Int Surg 2017;**102:**560–567
DOI: 10.9738/INTSURG-D-17-00092R1

Treatment Efficacy of Different Surgical Procedures for Postoperative Residual Rectourethral Fistula Following Anoplasty

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Background/Purpose: This study summarizes the surgical treatment options and their clinical efficacy for postoperative residual rectourethral fistula following anoplasty in patients treated in our department from July 2005 to July 2012.

Methods: For 38 cases of postoperative residual rectourethral fistula following anoplasty, according to differences in anal appearance and functionality, either an anterior sagittal or a posterior sagittal surgical rectourethral fistula repair procedure was performed.

Results: The residual rectourethral fistula following anoplasty was an obvious tube-like structure with a length of 0.9 ± 0.4 cm. Healing after the 1-stage operation was achieved in 37 cases. Among these cases, 35 were followed up and showed no urethral stricture and no diverticulum. The clinical score for anal function was excellent in 31 cases and good in 4 cases, with significant differences compared with the scores before surgery ($P < 0.05$). The respective operative times for the 2 surgical procedures were 74.6 ± 10.1 minutes and 105.6 ± 14.6 minutes ($P < 0.05$).

Conclusions: The appropriate choice of surgical procedure was dependent on the patients' anal appearance and functionality. Posterior sagittal anorectoplasty was suitable in cases with a severely disordered perineal appearance, and it was a relatively difficult operation. In contrast, anterior sagittal anorectoplasty was best applied in patients with minor alteration in perineal appearance, and it had a clear surgical field and was easy to perform. In this study, the repair of cases of residual rectourethral fistula following anoplasty using anterior sagittal or posterior sagittal anorectoplasty showed high success rates, and anal function was significantly improved.

Key words: Anorectal malformation – Anal atresia – Rectourethral fistula

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Postoperative residual rectourethral fistula following anoplasty is a severe complication resulting from errors during surgery for rectourethral fistula in cases of congenital anorectal atresia. Anorectal atresia with rectourethral fistula is the most common anorectal malformation in male infants, and anoplasty with a posterior sagittal approach (Peña surgery)[1] or laparoscopically assisted anorectal pull-through anoplasty[2] is often recommended. However, if the incorrect surgical procedure for the treatment of anorectal atresia is chosen, then a postoperative residual rectourethral fistula following anoplasty may occur; this condition manifests as a confluence of urine and feces and is often combined with poor anal appearance and function of the anus, potentially including perineal scars, rectal retraction, anal stenosis, uneven distribution of the external sphincter, bowel mucosa exstrophy, feces defile, or fecal incontinence. In these cases, surgery is the only treatment option. The commonly used procedures include rectourethral fistula repair by posterior sagittal anorectoplasty or anterior sagittal anorectoplasty. From July 2005 to July 2012, 38 pediatric patients with residual rectourethral fistula following anoplasty were treated in our department. According to differences in anal appearance and functionality, different surgical procedures were applied, and the surgical efficacy was satisfactory. The details are described as follows.

Methods

General information

From July 2005 to July 2012, 38 male pediatric patients with residual rectourethral fistula following anoplasty were treated in the Department of General Surgery of Beijing Children's Hospital. The patients' age range was 6 months to 14 years old, and the mean age was 3.6 ± 2.9 years. Among the patients, 30 showed clinical manifestations of confluence of urine and feces in the urethra and the anus, and 8 patients showed only anal urination, with no defecation via the urethra, including 5 patients who showed no urethral urination. The anal appearance was basically normal in 19 cases, with Lizheng's anal function scores[3] of 5 to 6 in 10 cases and 3 to 4 in nine cases. Moreover, 19 pediatric patients showed a poor anal appearance and functionality, including varying degrees of scarring, uneven distribution of external sphincters, anal stenosis, anal mucosa eversion, and or a retracted rectum. These patients had Lizheng's anal function scores of 0–4, including anal incontinence in three cases (score of 0). Preoperative voiding urethrography revealed a urethral fistula in all 38 pediatric patients (Fig. 1), and lower gastrointestinal contrast radiography revealed a urethral fistula in 36 cases (Fig. 2). All 38 pediatric patients underwent the first surgery within 1 week after birth because of aproctia, all with perineal anoplasty as the surgical procedure. A history of urethral discharge of stool was found in 23 children after birth and before the surgery, whereas this preoperative symptom was not found in 15 cases.

