



Recurrent rectourethral fistula repair: A novel technique of gracilis muscle interposition^{☆,☆☆}

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ABSTRACT

Background: A perineal approach with gracilis muscle interposition (GMI) remains the most common in recurrent rectourethral fistulas (RUFs). The closure failures in 10%–20% cases and urinary incontinence in 10%–80% cases referred to the disadvantages of the perineal approach owing to neurovascular damage. This article shows that the retraction of a survived muscle gracilis flap is one of the causes of fistula recurrence, and a simpler technique of the flap interposition is presented, requiring a less invasive perineal access.

Methods: Three consecutive patients with RUF at the age of 5, 13 and 15 years who underwent multiple (3, 4 and 5) unsuccessful reconstructive attempts were referred to our clinic. The last procedure was performed with GMI. In all cases, these were acquired iatrogenic fistulas that occurred after operations for Hirschsprung's disease (2) or anorectal malformation (1), and subsequent dilatation of the anus. All patients had a colostomy several years ago. Reinterposition and distant anchoring of the survived gracilis flap were performed through the perineal approach, avoiding lateral and anterolateral dissection.

Results: The early postoperative period was uneventful. In one patient, a stricture of the posterior urethra formed. Urethrotomy was performed in 3 months, with complete restoration of micturition. The colostomy was closed in all patients after 3–4 months. In the long term follow up within 4–8 years, no fistula recurrence was noted.

Conclusions: The suggested traction technique of GMI allowed: to minimize the perineal access size, hence minimizing the injury and the operating time; to place the anchoring suture outside of the inflammation zone, ensuring secure attachment; to provide precise flap positioning and intimate contact between the flap and the urethra.

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Recurrent rectourethral fistulas (RUFs) after failed acquired or recurrent fistula repairs are considered complex fistulas, representing a challenge to a surgeon regardless of the initial condition (anorectal malformation, Hirschsprung's disease, Crohn's disease, trauma or others) [1,2].

A perineal approach with gracilis muscle interposition (GMI) remains the most common approach to close complex fistulas, being successful in 70%–90% of cases [3]. In a meta-analysis involving 26 studies of adult population, Hechenbleikner et al. [4] collected data from 416 patients with acquired rectourethral fistulas. Most patients (65.9%)

underwent transperineal repair. Tissue interposition flaps, predominantly gracilis muscle, were used in 72% of repairs. Most high-volume centers (>25 patients) performed transperineal repairs with tissue flaps in 100% of cases.

A wide space for manipulation with good visualization, the ability for simultaneous urethral reconstruction and a flap interposition are thought to be the key benefits of the perineal approach. However, among its drawbacks, the risks of urinary incontinence and erectile dysfunction, which most experts believe to be owing to nerve branch damage during the course of massive dissection between the rectum and the urethra, are emphasized [5,6,7,8].

After GMI failure, it is difficult for the patient to agree to reoperation and for the surgeon to make a decision to redo the repair, especially since the choice of treatment options is usually very limited – permanent urinary and fecal diversion or reinterposition of the gracilis muscle, taken from the opposite hip [9].

Acquired rectourethral fistulas are not routinely encountered in pediatric surgical practice [2]. Only a few successful repairs of repeatedly operated recurrent RUF in children with anorectal malformation and Hirschsprung's diseases are described. The interposition of the gracilis flap in boys with RUF has not been reported in published studies.

Abbreviations: RUF, recurrent rectourethral fistulas; GMI, gracilis muscle interposition.

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This article shows that the retraction of a survived muscle gracilis flap is one of the causes of fistula recurrence, and a simpler technique of the flap interposition and distant anchoring is presented, requiring a less invasive perineal access.

1. Patients and methods

Three patients were referred to the clinic after repeated failed attempts to close the rectourethral fistula. All patients had a colostomy prior to the first fistula repair several years ago.

