



Modified Turnbull–Cutait anastomosis with fasciocutaneous flap reconstruction for radical excision of vulvar and anal intraepithelial neoplasia

A. C. Rogers¹ · M. Brophy¹ · T. A. Walsh¹ · R. P. Hanson¹ · A. E. Brannigan¹

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Introduction

Intraepithelial neoplasia of the lower genital tract, perineum and perianal area is most often linked to the prevalence of human papillomavirus (HPV) infection [1]. HPV predisposes to precancerous lesions involving the cervix, but less commonly the vagina, vulva and anus. Left untreated, anal intraepithelial neoplasia (AIN) and vulvar intraepithelial neoplasia (VIN) can progress to invasive cancer; with long-term progression of approximately 10% [2]; risk is highest in those who smoke, have multifocal disease or are immunocompromised [1].

Treatment strategies vary, and range from watchful waiting to radical excisional surgery, but irrespective of modality, up to 50% of patients will develop recurrence [3]. Recurrence is significantly lower with clear surgical margins [3], although lymph node clearance is not necessary.

In the current study, we report a novel therapeutic strategy for extensive multifocal high-grade intraepithelial neoplasia involving complete surgical resection of disease with perineal reconstruction and formation of a neo-anal opening.

Case history

A 37-year-old woman attended for surveillance of VIN, with a previous diagnosis of human immunodeficiency virus (HIV). Despite multiple attempts at topical and ablative therapy, the VIN remained high grade (VIN 3), and progressed to involve her entire vulva, perineum and perianal region.

Biopsies of the anal region confirmed AIN 3 circumferentially and to the dentate line (Fig. 1).

In consultation with the patient and multidisciplinary team members, a decision was made to surgically resect the volume of disease, requiring multimodal input from gynaecology, colorectal and plastic surgery teams. The patient requested an alternative to a non-restorative resection, which would normally involve removing her anal skin and dentate line with an abdominoperineal resection. To facilitate a clear anorectal margin, a rectal mobilisation is required; however, her perianal and perineal skin also required resection and reconstruction, rendering an anastomosis of her rectum to her reconstructed neo-anal skin difficult. Thus, after an extensive skin resection, her perineum and neo-anus were reconstructed with a Turnbull–Cutait pull-through anastomosis and fasciocutaneous flaps.

Surgical technique

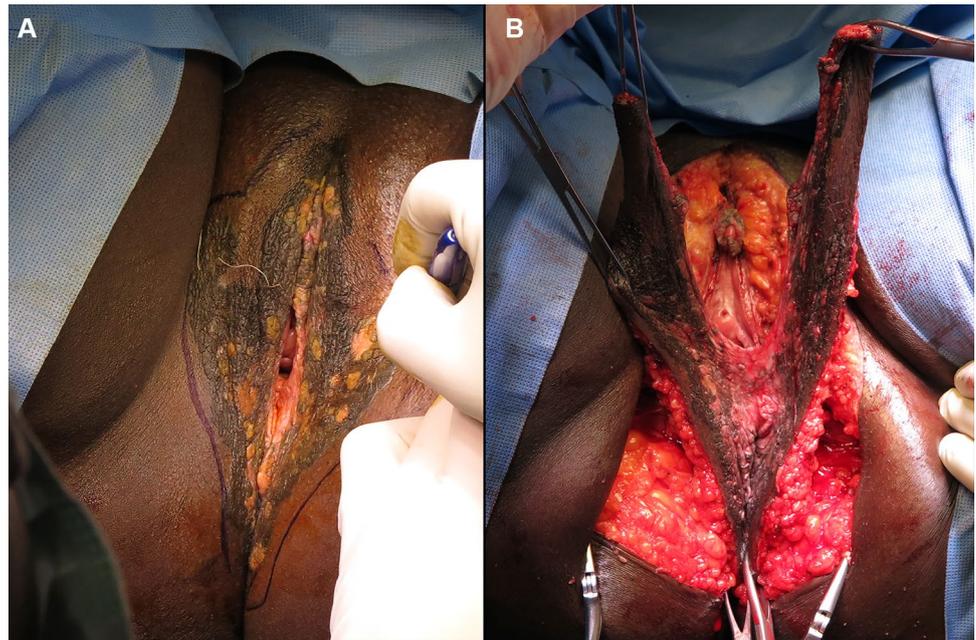
The patient received mechanical bowel preparation, perioperative antibiotics and thromboprophylaxis. The rectum was laparoscopically mobilised to the pelvic floor and vagina dissected from the rectum to facilitate mobilisation anteriorly. Dissection to the origin of the inferior mesenteric artery was completed for optimal mobilisation, however, no vessels were divided. A defunctioning loop ileostomy was fashioned at the right iliac fossa.

Simultaneously, a radical bilateral vulvectomy with clitoral sparing was completed (Fig. 1b). The clitoris was later treated with laser ablation. The cutaneous aspect of skin up to and proximal to the dentate line was then dissected free from the deeper tissues. The anorectal excision was made above the dentate margin, sparing the fibres of the internal anal sphincter, while achieving a margin which was macroscopically clear of disease. The rectum was transected with a laparoscopic stapler (Endo GIA™, Medtronic plc, Minneapolis, MN, USA) above the sphincter, and pulled downward

✉ A. C. Rogers
ailinrogers@gmail.com

¹ Department of General and Colorectal Surgery, Mater Misericordiae University Hospital, Eccles Street, Dublin 7, Ireland

Fig. 1 Extent of vulvar and anal intraepithelial neoplasia being marked to ensure adequate excision margins (left), with subsequent en bloc excision after rectal transection, to include perineal skin and anoderm (right)



through the perineum. Polydioxanone (PDS) sutures were applied to hitch the rectal muscle to the anal sphincter complex and secure it in place, leaving the mucosa unattached for the review procedure (Fig. 2).

