

# Considerations before repair of acquired rectourethral and urethrovaginal fistulas in children

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**Background:** Acquired rectourethral or urethrovaginal fistula between the rectum or vaginal and lower urinary tract is an uncommon entity, which occurs as a consequence of pelvic disorder, including trauma, iatrogenic injury, inflammatory bowel disease, pelvic neoplasm or infection. But when is it appropriate to repair the fistula and what methods to be chosen? There has been no consensus on them. This study was undertaken to determine the timing of the procedure and the repair of rectourethral and urethrovaginal fistula.

**Methods:** From 1998 to 2006, we treated 19 children with rectourethral or urethrovaginal fistula, including rectourethral fistula in 15 boys and urethrovaginal fistula in 4 girls. The mean age of the patients was 6.2 years (range, 8 months to 11.5 years). The fistula occurred after pelvic fracture in 10 patients, and after iatrogenic injury in 9 including 4 after radical operation for Hirschsprung's disease and 5 due to anorectal malformation. Preoperatively, the general and local infections were controlled thoroughly, and complications such as urethral stricture and secondary megacolon were treated at first. At least 6 months after the last procedure, all patients underwent the 1-stage York-Mason procedure (via parasacrococcygeal incision) without colostomy and suprapubic cystostomy. Intraoperatively, the entire fistulous tract was excised completely.

**Results:** Infection and partial dehiscence of the wound occurred in 2 patients respectively. All fistulae were closed successfully without fecal incontinence or postoperative anal stricture. No patient suffered from urinary incontinence after fistula repair. The scars around the fistula were removed because they would shrink and lead to subsequent urethral occlusion or stricture.

**Conclusions:** The timing of operation for acquired rectourethral or urethrovaginal fistula is appropriate at least 6 months after the last procedure. The 1-stage York-Mason procedure for the repair of the fistula is feasible and effective.

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**Key words:** fistula;  
rectourethral fistula;  
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## Introduction

Acquired rectourethral or urethrovaginal fistula between the rectum or vaginal and lower urinary tract is an uncommon entity, which occurs as a consequence of pelvic disorders, including trauma, iatrogenic injury, inflammatory bowel disease, pelvic neoplasm, or infection. The fistula presents a particular challenge to surgeons. If inappropriate measures are taken, the patients may suffer from failures. There is no clear consensus on the timing of operation and operative methods. When is it appropriate to repair the fistula with 1-stage or 3-stage procedures? Is it necessary to do colostomy and suprapubic cystostomy? Which surgical technique is chosen to repair rectourethral fistula? All of these issues must be considered before repair of the fistula.

## Methods

We reviewed the data of 19 patients who had received repair of the acquired rectourethral or urethrovaginal fistula at our hospital from 1998 to 2006. Rectourethral fistulae occurred in 15 boys, and urethrovaginal fistulae in 4 girls. The mean age of the patients was 6.2 years (range, 8 months to 11.5 years). Of the 19 patients, 10 had pelvic fracture and 9 had iatrogenic injury including radical operation for Hirschsprung's disease (4), and anorectal malformation (5). The patients with pelvic fracture underwent one or several urethral operations.

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Of the 4 girls with urethrovaginal fistula, 3 were complicated by vaginal dilation and dropsy because of pelvic fracture. Of the 19 patients, 7 underwent one fistula repair via the transanal approach and 2 via the transperineal approach, but 2 patients underwent two fistula repairs via the transanal approach. Diagnosis was based on pneumaturia or feces in the urine. Rectal examination was performed to search for the fistula. Endoscopy, voiding cystourethrography and thin barium enema examination were used to show the localization of fistula and the orifice, and to observe whether urethral stricture or megacolon existed. Anorectal manometric studies were performed to estimate the sphincteric function.

### Preparation before operation

The timing of operation was at least 6 months after the last procedure. If the urinary tract was infected severely, intravenous antibiotic was given and a Foley catheter should be inserted until the infection was controlled. Preoperatively, all patients took fluid food, while metronidazole and neomycin were given orally. The patients were subjected to daily enema for 7 to 10 days and intravenous antibiotic prophylaxis. A clean colon enema was given before the operation.

### Management during and after operation

All patients underwent the 1-stage York-Mason procedure without colostomy and suprapubic cystostomy.

The patient was placed prone in the jackknife position with the buttocks separated with adhesive tape. An incision was made in the midline from the tip of the coccyx to the anal verge, and subcutaneous tissue was divided in the midline. The muscle bundles of the anus were divided layer by layer, and each sphincter muscle was tagged with color-coded suture for the following reconstruction of the anus. The rectal mucosa was then opened along the entire incision length. The orifice of the fistula was directly visualized and probed to determine the direction, size and orientation of the fistula. The anterior rectal orifice and the entire fistulous tract were completely excised using a scalpel. All inflammatory tissues in the region of the fistula were also excised, allowing for reapproximation of healthy tissues. The fistula was closed with 5-zero interrupted absorbable sutures. The anterior rectal wall musculature was closed with 4-zero interrupted silk sutures. The anterior and posterior rectal mucosa was closed in a single layer with 3-zero chromic sutures. The paired sutures of the anal sphincter were then tied and the subcutaneous tissues were closed with absorbable suture. A drain was left under the presacral fascia. The skin was closed with absorbable sutures.