Clinical assessment included urethrocystorectoscopy with simultaneous digital rectal examination, voiding cystourethrography (VCUG), blood tests and ultrasound renal and bladder scan. Routine urine culture was taken and preoperative antibiotics were administered for 2–5 days. The operations were performed by a surgeon familiar with the techniques and approaches used in urology and coloproctology.

1.1. Procedure

1.1.1. Patient preparation

The patient was in the lithotomy position. During urethroscopy, a 5 F catheter was inserted to the rectum through the fistula, and a guidewire was introduced into the bladder, over which the 10–12 F Foley catheter was introduced.

1.1.2. Dissection

The rectum was separated from the posterior urethra by perineal access through the available scar. Dissection was carried out, creating a bed for the gracilis flap, only along the surface of the flap and the anterolateral aspects of the rectal wall, avoiding further lateral dissection. The fistula was transected focusing on the catheter inserted into the urethra. Dissection stopped 2 cm above the upper edge of the fistula. Thus, the muscle flap was completely isolated and pulled out without the massive dissection between the rectum and the urethra.

1.1.3. Closure

The rectal opening was closed with a longitudinal running suture (3/0), and the urethral opening was closed with interrupted sutures (polydioxanone 4/0) in the oblique direction to reduce tension.

1.1.4. Gracilis muscle interposition

The anterior abdominal wall was pierced above the pubic bone on the opposite side from the base of the flap, using curved forceps inserted through a small skin incision in the projection of the external inguinal ring. Next, the forceps were advanced to the posterior surface of the vertical branch of the pubic bone, perforating the endopelvic fascia, orienting on the tip of the finger inserted paraurethraly at the apex of the wound. Thus, the forceps jaws passed into the perineal wound (Fig. 1). The end of the muscle flap was firmly stitched with a figure-eight suture (thread, Vicryl 1.0). Then the free end of the thread was caught and pulled out by the forceps, drawing the gracilis flap into the bed until it stopped (Fig. 2A), after which the thread was anchored to the aponeurosis on the anterior abdominal wall (Fig. 2B), in a position of slight tension, to form a traction suture (Fig. 3). As a result, the muscle flap occupied an oblique position snuggling tightly side by side with the urethral closure site without additional dissection, necessary to secure the muscle flap with sutures.

1.2. Postoperative period

All patients received broad-spectrum antibiotics for 10–4 days. Drains were removed on day 8–10. The urethral catheter was removed and a cystography was performed at 21–28 days. With free urination and the absence of leakage, the cystostomy catheter was removed.

