CASE REPORT

Refractory diversion neovaginitis in a sigmoid-colon-derived neovagina: clinical and histopathological considerations

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ABSTRACT

Colonic segments are being used as pedicled grafts in neovaginoplasty, a surgical procedure to (re)construct a (neo)vagina. A disadvantage of using colonic grafts is the potential occurrence of neovaginal complications due to diversion from the faecal stream. Here, we report a case of severe, refractory diversion colitis of the sigmoid neovagina, so-called 'diversion neovaginitis', in a 42-year-old woman with complete androgen insensitivity syndrome. Neovaginal biopsy specimens showed colonic-type mucosa with strong increase of lymphoplasmacellular infiltrate in the lamina propria, ulceration with fibrinoid deposition and some crypt irregularity. Endoscopy showed erythematous mucosa, superficial ulceration, mucus discharge and multiple pseudopolyp-like lesions. Local application of mesalazine foam enemas and sodium butyrate enemas initially gave symptom relief. However, this was a temporary effect, ultimately necessitating removal of the neovaginal construct. It is important that all patients are informed about neovaginal bowel complications, for example, diversion neovaginitis. Regular medical and endoscopic follow-up appears recommendable.

BACKGROUND

Neovaginoplasty, the surgical construction of a vagina, is performed in male-to-female transgenders and biological women with congenital or acquired absence of a functional vagina.¹ In sigmoid neovaginoplasty, a segment of the sigmoid colon is mobilised, isolated and transferred caudally on its vascular pedicle to form the neovaginal cavity. Using an intestinal segment for this

procedure has advantages when compared with other types of grafts, such as inverted penoscrotal skin, peritoneal tissue or pedicled regional skin flaps. The intestinal segment provides natural lubrication, adequate neovaginal depth and has little tendency to shrink and form stenosis.¹ The main disadvantages of using an intestinal segment as graft comprise the necessity to perform intestinal surgery and the risk of neovaginal complications due to diversion from the faecal stream.^{1 2} As such, diversion of colonic tissue may lead to diversion colitis (DC), a sometimes clinically challenging entity. DC is inflammation of a diverted, defunctionalised colonic segment, first recognised in remaining distal colonic segments following colostomy or ileostomy.³ Most patients do not experience symptoms, but symptoms may include mucous discharge, bleeding, pain and tenesmus.⁴ In sigmoid vaginoplasty, the intestinal segment is diverted from the faecal stream, however, not entirely defunctionalised because operated subjects are able to engage in neovaginal penetrative sexual intercourse. Mild DC of the sigmoid neovagina has been sporadically reported.^{2 5–7} In this case report, we focus on the histopathological and endoscopic features and management of a severely active diversion neovaginitis, being refractory to standard medical treatment.

CASE REPORT

A 42-year-old woman with complete androgen insensitivity syndrome was

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referred to our hospital with neovaginal pain and malodorous neovaginal discharge since 1 year. Her surgical history included an orchidectomy, a bilateral breast augmentation, both at the age of 16, and a sigmoid neovaginoplasty via laparotomy, at the age of 28, all performed elsewhere as part of the gender-assignment process. She had comorbid mineralocorticoid hypertension and used antihypertensives (amlodipine and perindopril). She had no history of chronic (inflammatory) bowel disease. She was sexually attracted towards men, and had been sexually active with one male partner the last year, but her symptoms were impeding a healthy and satisfying sexual life. Physical examination of the neovagina showed brown, viscous, malodorous neovaginal discharge. The speculum examination was painful. The insertion depth of the neovagina was 20 cm. Neovaginal endoscopy, performed and evaluated by a gastroenterologist experienced in neovaginal endoscopy, showed multiple pseudopolyp-like lesions and mildly erythematous mucosa, as commonly ascribed to chronically active inflammation of the mucosa. There were no signs of active bleeding. Concurrent sigmoidoscopy revealed no concurrent abnormalities of the remaining proctosigmoid. Random biopsies were taken of the sigmoid neovagina and histopathologically examined: colonic tissue with chronic, focally active, erosive inflammation was observed (figure 1). DC of the sigmoid neovagina, ‘diversion neovaginitis’, was diagnosed. Treatment consisted of neovaginally applied mesalazine foam enemas (Salofalk, 1 g/application, Dr Falk Pharma GmbH, Freiburg, Germany) administered every other day and sodium butyrate enemas (50 mL of 40 mM) daily. We started with this combination of local therapies rather than solely mesalazine foam enemas due to the severity and duration of her symptoms.

