Ultra-low anterior resection with coloanal anastomosis for recurrent rectal prolapse in a young woman with colitis cystica profunda

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ABSTRACT

This case demonstrates the successful treatment of a young female patient with colitis cystica profunda causing rectal prolapse, after primary treatment with a Delorme procedure had failed. An ultra-low anterior resection with a temporary defunctioning ileostomy was carried out with good postoperative results. This case illustrates the possibility of carrying out sphincter preserving surgery rather than an abdominoperineal resection in the treatment of this condition, which may be preferable for patients.

KEYWORDS

Colitis cystica profunda – Rectal prolapse – Anterior resection

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Colitis cystica profunda (CCP) is a rare benign condition that was first described by Virchow in 1863. Fewer than 200 cases have been reported in the literature. It is characterised histologically by the formation of multiple submucosal cysts (Fig 1), with the muscularis propria and serosa rarely affected. Macroscopically, at colonoscopy it may appear erythematous, polypoid or ulcerated, and can appear as a mass and mimic malignant lesions.¹ It most commonly affects the rectum, particularly the anterior wall, but may be found anywhere in the colon.²

It shares histological features with solitary rectal ulcer syndrome (SRUS) and the overlap between these conditions is not fully understood. Some authors have proposed that it may be ischaemic, inflammatory or post-traumatic in aetiology.³ However, given the frequent association with rectal prolapse, it has been suggested that these disorders all fall into the spectrum of one disease process.⁴ In an effort to encompass both CCP and SRUS, du Boulay proposed the term ‘mucosal prolapse syndrome’ but this term does not seem to be widely used today.

Patients may present with a variety of symptoms, the most common being rectal bleeding, mucus discharge, tenesmus, abdominal pain and constipation. Symptoms of obstructed defecation are present in many patients and the incidence of demonstrable prolapse has been reported as being as high as 54% by some authors.⁵ The average age of presentation is around 30 years (range: 4–76 years), with males and females equally affected.⁵ Treatment depends on the severity of symptoms but generally focuses on correction of obstructed defecation. Some patients may improve with laxatives and

dietary modification or biofeedback but others may require a surgical intervention either via a perineal (Delorme/Altemeier) or abdominal (rectopexy) approach. When these treatments fail, more radical procedures may become necessary, most commonly abdominoperineal resection or a permanent defunctioning colostomy.⁶

We present the case of a young female patient with CCP who underwent successful definitive surgery with an ultra-low anterior resection after conservative measures and less radical surgery had failed.

Case History

A 28-year-old, nulliparous woman with no past medical history presented in 2009 with longstanding symptoms of obstructed defecation. Her main symptoms were straining to pass stools with multiple attempts per day as well as passing fresh blood per rectum. She was assessed initially in another specialist centre and after a pelvic floor workup, a stapled transanal rectal resection was planned to correct a symptomatic high grade internal rectal prolapse. However, examination on the day of surgery revealed that the mucosa was significantly thickened and appeared inflamed. The procedure was therefore abandoned. At this point, the patient decided she did not want to pursue any further treatment.

She presented to our unit in early 2010 with ongoing symptoms and on clinical examination, a full-thickness rectal prolapse was demonstrated. A colonoscopy was essentially normal, and repeat biopsies showed inflammation and fibrosis with non-specific changes in the rectal mucosa. The

options of either a Delorme procedure or a laparoscopic ventral mesh rectopexy were discussed with the patient. Given her young age and the fact that she had not yet started a family, a perineal approach was thought to be preferable in the first instance while accepting a higher risk of recurrence. A Delorme procedure was carried out in March 2010 and at the time of surgery, she had significant inflammation of the rectal mucosa, particularly of the anterior rectal wall with visible submucosal cysts. Histology of the stripped mucosa subsequently confirmed CCP.

The patient had an uneventful postoperative recovery and complete resolution of her symptoms. Unfortunately, she re-presented in January 2012 with new symptoms of obstructed defecation syndrome after a first pregnancy. Repeat flexible sigmoidoscopy and magnetic resonance imaging showed similar features to her initial presentation with inflammation extending from 1cm above the dentate line and continuing 4cm proximally.

She was referred to the regional tertiary referral centre and discussed in their pelvic floor multidisciplinary meeting.

It was felt that she would need an abdominoperineal resection. The patient was unwilling to have an operation resulting in a permanent stoma, and was therefore given the option of a laparoscopic ultra-low anterior resection with a coloanal anastomosis and temporary defunctioning loop ileostomy. This was carried out uneventfully in October 2012. Postoperative histology confirmed CCP with clear resection margins. In January 2013 she was readmitted for closure of her ileostomy following a water soluble contrast enema that revealed an intact coloanal anastomosis. She was seen recently in clinic and had a follow-up colonoscopy (August 2013), which showed no new abnormalities. She has good bowel function and no ongoing obstructed defecation syndrome symptoms.

Discussion

This case highlights the possibility of carrying out restorative sphincter conserving surgery in selected patients with CCP causing prolapse and obstructed defecation in whom less radical surgery such as a Delorme procedure has failed. Previous studies have reported a low success rate when anterior resection is carried out after other surgery has failed and suggest that a defunctioning colostomy may be a better option.⁷ However, owing to the rarity of the condition, only small numbers of patients have ever been studied and no prospective data are available.

Conclusions

In young patients who find the concept of a permanent colostomy unacceptable, we feel that restorative surgery should be considered. Thorough discussion with patients regarding the likely complications and functional outcome is of paramount importance at an early stage so all options can be considered.

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