,000), mainly Crohn's disease with LAP 85%.

Another recent controlled study of 53,568 patients with UC and CD indicated that the prevalence of cancer was higher in patients with stenosis (25). This requires regularly monitoring for patients with crohn’s anal lesions, and to perform biopsies (26) (Fig4)

In case of stenosis with ano-perineal supputation, it is recommended before dilating to treat luminal disease and associated ano-perineal lesions by optimizing background treatment and draining the suppurations.

Conclusion

Anal stenosis usually occurs after several flare-ups inflammatory and is often late during the disease. The therapeutic strategy requires medico-surgical concentration.

The increased risk of dysplasia in these patients requires regular monitoring with biopsies.

In the case of inflammatory stenosis, a basic treatment of CD is proposed as a first intention before dilation in the event of failure.

Dilatation is proposed as a first line in case of fibrous, symptomatic and short stenosis or when it makes endoscopic monitoring impossible and / or when it does not allow the drainage of suppuration.

References

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