Island fasciocutaneous flaps were raised over perforator-rich areas in the medial thighs. These were designed as Keystone flaps and transposed into the perineal defect bilaterally, attaching medially to the vaginal mucosa and securing in the midline anteriorly and posteriorly. A corrugated drain was placed behind the transected rectum through the perineal flap (Fig. 3).

One week later, the patient returned to theatre for completion of the reconstruction. The flap anastomosis was reopened in the midline, just over the point where the stapled rectum lay, to create a neo-anal orifice. The closed distal rectum was opened by resecting the staple line and delivered through the neo-anus. The rectal seromuscular sutures were reinforced to the anal sphincter with PDS, and the mucosa was anchored circumferentially to the skin flaps with 3–0 polyglactin 910. The corrugated drain was left in situ, and the island fasciocutaneous flaps were closed around the newly formed neo-anal opening.

A small wound dehiscence posterior to the neo-anal opening prompted a return to theatre a week later. The skin flaps were partially closed at the areas to approximate the soft tissues and two bolstering sutures were placed to secure the rectum to the skin. The patient was discharged on postoperative day 17 and the wound fully healed by 4 weeks (Fig. 4).

Histopathologic assessment demonstrated extensive VIN 3 and AIN 3 in the resected specimens, with clear margins and no invasive carcinoma. The patient underwent intensive pelvic floor physiotherapy during her recovery period

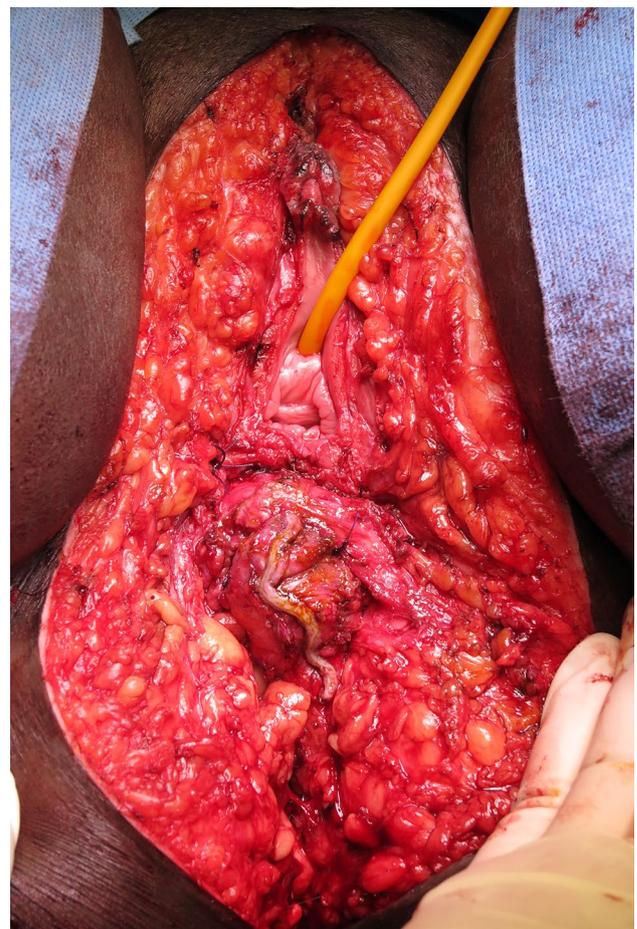


Fig. 2 Completed excision showing clitoral sparing and overstapled rectum



Fig. 3 Perineal flap reconstruction with corrugated drain exiting immediately posterior to rectal stump

and had a gastrografin enema test to confirm continence at 3 months postoperatively. She then underwent reversal of the loop ileostomy and recovered well, with good continence for solids, liquids and gas. She will remain under indefinite surveillance to ensure the disease does not recur.

Discussion

The current case demonstrates the challenges of managing extensive VIN and AIN, requiring multidisciplinary input, balancing the risk of progression to malignant transformation and consideration of patient wishes. HIV infection carries an increased risk of intraepithelial neoplasia, which affects up to a third of HIV positive patients [4]. The prolonged immunosuppression associated with HIV increases susceptibility to HPV and its associated cellular changes, including progression to invasive disease [5]. In fact, the increasing prevalence of anogenital dysplasia can be partly attributable to the improved survival expectancy of those with HIV in modern times, with the successful widespread use of antiretroviral therapy [2].



Fig. 4 Perineum at 4 weeks. Rectal mucosa can be seen with a scarred area posteriorly—this was the site of a small wound dehiscence which healed well

This patient was immunocompromised, had progressed despite multiple previous topical treatments and had multifocal extensive VIN and AIN, thus it was decided that a complete excision of disease with clear margins would give the best prognostic outcome. A local excision to the dentate line was felt to offer too high a risk of margin-positive disease, and thus a resection above the anorectal junction was deemed necessary. The patient wished to avoid a stoma, thus an abdominoperineal resection (APR) was deferred in favour of a reconstructive technique. With preservation of her anal sphincter and continued pelvic floor exercises, she underwent a successful reversal of her ileostomy and maintained continence.

This novel approach of radical soft tissue and rectal resection, with extensive perineal reconstruction and formation of a neo-anus demonstrates an alternative treatment option for those with extensive disease and high risk of malignant transformation.

Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki Declaration and its later amendments or comparable ethical standards.

Informed consent The patient gave full written and informed consent for her phase and photographs to be presented internationally and in publication form.

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