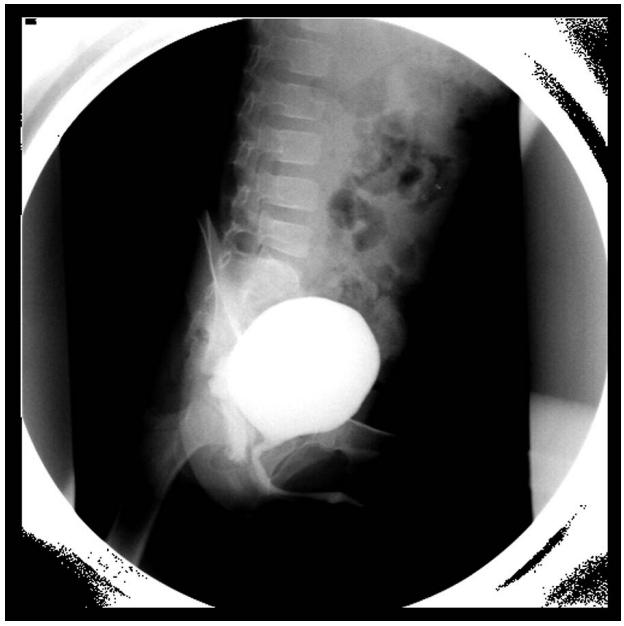

Fig. 1 Voiding cystography: a rectourethral fistula was observed preoperatively.

Surgical methods

Depending on the appearance and functionality of the anus, 2 different surgical procedures were applied. Among 6 of the pediatric patients, 5 exhibited severe rectal retraction, and 1 exhibited forward shifting of the anal opening (the external sphincter was located to the rear of the anal opening). The anal function scores ranged from 0–4 points (0 in 3 cases, 2 in 2 cases, and 4 in 1 case). Rectourethral fistula repair by posterior sagittal anorectoplasty was performed in these 6 cases. The remaining 32 patients had a good anal appearance and functionality, and rectourethral fistula repair using an anterior sagittal procedure was applied.

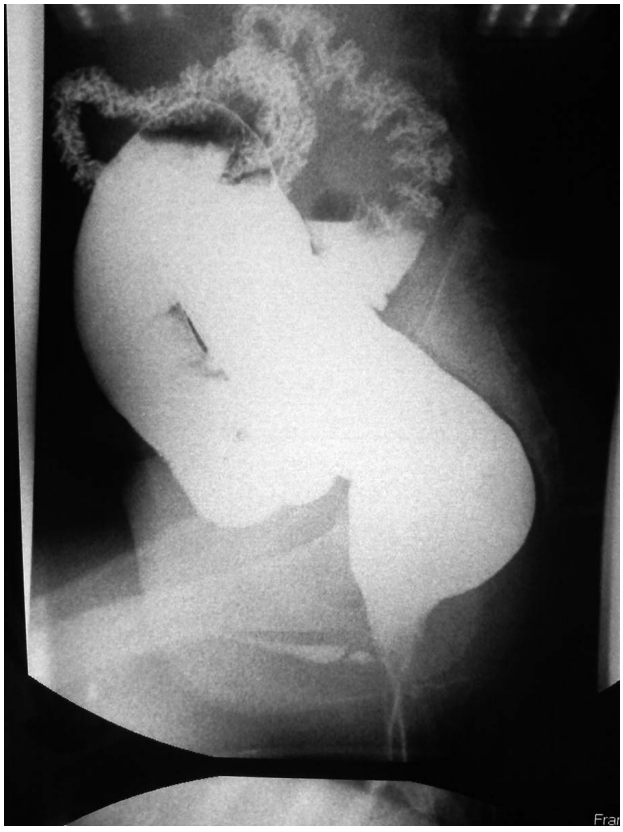

Fig. 2 Lower digestive tract contrast radiography: a rectourethral fistula was seen.

