CLINICAL CASE

Fibrin glue sealing in the treatment of a recto-urethral fistula in Crohn’s disease: A case report

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Summary Recto-urethral fistulas in Crohn’s disease are rare, and managing them is difficult. The various surgical techniques are not reliably effective and are associated with a significant risk of morbidity. The rectal mucosal transposition flap technique, which is used most frequently, requires the rectal mucosa to be in a healthy condition. We report here on a case where treatment was by injecting fibrin glue into a complex fistula with a single anorectal point of origin but combining a median recto-urethrocutanéo-rectoperineal tract with two deep lateral rectoperineal tracts. The patient had presented with active rectal Crohn’s disease. This treatment produced complete closure, verified by MRI, of all the fistula tracts, which was still maintained after three years, and with normal anal continence. When confronted with this type of fistula, and particularly when the condition of the rectal mucosa is poor, the specialist should be encouraged by this good result to consider the injection of fibrin glue, a technique without risk of morbidity, as a first course of action.

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Résumé Les fistules recto-urétrales de la maladie de Crohn sont rares et de prise en charge difficile. L’efficacité des différentes techniques chirurgicales est inconstante et associée à un risque de morbidité non négligeable. Le lambeau muqueux rectal d’abaissement, la technique la plus employée, nécessite l’intégrité de la muqueuse rectale. Nous rapportons un cas de traitement par l’injection de colle biologique d’une fistule complexe, à point de départ anorectal unique, associant un trajet médian rectocutanéo-urétro-rectoperineal à deux trajets latéraux profonds...
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Introduction

Recto-urethral fistulas in Crohn’s disease are rare, and managing them is difficult [1—12]. The various surgical techniques are not reliably effective and are associated with a significant risk of morbidity. The rectal mucosal transposition flap technique, which is used most frequently, requires the rectal mucosa to be in a healthy condition [1,5,8]. We report here on a case where treatment was by injecting fibrin glue into a complex Crohn’s fistula with a single ano-rectal point of origin but combining a median recto-urethrococutaneous tract with two deep, more lateral, rectoperineal tracts in a patient with active colorectal Crohn’s disease.

Case report

A 40-year-old man presented in 1996 with a perineal abscess which was laid open without draining. Ten months afterwards, Crohn’s disease was diagnosed, with localised rectal and colon involvement, and complex anterior anoperineal suppurative processes; three fistula tracts were laid open. The patient was lost to follow-up until September 2002, when he once again presented with colon and rectal lesions observed by colonoscopy to be predominantly in the sigmoid, with anoperineal involvement. Fistulography showed three anterior perineal fistula tracts converging as they crossed the anal sphincters into a single tract communicating with the rectum. The medical treatment (metronidazole and 5-aminosalicylic acid) improved the symptoms of the colon disease. Two months later pneumaturia developed. During an examination under spinal anaesthesia in January 2003, the following were observed:

• a median fistula tract, the external opening of which was at the base of the scrotum;
• two other tracts orientated laterally then opening into the anterior section of each ischioanal fossa;
• a single point of origin (common internal opening) situated in ulceration anterior to the anorectal junction.

After occluding the three external cutaneous orifices, methylene blue was injected through the internal orifice. Its reappearance in the urinary lumen confirmed communication with the urinary apparatus. In order to drain them, non-cutting setons were placed in these three tracts which crossed the upper part of the anal sphincters. Magnetic resonance imaging (MRI) confirmed the existence of a recto-urethral fistula involving membranous urethra, developing from the median fistula tract and communicating with the external orifice situated at the base of the scrotum via a complex bifurcated tract (Fig. 1).

In March 2003, this patient, who weighed 65 kg, was given azathioprine (150 mg per day). From June to July 2003 the patient received three infusions of infliximab (5 mg/kg). The setons were removed between the 1st and 2nd infusion. Infliximab did not induced closure of any fistula and was not

Figure 1 Initial MRI. T2 sagittal and axial sections. The anal canal is shown by a small catheter containing water (water appears as a white hypersignal on T2-weighted images); a: Sagittal section showing a portion of the anterior tract of the fistula (white arrow) extending towards the membranous urethra (the prostate is indicated by the white star, the bladder by the black star); b: Axial section passing through the inferior part of the anal canal showing two anterior tracts (white arrows) situated between the anal canal and the root of the penis (corpus spongiosum = white diamond). The ischia are indicated by the black diamonds.
Figure 2  MRI after treatment: T2 sagittal (a) and axial (b) sections. The fistula tracts have disappeared. There is just fibrosis, observed as a hyposignal, with the root of the penis appearing deformed (white arrow). The prostate is indicated by the white star, the bladder by the black star, the ischia by the black diamonds, and the penis by the white diamond.

used again. One month later a suprapubic cystocatheter was inserted because of urinary retention involving an infection.

