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CASE REPORT

Surgical management of a giant condyloma of Buschke-Löwenstein in a patient with Netherton syndrome using the pedicled anterolateral thigh flap — a case report☆

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Summary The surgical management of a giant condyloma of Buschke and Löwenstein poses particular reconstructive challenges, given the wound size, depth, and infection risk. We present a case where a pedicled anterolateral thigh flap is used to reconstruct a complex wound following resection of a giant condyloma of Buschke and Löwenstein arising in a patient with Netherton syndrome. Our operative technique re-iterates the utility of this flap in perineal reconstruction and demonstrates the possibility for an 18 cm wide arc of rotation. To our knowledge, this is the first report of a successful complex excision of a giant condyloma in Netherton syndrome, the subsequent reconstruction, and periodic maintenance with topical therapies.

Introduction

The giant condyloma of Buschke and Löwenstein (GCBL) is a rare, large, cauliflower-like anogenital lesion.[1] GCBL has been associated with human papilloma virus (HPV) and immunosuppression or immunodeficiency. While there is no consensus regarding treatment of GCBL, wide surgical

☆ This work has not been presented, wholly or in part, at any meeting.

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excision is preferred due to its size, capacity for local destruction, and high risk of recurrence (50%).[1,2]

Netherton syndrome (NS) is a rare autosomal recessive disorder resulting from mutations in the SPINK5 gene and subsequent dysfunction of the encoded serine protease, LEKT1.[3] Clinically, it is characterized by trichorrhexis invaginata, ichthyosis linearis circumflexa, erythroderma, atopic diathesis and variable immunodeficiency.[4] An undefined susceptibility of patients with NS to various serotypes of HPV can result in extensive wart infections including GCBL, necessitating aggressive management.[5,6]

We present a case where a pedicled anterolateral thigh (ALT) flap is used to reconstruct a complex perianal, perineal, and perivaginal wound following resection of a GCBL arising in a patient with NS.

Case report

A 26 year-old woman with NS presented with a seven-year history of a massive perineal condyloma. She had not been sexually active. The lesion was enlarging, extremely painful, and foul smelling. Sitting was impossible; the odor created a social stigma. It extended from the sacrum through the gluteal cleft, encircled the anal sphincter and invaded the posterior reaches of the vulva (Figure 1). A diminished cutaneous immune response made her prone to skin infections. Deep infection was prevented with rotating prophylactic ciprofloxacin and trimethoprim and sulfamethoxazole.

Sigmoidoscopy revealed no anal canal involvement. A liquid-based Pap smear ruled out infection with oncogenic HPV sub-types but demonstrated the presence of atypical degenerated squamous cells suggestive of vulvar intraepithelial neoplasia (VIN I). Non-surgical, topically-based therapies were unsuccessful at containing the lesion.

Operative technique

Colorectal surgeons performed a temporary laparoscopic ileostomy prior to excision. Under general anesthesia in the lateral decubitus, the condyloma was resected in a subcutaneous plane, leaving a defect measuring 45 × 25 cm. Uninvolved anal sphincter skin was spared to preserve function and avoid cicatricial contracture (Figure 2).

Cadaveric allograft was used as a temporary closure to test the likelihood of graft take. The allograft was covered with Acticoat Ag® and bolster dressed with burn gauze and a tie-over. The patient went back to theater at day 5 post-op for a dressing change; there was greater than 60% graft loss due to infection.

Treatment of the infection necessitated multiple debridements in the main operating room. Once controlled, dressing changes were performed under conscious sedation until healthy granulation of the wound bed was evident. Bilateral gluteal and bilateral upper medial thigh flaps were raised and inset to decrease surface area. Adhesive patch testing was performed. A VAC® dressing was secured to the surrounding skin and a rectal tube to temporarily close the remainder of the defect until these local flap suture lines were adequately healed. This defect measured 20 × 12 cm and was posterior to the rectum.

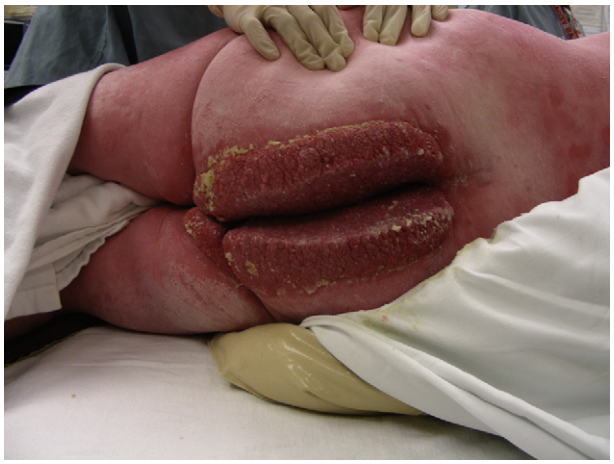

Figure 1 Preoperative view of the giant condyloma of Buschke-Löwenstein: the lesion extended from the sacrum through the gluteal cleft, encircled the anal sphincter and invaded the posterior reaches of the vulva.

A 21 × 9 cm sub-fascial ALT flap was elevated with an 18 cm pedicle. To maximize pedicle length and reach, the paddle was designed with the distal-most two identified perforators lying proximally in the flap; dissection was carried to the profunda junction.

The patient was transitioned to lithotomy. An 8 cm wide tunnel was made deep to rectus femoris and sartorius, then superficial to gracilis; two sartorius pedicles were taken.

The flap was transposed to the perineum. The most anterior aspects were aggressively thinned and secured to the anal sphincter remnant (Figure 3).

The donor site was closed primarily. The patient was taken back periodically for dressing changes in the subsequent weeks. Minor delays in primary healing were addressed with re-advancements. The patient was discharged home three weeks after reconstruction and re-referred for an ostomy reversal (Figure 4).

Over the next several months, the patient had complete healing of the area. Minor recurrences of condylomatous growths within the site responded to topical imiquimod and cryotherapy.

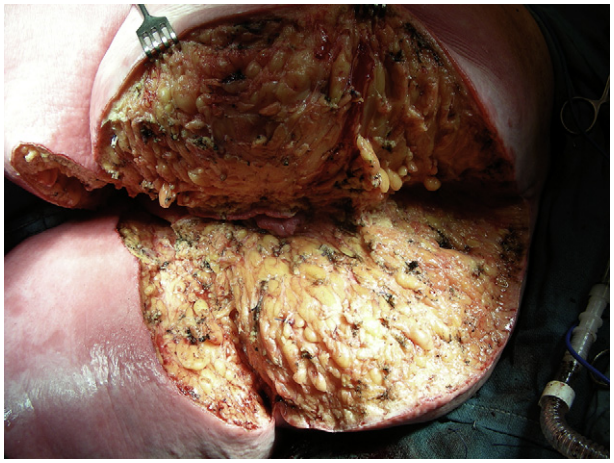

Figure 2 Perioperative view: the defect measured 45 × 25 cm.