Rectourethral fistula repair by posterior sagittal anorectoplasty

A silicone Foley catheter was preoperatively applied. The patient was placed in the prone position. With a sagittal median approach from the bottom tip of the coccyx to the front of the anal opening, the circumference of the anal opening was cut at the junction of the mucosa and skin. With guidance from an electrical stimulator, an incision was created at the posterior midline of the sphincter complex and the levator ani muscle with marking, and the rectum was separated inward toward the proximal end. During the separation, the opening of the fistula in the rectum was probed. When the probe reached the rectal fistula, the distal rectal fistula was longitudinally cut at the median of the anterior rectal wall. Under direct visualization, the fistula opening was disassociated, and a pulling wire was sutured as the marker. The rectum was continuously separated inward toward the proximal end, with a length of approximately 5 cm, to allow the terminal rectum to reach the perineal skin with no tension. The rectal opening of the fistula was lifted to separate it toward the urethra. After the urethral opening was exposed, the fistula was resected. The rear wall of the urethra was released to allow transverse interrupted suturing of the full thickness of the fistula with no tension. The tissues surrounding the urethra were fully dissociated, which may have included the deep transverse perineal muscle and the urethral sphincter, and the urethral incision was longitudinally sutured and covered. Furthermore, the tissues of the levator ani muscles on both sides were sutured, and the urethra was covered again. The full thickness of the rectal wall received interrupted sutures in the submucosa. The retracted perianal skin and the external sphincter were released. With guidance from an electrical stimulator, the distribution of the external sphincter was redesigned. The terminal rectum was placed in the center of the external sphincter and was sutured to the released skin in the anal recess and the sphincter. The sphincter complex and levator ani muscle were resutured, forming the anus. The incision was then closed in layers.

Rectourethral fistula repair by anterior sagittal anorectoplasty

A catheter was preoperatively placed in the urethra; in certain cases, it may have entered the rectum (Fig. 3). The patient was placed in the lithotomy position. With the anterior anal sagittal approach, an incision was created from the rectal anterior wall to the rear of the scrotum. Subsequently, the skin, muscle tissue (which may have included the superficial transverse perineal muscle, the bulbospongiosus muscle, the deep transverse perineal muscle, the urethral sphincter muscle, and the anterior levator ani muscle) and the anterior rectal wall were cut from the superficial layer to the deep layer. In general, the rectal opening of the rectourethral fistula was first found within 3 cm of the anus. If the catheter had entered the rectum, the fistula was easier to find. A longitudinal incision was created in the rectal wall to reach the rectal opening of the fistula, and a pulling wire was then sutured around the rectal opening of the fistula with separation (Fig. 4). Subsequently, the rectourethral gap was separated close to the serosa on the anterior rectal wall toward the proximal rectum by approximately 2 cm. The pulling wire at the rectal opening of the fistula was lifted to separate the fistula from the posterior wall of the urethra. After the urethral opening was exposed, the urethra around the fistula was disassociated (Fig.

