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VY anoplasty for ectropion of anal mucosa in an adult with anorectal malformation

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ABSTRACT

Ectropion of anal mucosa (AME) is a possible complication following anorectal surgery and it is often cause of symptoms such as soiling, pain and bleeding affecting the patients' quality of life. Two-flaps anoplasty has been previously described for the correction of AME. We herein report a case of an adult patient who underwent a successful double V/Y-flaps for mucosal ectropion as consequence of repair of anorectal malformation (ARM).

A 25 year-old woman with history of ARM presented with an extensive AME causing bleeding, pain, soiling, and dyspareunia. This condition prevented her from an effective rectal nursing and a satisfactory social life.

The patient underwent the resection of ectopic mucosa and the creation of two V-shaped skin flaps to cover the skin gap. A minor dehiscence of one of the flaps was treated conservatively. At two years follow up the patient is symptoms free, has a good quality of life and can perform an effective bowel management.

AME is a possible complication after anorectal surgery for ARM and the multiple V-Y plasty is a viable procedure for its treatment. A multidisciplinary approach in these cases is recommended.

Level of Evidence V.

1. Introduction

Ectropion of anal mucosa (AME) is a possible complication in patients with previous history of anorectal surgery. It is characterized by anal mucosa lining perianal skin beyond the anal canal. Patients affected by AME may suffer from soiling, anal pain and bleeding. These symptoms lead to variable degrees of quality of life detriment.

Many techniques have been reported for surgical correction of perianal lesions in adult patients, with variable results. In particular "flaps" techniques have been described to prevent anal stricture and muscles lesions [1–3] as well as the V-Y anoplasty for anal stricture or mucosal ectropion has excellent results.

We report the successful use of multiple V-Y flaps technique in an adult formerly affected by anorectal malformation (ARM) associated with VACTERL and Triple X Syndrome who presented with symptomatic AME.

2. Case

A 25 year-old woman affected by VACTERL association and 47-XXX chromosomal arrangement was treated, during the neonatal period, for type III esophageal atresia and ARM with rectovestibular fistula, this last requiring a redo anorectoplasty during childhood. She presented with an extensive AME causing bleeding, pain and soiling. Furthermore, the condition prevented the patient from daily rectal enemas required to effectively manage her fecal incontinence. Her social life was consequently heavily affected.

A multidisciplinary evaluation, involving paediatric and plastic surgeons, a stoma therapist and a psychologist was conducted and a surgical approach offered. After bowel preparation, using polyethylene glycol solution and antibiotic prophylaxis, under general anaesthesia, the patient underwent examination and sphincter electro-muscle stimulation. Absence of stricture and the correct position of the anal opening were ascertained. Ectopic mucosa was excised to the muscles plane (Fig. 1A and B); two skin flaps (4 by 1,5 cm and 2 by 1 cm respectively) were created via V incisions and Y sutures using interrupted 4-0 resorbable sutures (Fig. 1C and D).

Fasting, total parenteral nutrition and intravenous Cefazoline, Gentamicin and Metronidazole were prescribed for 4 days.

At the 6 months follow up visit the patient reported the effectiveness

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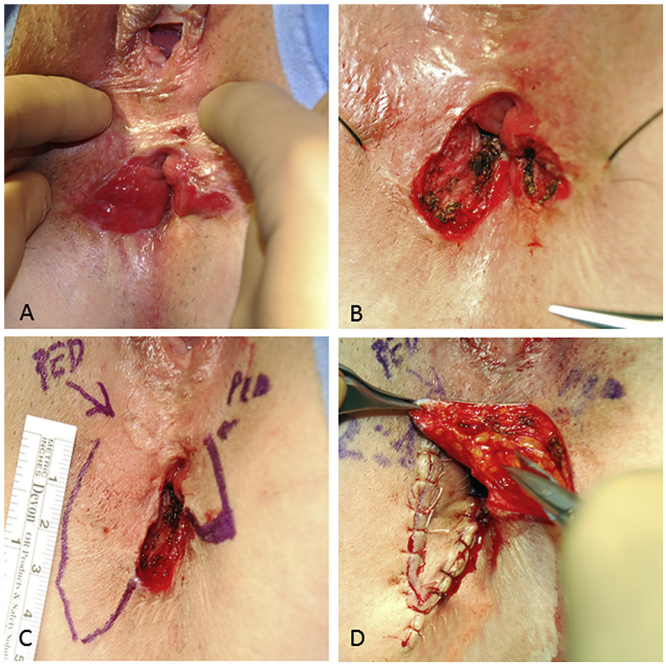

Fig. 1. A. Extensive portion of anal mucosa slips out of the anal canal lining the perineal surface; **B.** Resection of ectopic mucosa preserving muscles plane; **C.** Two 4 by 1,5 cm and 2 by 1 cm flaps skin drawing; **D.** V incision and Y suture; interrupted 4-0 resorbable sutures were used; preserving enough subcutaneous tissue provided a good blood supply.