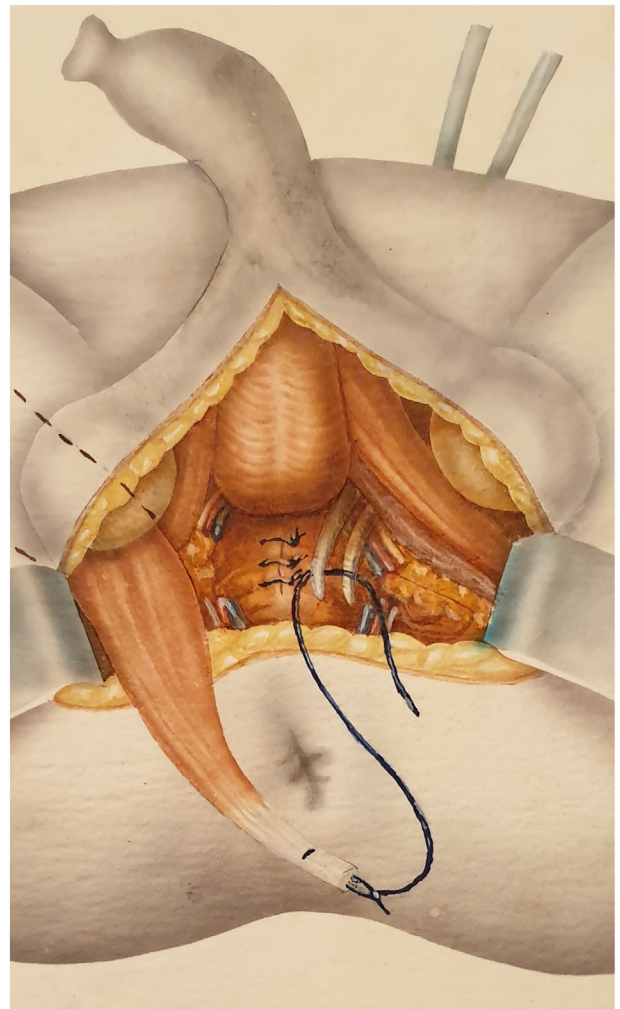


Fig. 1. The forceps passed into the perineal wound and the free end of the thread was caught.

Colostomy reversal was performed after 3–4 months. The patients underwent a thorough medical examination after 6 and 12 months with long-term results evaluated after 4–8 years.

2. Results

2.1. History

Three children with RUF at the age of 5, 13 and 15 years, who underwent multiple [3–5, and] failed fistulas repairs were referred to our clinic. In all cases, these were iatrogenic fistulas that occurred after operations for Hirschsprung's disease [2] or anorectal malformation [1], and subsequent dilatation of the anus. Two patients with Hirschsprung's disease had undergone redo abdominoperineal pull-through procedures, with subsequent failed attempts to close the fistula through the perineal approach and gracilis muscle interposition. In the youngest patient, GMI was performed with a walking flap [10]. The patient with anorectal malformation, presenting with obesity, had undergone five attempts to close the fistula via the perineal approach, including redo rectal perineal pull-through procedure. His anus was well-formed and correctly positioned.

Colostomy was in place in all three of the cases prior to the first attempt at rectourethral fistula repair. The fistulas had existed for 4, 11 and 13 years. The two teenagers had weak erections and one suffered from recurrent orchiepididymitis which led to bilateral vas ligation. The diagnosis of RUF was undisputed, since all of the patients presented