In November 2003, a urinary catheter had to be put in place owing to complete inflammatory obstruction of the bulbomembranous urethra. Two weeks later, a cystoscopic examination (under spinal anaesthesia) showed that the inflammatory obstruction, located at the point where the fistula opened into the urethra, had partly disappeared. Moderately active proctitis was observed. During the same procedure, the medial fistula tract was catheterised, starting from the anorectal ulceration and going as far as the urethral opening, using a catheter mounted on a semi-rigid guide (Duplocath 35°, Tissucol Kit®), as were the two lateral fistula tracts, using a flexible catheter (Duplocath 25°, Tissucol Kit®) and starting from the points where they opened to the exterior. As these catheters were gradually withdrawn, fibrin glue (Tissucol®, Baxter, Maurepas, France) was injected (2–3 mL per fistulous tract). Glue issuing through the urethral opening was monitored by simultaneous cystoscopy. At the end of the procedure, a trans-urethral double-lumen catheter was put in place. Bed-rest was strictly enforced for the two hours following the procedure.

Only the part of the tract adjacent to the urethral opening appeared to be sealed, as no pneumaturia recurred when the urinary catheter was withdrawn two weeks later, but the three cutaneous openings still leaked. There was still moderate proctitis. In February 2004 the three remaining tracts were again drained by setons inserted into the three cutaneous openings, and issuing through the common anorectal opening. A new attempt was made in September 2004 to inject glue, using the same technique as in the first procedure.

In November 2005, MRI revealed no fistulous tracts at all (Fig. 2). At the end of the follow-up period, in October 2007, the patient still receiving azathioprine, had no pneumaturia, no urination disorder, no urinary infection, or perineal wetness. Anal continence was normal. During the clinical examination, there was a complete absence of any form of anorectal/perineal suppuration with total closure of both the external openings and of the internal anorectal opening. In December 2007, the patient died during a massive digestive haemorrhage. No autopsy was performed.

Discussion

Recto-urethral fistulas are rare in Crohn’s disease; less than 30 cases have been reported in English and French literature [1–12].

Their treatment, for which there are no agreed recommendations, is difficult: it ranges from no treatment to proctectomy with coloanal anastomosis, or even abdominoperineal resection of the rectum. Giving no treatment appears to be justified when the symptoms are unobtrusive, because theoretically there is no risk of ascending urinary tract infection, owing to the action of the urinary sphincters [1]. No medical treatment was offered, except for metronidazole in two cases which were given inconsequentially and to no effect [2,10]. To our knowledge, no one has reported using anti-TNF-alpha antibodies for this indication. The choice of surgical technique is guided by the condition of the rectal mucosa. The rectal mucosal transposition flap technique is the technique most used when there are no active lesions of the rectal mucosa, with seven cases reported [1,5,8]. Establishing a temporary faecal or combined (faecal and urinary) diversion prior to performing this technique seems to favourably influence the results [8]. The result of one of the seven cases is not specified [5]. For two of the remaining six cases the rectal mucosal transposition flap technique was a failure, but was effective in four patients, with one or two procedures being necessary, this efficacy being observed after 46 months in one case [1], and an imprecisely specified period of time in the other three cases [8].

Other surgical techniques are reported which are however by no means always effective nor without risk of morbidity: primary repair via various approaches [1,2,4,9], cutaneous advancement flap repair, on its own [8] or combined with other surgical procedures [1,2], gracilis muscle...
transposition [7], proctectomy with coloanal anastomosis [8], abdominoperineal resection of the rectum [3,12], and isolated urinary (cystostomy) or digestive (ileostomy, colostomy) diversions [1,2,4,6,11].

Seven observations of recto-urethral fistulas not linked to Crohn’s disease and treated by the injection of fibrin glue have been reported [13–15]. Of the six cases treated solely by injection of fibrin glue, two involving urethrovesicorectal fistulas proved to be failures [13], while the results are not given individually for the other four, who were part of a series of 37 anal fistulas treated using this technique [14]. The seventh case was successfully treated by a combination of endoscopic fibrin glue injection and clipping, via the endorectal route [15].

We chose the injection of fibrin glue for our patient because:

- the three perfusions of infliximab had clearly been ineffective; moreover in the prospective study by Present et al. [16] the median time to achieve complete closure of the fistulous tracts was two weeks after the end of treatment;
- there were active lesions of the anal and rectal mucosa;
- there is no risk of morbidity with this technique [17];
- it would be possible to use all the other techniques available later, if it failed.

This good result was probably favourably influenced by the prolonged preliminary draining and by the perfusions of infliximab. Indeed, infliximab reduces local inflammatory activity, and thus, along with drainage, forms a stage in the medical/surgical strategy for managing complex anal fistulas in Crohn’s disease [18,19].

Conclusion

After three years and with the closure of all the tracts confirmed by MRI, we are encouraged by this good result to offer the injection of fibrin glue, following, at the least, prolonged draining, and possibly the use of infliximab, as first line treatment for recto-urethral fistulas in Crohn’s disease. The injection of fibrin glue causes no morbidity and does not, should it fail, prevent later use of other surgical forms of treatment, the efficacy of which is less reliable and the risk of its causing potential morbidity, higher.

Conflict of interests

None.

References