Postoperatively, the patient was placed in a prone position with the wound exposed and a headlamp lighting the wound continuously. The wound was given intensive care. The patient was maintained with no oral intake for 3 days. Drains were removed on postoperative day 3 to 5. The urinary bladder catheter was pulled out after 14 days. After two weeks of the operation, the anus was dilated for 3-6 months.

### Results

The time for fistula repair was 1.5 to 2 hours in all patients. The loss of blood ranged from 50 to 150 ml. No transfusion was required. Infection and partial dehiscence of the wound occurred in 2 patients respectively. All fistulae were closed successfully without fecal incontinence or postoperative anal stricture. No patient suffered from urinary incontinence after fistula repair.

### Discussion

Rectourethral or urethrovaginal fistula in children may have a congenital or acquired origin. Congenital rectourethral fistulae are usually associated with other anorectal abnormalities, while the etiologies of the acquired cases include pelvic fracture, redo pull-through in Hirschsprung's disease, redo operation in anorectal malformation, and operation for tumor. Owing to pelvic fibrosis and adhesions resulted from previous disorders and their complications, the acquired rectourethral fistula presents a challenge for the surgeon.<sup>[1]</sup> The following should be considered before the repair of the rectourethral or urethrovaginal fistula.

First, when is it appropriate to repair the fistula? Actually, there is no clear consensus on the most appropriate timing of fistula repair. Fengler et al<sup>[2]</sup> thought that immediate repair is appropriate if no sepsis is present and the fistula has manifested itself more than 6 to 8 weeks after surgery. Hanus<sup>[3]</sup> believed that the timing of repair should be individualized, mainly according to the etiology, delay of diagnosis, size of the fistula, the first or subsequent repair, and the general condition of patients. But we think that the timing of fistula repair depends on not only the general condition of patients, local condition of the fistula, but also the urethral stricture or secondary megacolon. Acquired urethrorectal fistulae are the complications of previous diseases, and there are often severe pelvic fibrosis and adhesions. In the early period, scars around the fistula are stiff and fragile. At the same time, tissues around the fistula often have chronic infection. Therefore, it

is advisable to repair the fistula after the scars become soft and infection disappears. We found that it was appropriate to repair the fistula after 6 months since the last operation or urinary infection being controlled. Urethral stricture or secondary megacolon, if present, should be cured at first before closure of the fistula. Pelvic fracture is often associated with severe posterior urethral injury. Before fistula repair, reconstructing an unobstructed urethra is very important. In pediatric patients, one of the etiologies of acquired urethrorectal fistula is redo operation in anorectal malformation. After anorectoplasty in neonates, some patients are possibly complicated by anal stricture or secondary megacolon. If urethral stricture or constipation is still present, the subsequent procedure may be at the risk of failure. In our series, 5 patients who were complicated by urethral or anal stricture underwent fistula repair at first, and they all failed.

Second, is it necessary to do colostomy and suprapubic cystostomy? Generally, acquired rectourethral or urethrovaginal fistula is repaired often by standard three-stage procedure (colostomy and cystostomy, fistula repair, and colostomy takedown). Fecal diversion and urinary diversion are thought to be done with the hope of spontaneous closure of fistula and protection of subsequent incision. Some fistulae, especially some small fistulae may close spontaneously after double diversion. Noldus et al<sup>[4]</sup> revealed that conservative approach is the first choice because about 50% of fistulae can be cured. However, according to Al-Ali et al,<sup>[5]</sup> in 30 patients who underwent double diversion, spontaneous closure of fistula occurred in 14, in whom 5 had a patent urethra and 9 had a occlusive or strictured urethra. In fact, it is more difficult to treat urethral occlusion or stricture than to treat urethrorectal fistula. Hence, it is unreasonable to perform double diversion only for the spontaneous closure of fistula. It is better to remove the scars around the fistula, which may contract and lead to subsequent urethral occlusion or stricture. Therefore, it is more advisable to repair the fistula actively than to wait for the spontaneous closure of fistula. A fecal diversion is helpful to prevent infection of subsequent incision.

Our former series<sup>[6]</sup> showed that an enterostoma can help to reduce the frequency of wound infection and wound dehiscence of the future operative site. Nevertheless, the enterostoma also brings about many serious complications, including stoma prolapse, intestinal obstruction, and skin dehiscence. Of our previous 48 patients with enterostoma, 39.6% had one or several complications.<sup>[6]</sup> The same results were also found by Patwardhan et al<sup>[7]</sup> and Nour et al.<sup>[8]</sup> Of those patients who have received colostomy, the treatment proceeds in three stages (diversion-bowel,

closure technique, and undiversion). Obviously, one-stage repair procedure without colostomy reduces the risks of multianesthesias and multioperations. Furthermore, it eases the physiological, psychological, and economic burdens of the patients and their parents. Economically, the one-stage procedure is especially applicable in the developing countries. As surgeons become more experienced, bowel preparations become standardized, and effective antibiotics are developed, it is feasible to repair the rectourethral or urethrovaginal fistula. Renschler and Middleton<sup>[9]</sup> thought that it is of great benefit to the patients when the one-stage procedure is successful. Wood and Middleton<sup>[10]</sup> repaired rectourinary fistulae using the single-stage transrectal transsphincteric (modified York-Mason) procedure, and thought that the procedure is more simple and effective than the standard three-stage procedure. In our present series, all patients who underwent the one-stage fistula repair achieved the same results as Renschler and Middleton's.<sup>[9]</sup> Of course, when antibiotics alone cannot control the inflammation and infection associated with the fistula, colostomy may be a good choice. Fortunately, all of our patients succeeded in the one-stage procedure without colostomy.