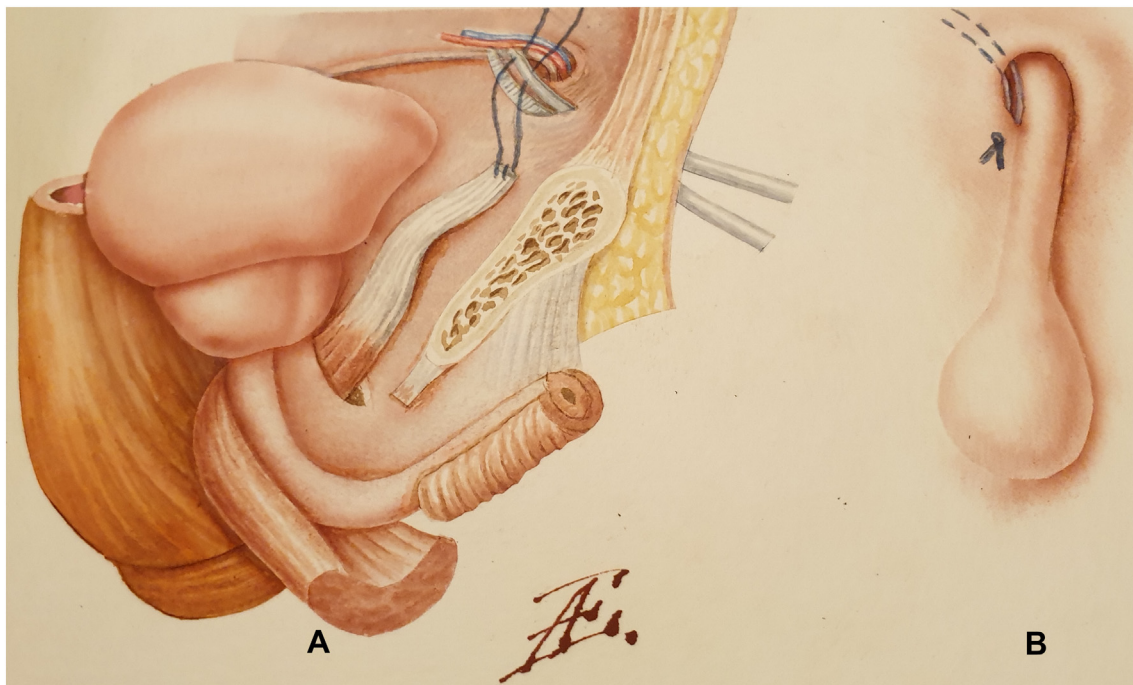


Fig. 2. Drawing the gracilis flap into the bed. (A) Pulling the traction suture with the gracilis flap. (B) The traction suture anchored to the aponeurosis.

with anal urine leakage during urination. Recurrent urinary tract infections were noted in all of the patients.

2.2. Examination results

Anus was well-formed in the patient with anorectal malformation. The location and the shape of the fistula were clearly visible with urethroscopy. The fistula in each patient was located in the prostate-membranous part of the urethra and had the form of a fissure 1–1.5 cm long. In the rectum, the fistula was located on the anterior wall at a depth of 3–5 cm. Moreover, in two boys with Hirschsprung's

disease the fistula was located directly above of the dentate line. Digital rectal examination showed the gracilis muscle palpable in the form of a longitudinal bulge under the anterior wall of the rectum, located distally to the fistula (2 patients). In the third patient, the muscle was displaced from the midline laterally and distally towards the base of the muscle. Urinary bladder mucosa showed signs of chronic inflammation in all of the patients.

2.3. Surgeries

The flap was separated from its bed without notable bleeding. Blood loss was 10, 20 and 40 mL. The operations took 165, 110 and 140 min, respectively. The space between the gracilis muscles and the surrounding tissues was soft enough, without significant fibrosis. The flap in all patients was alive, with no signs of circulatory impairment or significant scarring, although dense scars were palpated around it. The muscle flap interposition using the traction suture was safe and took no more than 10–12 min.

2.4. Postoperative period

The early postoperative period in all cases was uneventful. After removing the urethral catheter on day 21–28 and clamping the cystostomy catheter, urination was restored in two patients. Voiding cystography did not demonstrate contrast leakage. In one patient, a stricture of the posterior urethra formed, 0.5 cm long, and readmission occurred after 3 months. A urethrotomy was then performed, with complete restoration of the passage of urine through the urethra.

The colostomy was closed in all patients after 3–4 months.

In the long term follow up, no fistula recurrence was noted. Erectile function and urinary incontinence manifestation did not change notably.

3. Discussion

In the management of boys with anorectal malformations, acquired, persistent or recurrent rectourethral fistulas may form [2]. This complication appears to most commonly result from an inadequate operative

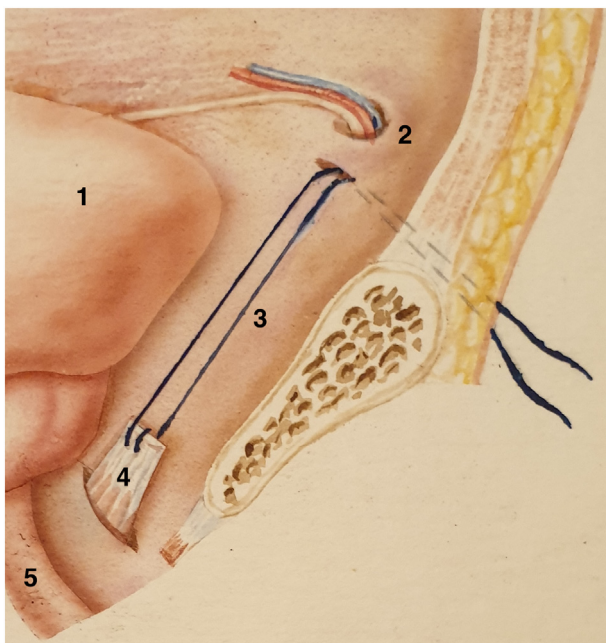


Fig. 3. Final position of the flap with the traction suture. (1) Urinary bladder, (2) internal inguinal ring, (3) traction suture, (4) gracilis flap, and (5) urethra.