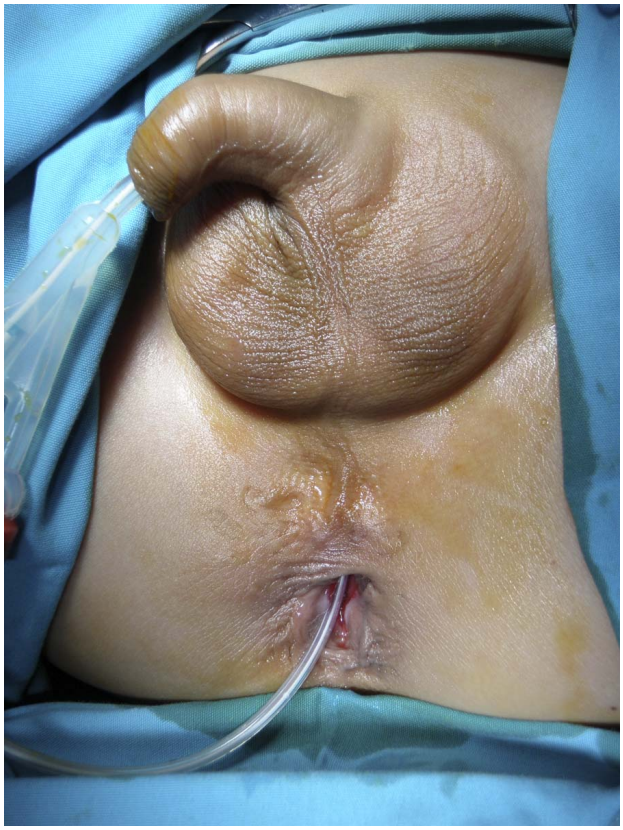

Fig. 3 A catheter was placed preoperatively into the rectum from the urethra.

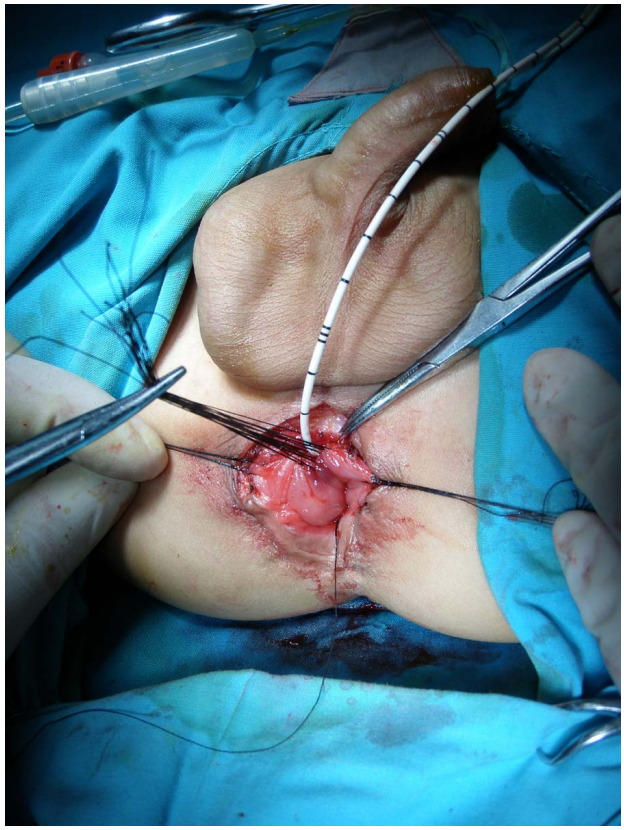

Fig. 4 The anterior sagittal approach was used to open the anterior wall of the rectum at the distal end of the fistula, expose the rectal fistula, and mark the suture site.

5). After the excision of the fistula, the full thickness of the urethra was transversely sutured for tension-free closure of the fistula. The perineal muscle tissue was sutured to cover the incision in the urethra. The levator ani muscles on both sides of the rectum were released, and the levator ani muscles (primarily the pubococcygeal muscle) were sutured to separate the urethral and rectal incisions. The longitudinal incision in the anterior wall of the rectum was sutured to the submucosal layer, and the dissociated rectum was pulled down so that the intact anterior rectal wall would cover the incision in the urethra. Under the guidance of an electrical stimulator, the open sphincter complex was distributed and sutured. The terminus of the rectum was sutured to the skin and the external sphincter to form an anus (Fig. 6).

Perioperative management

The patient fasted the day before the surgery, although drinking water was provided. Postoperatively, the patient fasted with no water for 3 days before defecation; eating was allowed after defecation. Anal treatments and therapies began on the second day after surgery to keep the incision clean and dry. Intravenous cephalosporins and metronidazole were applied for 5–7 days. Approximately 2 weeks after the surgery, the catheter was removed, and anal dilation and defecation training was initiated. In general, the anal dilator was gradually increased from size 9 (9 mm in diameter) to size 20 (size 16 for patients younger than 6 months). The training duration was 3–6 months.