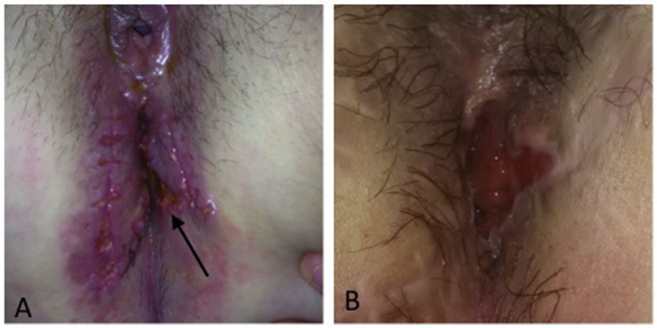

Fig. 2. A. Partial dehiscence of the minor flap (black arrow); **B.** Healed dehiscence with mucosa lining.

of resumed rectal washout with good bowels control. Eighteen months after surgery pain and soiling were resolved. Furthermore, she confirmed the complete acceptance of her perineum appearance (Fig. 2B).

3. Discussion

Patients affected by ARM receive their main surgical treatment during infancy. However, long term sequelae may require life-long invasive diagnostic and therapeutic procedures. Transitional care gained, in recent years, large speculative space in the specialized literature. Agreed pathways have still to be identified to address any possible issues that can affect these patients in adulthood.

Colorectal surgeons can be asked to treat an ARM patient presenting during the adult life with AME, its symptoms and consequent deterioration of patients' quality of life.

The occurrence of AME during the follow up of ARM patients varies grossly among series from 4 to 60% [2,4]. The variability on symptoms reporting, associated with different age and social life impact, could account for the wide range. Studies including adult patients, indeed, report higher incidence of AME compared with large studies focusing on children [5]. The case described had previous history of low ARM treated at another institution and needing a redo anorectoplasty during childhood. She presented to our colorectal clinic with anal discomfort and social impact of the ectropion, that was affecting her sexual life along with her self-confidence.

Several procedures have been reported to correct AME. Some authors described the S-plasty rotational advancement flaps [6,7], other reported good results using diamond-shaped flap with subcutaneous pedicle [8] while Oh et al. used the rotational advancement C-flap plasty [9].

Multiple flaps procedure with different arrangement of the flaps has been reported in a small series to treat both ectropion and rectal prolapse in patents with previous history of ARM [2].

Two-flaps anoplasty for ARMs can be applied for primary correction of some forms of ARM. It creates a sufficiently deep anus, preventing mucosal prolapse while preserving the anal sphincter [1]. More recently the risk of anal stricture has been outlined [5]. Large flaps size and attention to blood supply have been suggested to improve this aspect [10]. The same authors advise to restrict the defecation for 2 weeks prescribing fasting and parenteral nutrition for a week. Early postoperative feeding has been recently shown as preferable in infants with low ARM without covering stoma [11]. In the case herein described there was poor compliance to the prescribed fasting with occurrence of loose stools after surgery that may have contributed to a minor wound dehiscence. It is known patients with triple 47XXX may have mental disorder and do not follow recommendation after surgery [12,13].

Bowel management has been described as the treatment of choice in patients with previous history of ARM and unsatisfactory continence [14]. A recent multicentric study showed good results in terms of tolerance and quality of life in adults and children treated for ARM and using dedicated device for rectal irrigation [15]. The correction of AME, in this patient, was indicated also to allow her to resume this practice. Indeed, after surgery, she was able not only to successfully manage the transanal irrigations, but also to practice sports and to have regular intercourse.

4. Conclusions

AME is a possible complication after surgery for ARM. It can occur both in childhood and adulthood. Although it is not a life-threatening complication, it could affect the quality of life of patients. V-Y anoplasty is a suitable plastic surgical procedure. In this case it led to the complete resolution of preoperative symptoms. The multidisciplinary approach is mandatory to correctly indicate the best treatment for late sequelae of ARMs surgery.

Disclosures

- Consent to publish the case report was not obtained. This report does not contain any personal information that could lead to the identification of the patient
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- All authors attest that they meet the current ICMJE criteria for Authorship.
- All the authors have no financial disclosures: (ELP, FFL, FB. PM, PG).

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.epsc.2019.02.005.

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