technique [11]. Also, rectourethral fistulas have been reported to occur after commonly used pull-through procedures for Hirschsprung's disease — Soave and Swenson as a further complication of urethral injury, anastomotic disruption or after subsequent dilatations owing to tear of anastomotic stricture [12,13].

Recurrent RUFs following failed repair attempts are considered to be complex fistulas; the recurrence rate after the closure is about 50% in pediatric patients [14,15]. There is no gold standard operation, and a choice of repair technique depends on surgical specialty, expertise, the fistulas' features and associated problems such as anal or urethral stricture [16,17]. Operative management of recurrent RUF is technically challenging and can be categorized into 2 types of procedures, both of them basically aimed at separating the rectal suture line from the urethral sutures: a) various vascularized flaps interpositions and b) rectal wall reposition techniques, mainly redo pull-through procedures, utilizing laparotomy and posterior approach.

Endorectal pull-through procedure using the anterior perineal transsphincteric approach in RUF repair was proposed by Young and Stone [18] in 1917. In 1969, Kilpatrick and York-Mason described the posterior transsphincteric transrectal approach [19], which was advocated by de Vries and Pena in their technique for the treatment of imperforate anus [20]. Kubota et al. [21] performed an endorectal pull-through in pediatric recurrent RUF using abdominal and posterior sagittal access. Later, this access was used for the redo pull-through in a boy with Hirschsprung's disease and recurrent RUF [10]. Posterior sagittal anorectoplasty with ischioanal fat pad interposition was described by Levitt M.A. et al. [2].

Redo rectal pull through in fistula repair is a reasonable way in patients with rectal stenosis or incorrect position of the rectum outside of the sphincter complex. In boys with Hirschsprung's disease, the fistula is often located near the dentate line, so overlapping of the fistula by the rectal wall is possible only when the rectum is sewn below the dentate line, which can lead to fecal incontinence [16]. When the anus is normal or well-formed in the course of the previous anorectoplasty, the perineal route can be applied to the fistula repair.

The perineal approach is the most commonly used in complex RUFs in adults from published studies. A good fistula exposition, convenience of the flaps interposition, avoiding circular mobilization of multioperated rectum, and facilitating urethral reconstruction are considered as advantages of the perineal approach, while the risks of erectile dysfunction and urinary incontinence, owing to damage to the nerve branches, refer to its drawbacks [22,23,24]. A wide dissection between the rectum and the urethra is necessary, not only for exposure of the fistula and closure of the urethral and rectal openings, but also for anchoring a muscle flap 3 cm above the fistula level by several interrupted sutures [25]. Urinary incontinence following the perineal approach is observed in 10%–80% of cases [6,26]. The manifestation of urinary incontinence owing to the perineal approach is so significant that the artificial urethral sphincter implantation rate reaches 75% in the United States and Europe [22,27]. Causes of urinary incontinence are considered to be damage to the perineal branches of the pudendal nerve (neural impairment/injury) [7] and nerve branches in the apex of the prostate during dissection and anchoring [8,28].

A variety of interposition flaps through the perineal access have been shown, including muscle flaps — gracilis, gluteus maximus, levator ani, dartos, bulbocavernosus — as well as others flaps — island groin, tunica vaginalis, scrotal myocutaneous and ischioanal fat. Gracilis muscle is the most widely used one because of its regular blood supply and easy mobilization with minimal donor site morbidity. The perineal approach with GMI is usually the last choice for the recurrent rectourethral fistula, indicating the inefficiency of previous attempts using other methods.