Statistical processing and methods

The measurement data are presented as the mathematical mean ± SD. The scores for clinical anal function before and after surgery were classified as excellent, good, or poor. SPSS 19.0 statistical software was used for the statistical analysis, with t tests or χ^2 tests used for the data. A difference with $P < 0.05$ was considered statistically significant.

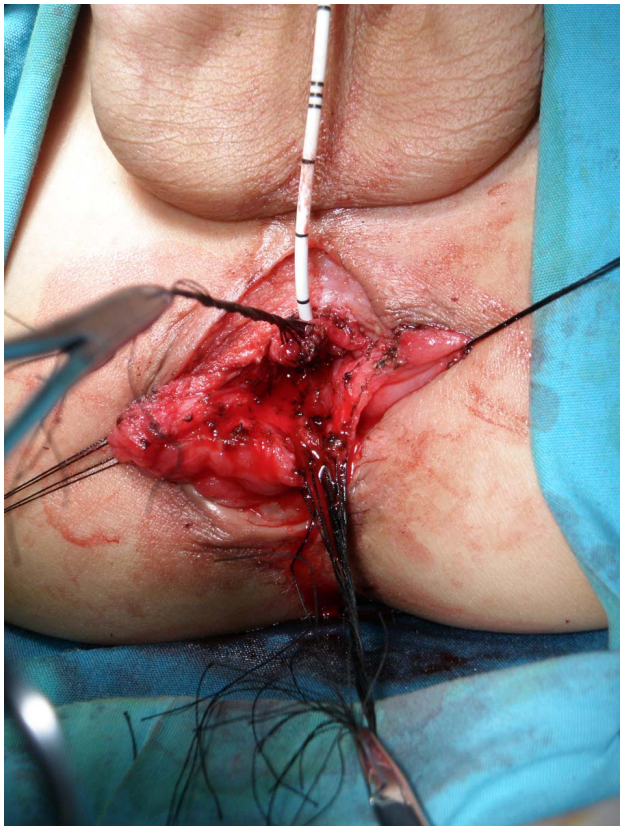

Fig. 5 Separation of the fistula.

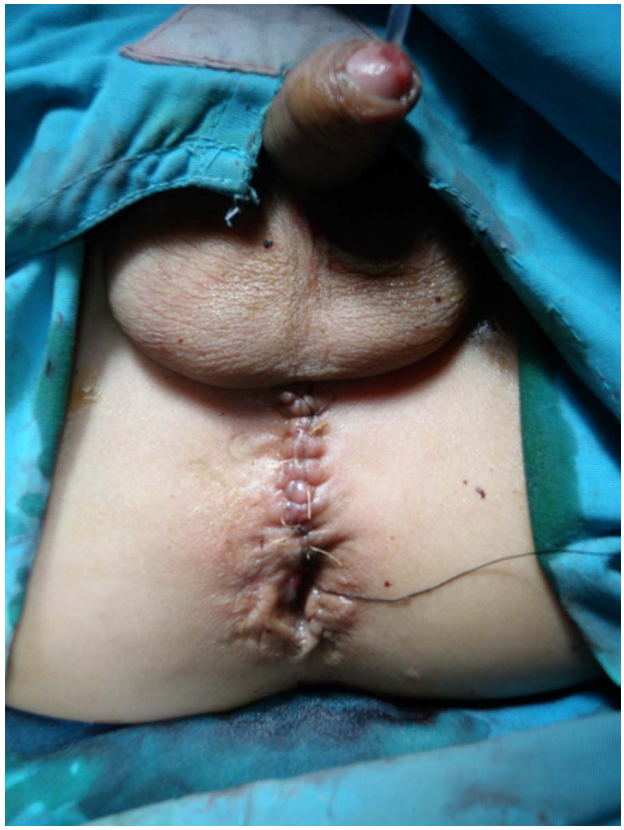

Fig. 6 At the end of the operation, the anus was located in the center of the external sphincter.