Initially, this induced symptom relief; she reported a reduction of neovaginal pain and discharge. However, the patient reported symptom recurrence 4 months later, despite ongoing intensive enema treatment (mesalazine foam enemas administered every other day and sodium butyrate enemas daily), to which she adhered consistently. Neovaginal endoscopy revealed (again) multiple pseudopolyps and mucosal erythema (figure 2A). Histological examination showed comparable inflammatory alterations as before, with similar signs of active and chronic inflammation. Sodium butyrate enemas were continued and cotherapy with budesonide foam enemas (Budenofalk, 2 mg/application, Dr Falk Pharma GmbH, Freiburg, Germany) and isotonic flushing of the neovaginal cavity was initiated. This intensified concomitant therapy did not induce symptom improvement in the subsequent period. To the contrary, in the following 3 months, symptoms worsened, as did the inflammatory aspect of the neovagina during endoscopy (figure 2B).

Because of the severity of symptoms and lack of improvement after medical therapy, the sigmoid

Figure 1 Histological examination of H&E-stained biopsy specimens of the sigmoid neovagina at presentation (A) and 4 months thereafter (B). Both biopsy specimens show colonic-type mucosa with strong increase of lymphoplasmacellular infiltrate in the lamina propria. (A) shows ulceration with fibrinoid deposition, (B) shows some crypt irregularity, consistent with chronic inflammation (comparable with inflammatory bowel disease histology).

neovagina had to be surgically removed, and a laparoscopic ileal neovaginoplasty was performed with an ileal segment of approximately 15 cm in the same session. Perioperative stress doses of corticosteroids were administered. One year postoperatively, the patient was symptom-free. The ileal neovagina had an insertion depth of 18 cm.

DISCUSSION

Multiple surgical techniques exist for vaginal (re)construction, each with its own advantages and

Figure 2 Neovaginal endoscopy performed 4 months (A) and 7 months (B) after initial presentation. (A) Erythematous mucosa, superficial ulceration, mucus-like discharge and multiple pseudopolyp-like lesions throughout the sigmoid-derived neovagina. (B) Lack of endoscopic improvement. The neovagina remained inflamed. Multiple pseudopolyps were observed in both the proximal and distal part of the neovagina, apparently to a greater extent as before. Clinically, a malodorous, brown discharge was prominent.

disadvantages.¹ For male-to-female transgenders, the most commonly used approach is penile skin inversion vaginoplasty in which a neovagina is created from an inverted pedicled penile skin flap. Postoperative intermittent dilatation is necessary to prevent neovaginal stenosis. This technique is not an option when insufficient penile skin is available. Other types of graft that can be used in vaginoplasty are non-genital skin flaps and pedicled intestinal segments.

Although DC following colostomy or ileostomy has extensively been studied, the exact aetiology remains unclear. Insufficient supply of luminal nutrients, especially short-chain fatty acids (SCFA), a crucial energy

source for colonocytes, seems to play a critical role.⁸ Deprivation of SCFA is associated with apoptosis of colonocytes (starvation), subsequently leading to intestinal epithelial barrier disruption, inducing inflammatory mucosal responses. The histopathological abnormalities observed in DC include mild-to-moderate chronic inflammation, atrophy, villous colonic surface, mild crypt architectural abnormalities, crypt abscesses, aphthous ulcers and sometimes follicular lymphoid hyperplasia.⁹ Though literature is scarce, the histological specificities of DC seem comparable with those seen in cases of diversion neovaginitis.^{2 5-7} During endoscopic examination of DC, mucosal friability, contact bleeding, mucous discharge, erythema, oedema, ulceration, filiform polyps and absence of mucosal vascular pattern may be observed.⁴ Endoscopic or histopathological signs corroborating the diagnosis of DC may be found in the majority of subjects after diverting surgery, however, remarkably, only approximately 30% of patients experience symptoms.¹⁰ Therefore, presence or severity of symptoms is not necessarily correlated with endoscopic or histopathological findings. If these data may be applied to the sigmoid-derived neovagina remains unclear. Traditionally, surgical restoration of bowel continuity and thereby the faecal stream is the preferred treatment option for DC. However, for obvious reasons, this is an undesirable option in patients with a sigmoid neovaginoplasty. SCFA or 5-ASA enemas and (local) glucocorticoids have sometimes been shown to be effective for both DC as diversion neovaginitis.^{2 5-7} Although adequate neovaginal depth is generally described as an advantage of intestinal vaginoplasty, an increased depth may be associated with increased mucus production, subsequently leading to stasis of neovaginal mucus and episodes of excessive, usually malodorous, mucus loss. Some authors claim that a shorter interposed intestinal segment may be preferred due to a less inconvenient neovaginal mucous drainage.^{2 11}

Based on limited evidence from retrospective studies, intestinal neovaginoplasty appears to be a safe procedure with few perioperative and postoperative complications.¹ The incidence and prevalence of diversion neovaginitis are unknown.^{2 5-7} It is important that all patients are informed about neovaginal bowel complications, for example, diversion neovaginitis, and regular medical follow-up appears recommendable to monitor symptoms and check for these complications. Currently, data on endoscopic and histopathological changes of the sigmoid colon used in a neovaginoplasty procedure are lacking, as are the implications of these findings for treatment and functional prognosis of the neovagina.

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