Rectourethral fistula recurrence after GMI seems to be a dramatic event, leaving almost no chance for successful reconstruction. Repeated muscle interposition from the contralateral hip is rarely employed, as both surgeons and patients no longer believe in the successful outcome [25,26]. Failures after GMI are usually attributed to inflammation, tissue

scarring or circulatory impairment in the flap. However, current observations have attributed the recurrence of fistulas to partial flap retraction owing to the muscle contraction. In all three cases, the muscle flap was not damaged, was viable, could be easily separated from the surrounding tissues, and was displaced towards its base, and the fistula was located cranially or craniolaterally from it. Retraction of the survived flap owing to its contraction has not been previously suggested as a cause of rectourethral fistula recurrence, although the possibility of gracilis retraction has been mentioned following the urethral reconstruction in a female [29].

We have described here the technique of reinterposition of the surviving gracilis muscle as a brief solution to a complex problem, when it is hard to decide on a new operation and urinary and fecal diversion might seem like a natural choice.

A number of factors contributed to the *M. gracilis* reusing in this group. Repeated failed attempts at fistula closure, including redo pull-through procedures; a well-formed or normal anus; fistula location next to the dentate line in boys with Hirschsprung's disease and the presence of a well-vascularized muscle flap in the wound led to an understanding that the flap reintegration would be the least damaging and technically the easiest option.

The surgeries have been performed via a less traumatic perineal approach with dissection along anterolateral aspects of the rectal wall and the surface of the gracilis flap, excluding dissection in the anterolateral direction when separating the rectum from the urethra and extensive dissection behind the prostate gland. Using the traction suture avoids having a wide operating field and the need to secure the flap to the prostatic capsule with stitches, which diminishes the risk of compression of neural branches. Traction suture turned out to be a technically simple and reliable anchoring mechanism, compared to several stitches attaching the muscle flap to the prostatic gland. Additionally, the point of fixation of the traction suture is out of the inflammation zone, which ensures a more secure anchoring and reduces the risk of suture failure and flap retraction.

Zmora et al. [25] have emphasized that, during the fistula closure, more attention should be paid to the high pressure area occurring in the urethra. In addition, it should be noted that cystostomy is usually reversed prior to the colostomy being closed and urethral sutures become subject to increased pressure during micturition, while the closed area in the rectum is still resting. Hence, tight contact between the muscle flap and the urethra, thanks to the traction suture described in this method, apparently contributes to faster healing of the urethral closure and secure fistula closure.

In one of the observed cases, botulinum toxin has been employed in order to achieve myoplegia and immobilize the muscle flap, when it appeared too short. Since botulinum toxin injections into the muscle prevent it from contracting and retracting, it seems reasonable to use it in GMI where the muscle belly is short, there is increased muscle tone or immobilizing the hip postoperatively is impossible. We have not come across data on type A botulinum toxin use in GMI; however, there are experimental and clinical data on improved outcomes of skeletal muscle and tendon repair after therapeutic botulinum toxin myoplegia [30].

This report has some limitations, which include the retrospective nature of the study and a small sample size owing to the rarity of the pathology. At the same time, the described method made it possible to obtain good results in a difficult situation with minimal costs and risks, which allow it to be recommended for further use and study, in recurrent and other complex rectourethral fistulas.

4. Conclusion

Here is presented a simpler and less invasive technique for rectourethral fistula closure with GMI. It has been shown that muscle flap retraction following the GMI procedure is probably an underestimated cause for RUF recurrence. The suggested GMI technique allows: to minimize the perineal access size, hence minimizing

the risk of neural branches injury; to place the anchoring suture outside of the inflammation zone; and to provide secure attachment, precise flap positioning between urethral and rectal sutures, and intimate contact between the flap and the urethra. Indications for this procedure include primary and recurrent RUF located in the prostatic–membranous part of the urethra.

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