Third, which surgical technique is better for the repair of rectourethral or urethrovaginal fistula? Multiple methods have been reported, via the perineal, posterior sagittal transanal, transrectal or transabdominal approach. One of the major problems in the repair of urethrorectal fistula is good exposure. The perineal approach has the disadvantage of access through scarred tissue, affords limited exposure, and may endanger urinary continence. We found that in patients with pelvic fracture, redo pull-through in Hirschsprung's disease or redo operation in anorectal malformation, the distance between the orifice of the fistula and the dentate line was often much longer. Hence, it is difficult to get a good exposure via the transanal approach. The transabdominal approach may induce complications of any intra-abdominal procedures, and patients need a long time for recovery. While the posterior midline transsphincteric approach, first described by Bevan and later popularized by York-Mason,<sup>[11]</sup> may usually avoid the dissection through a previous surgical site and afford a rapid, bloodless exposure through unscarred areas. Moreover, it may prevent the damage to neurovascular bundles and pelvic structures, which are important for sexual function and urinary continence. Fengler,<sup>[2]</sup> Bukowski,<sup>[1]</sup> Fournier<sup>[12]</sup> and Dal Moro et al<sup>[13]</sup> all succeeded in the treatment of urethrorectal fistulae by the York-mason approach. Barisic and Krivokapic<sup>[14]</sup> believed that the York-Mason trans-sphincteric

approach offers a straightforward access through healthy tissues, and a good visualization of fistula, and that the suturing of anterior rectal wall flap can easily be performed for definitive closure of the rectourethral fistula.

For fear of incontinence or wound infection, however, some surgeons do not accept this approach. Clinical experience<sup>[15]</sup> and experimental studies<sup>[16]</sup> have proved that the posterior approach with a precisely midsagittal incision does not interfere with the mechanism of continence. In Renschler and Middleton's<sup>[9]</sup> and our series, no fecal incontinence or anal stenosis developed, and fistulae were repaired successfully by the York-Mason approach.

We think that the York-Mason approach is satisfactory to treat acquired urethrorectal fistula. During the operation, the layered muscle bundles of the posterior anus are divided layer by layer, and each sphincter muscle is tagged with color-coded suture for accurate reconstruction of the anus at the end of the procedure. The orifice of the fistula is directly visualized and probed to determine the direction, size and orientation of the fistula. The anterior rectal orifice and entire fistulous tract are completely excised using a scalpel. All inflammatory tissues in the region of the fistula are also resected for reapproximation of healthy tissues. However, the excision of the fistulae is not recommended by some pediatric surgeons,<sup>[17]</sup> who think that if the tract is excised, a small fistula becomes a large fistula, which can lead to tension on the closure and subsequent failure. And other disadvantages include bleeding of the freshly excised wound margins followed by coagulation, which can compromise healing and potentially damage the urethral orifices. Moreover, Raz<sup>[18]</sup> considered that the well formed fistula scar could protect the postoperative spasms of the bladder. But chronic inflammation and scars around the fistula are difficult to heal. Hence, it is important to debride scars and devitalized tissues at the site of fistula. As for a large fistula, it is filled with a rectal muscle flap and then the anterior rectal wall is sutured layer by layer. Urethrovaginal fistula is often associated with vaginal stricture or atresia, and the proximity of the vagina is often dilated and dropsied. During the procedure, the fistula and the vaginal stricture or atresia can be exposed satisfactorily via the side-rectum approach. However, we should not ignore the one-stage York-Mason approach responsible for a higher rate of wound infection. Effective measures must be taken to diminish the frequency of infection and dehiscence of the wound. The measures include protection of tissue, prevention of fat liquefaction, thorough hemostasis, elimination of dead cavity, and fasting.

Obviously, it is very important to choose appropriate timing of operation and appropriate procedures for the repair of rectourethral or urethrovaginal fistula. Our experiences have shown that complications such as urethral stricture and secondary megacolon must be treated before the repair of fistula. The fistula can be repaired satisfactorily after the scars around it become soft, and general and local infections are controlled. It is feasible to repair the rectourethral or urethrovaginal fistula by the one-stage procedure. The York-Mason approach allows the surgeon to repair the fistula with excellent visualization via nonoperated tissues. It is a highly effective and minimally invasive procedure for the repair of urethrorectal and urethrovaginal fistula.

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*Lotus and bird* by Zhu Da (1626-1705), Qing Dynasty