Results

The fistula between the rectum and the urethra observed during surgery was generally a horn-like structure with a large rectal fistula opening and a small urethral fistula opening, with lengths ranging from 0.5 to 2 cm and averaging 0.9 ± 0.4 cm. The urethral fistula openings were all located in the membranous portion or the distal side of the urethra, with diameters ranging from 0.5 to 1.0 cm and averaging 0.8 ± 0.2 cm. The distance from the perineal skin ranged from 0 to 3.5 cm and averaged 2.7 ± 0.6 cm. The rectal fistula diameter ranged from 0.5 to 2.5 cm and averaged 1.4 ± 0.6 cm. The distance from the anal skin ranged from 0 to 3.0 cm and averaged 2.1 ± 0.9 cm. Complete anterior urethral rupture occurred in 1 case, and urethral posterior wall breakage occurred in 1 case, with the proximal posterior wall of the urethra and the anterior rectal wall connected at the perineum.

The operative times for rectourethral fistula repair by posterior sagittal anorectoplasty and anterior sagittal anorectoplasty were 105.6 ± 14.6 minutes and 74.6 ± 10.1 minutes, respectively. A comparison of the operative times revealed a statistically significant difference ($t = 11.312$, $P = 0.031$, <0.05). The intraoperative blood loss was 1 to 3 mL, with no blood transfusion.

Two weeks after the surgery, the urethral fistulas were well healed in 37 cases. Recurrence of the urethral fistula occurred in 1 patient, whose condition was improved after 1 month according to telephone follow-up; the patient was subsequently lost to follow-up. Follow-up results were obtained for 35 patients after 1 month; these results revealed smooth urination, no urethral stenosis, and no diverticulum by voiding cystography (Fig. 7). The appearance of the anus was satisfactory, with no fecal incontinence. Lizheng's clinical anal function scores were 3 to 4 (good) in 9 cases and 5 to 6 (excellent) in 26 cases, with significant differences compared with the corresponding preoperative scores (Table 1).

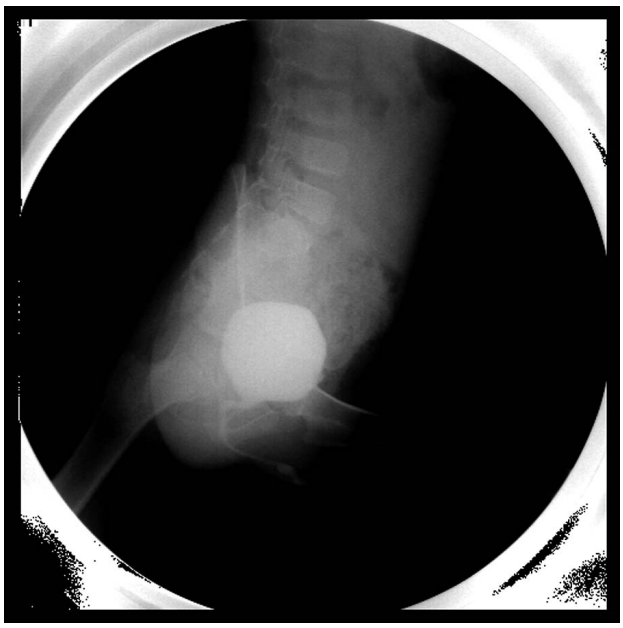

Fig. 7 Voiding cystography: no rectourethral fistula was observed postoperatively, and the urethra was slightly flexed, with no diverticulum or stenosis.

Discussion

Errors during surgery for male anorectal malformation with rectourethral fistula can cause residual rectourethral fistula following anoplasty. Certain pediatric patients are misdiagnosed with low anorectal malformation, and a simple perineal anoplasty is conducted. Certain surgeons deliberately use perineal anoplasty to treat median anorectal malformation combined with rectourethral fistula to form an anus first, and the urethral fistula is then treated in a second-stage operation. Performing the incorrect surgery for urethral fistula results in residual rectourethral fistulas. Perineal anoplasty only releases the posterior rectal wall, with insufficient separation achieved in most cases, which results in traction of the posterior rectal wall. Postoperative rectal retraction, anal stenosis, perineal scars, urethral injury, and other complications are frequently seen, and improper handling of the striated muscle complex could cause uneven distribution of the external sphincter, leading to anal dysfunction.

All pediatric patients included in the present study underwent perineal anoplasty in their first surgery, whereas only 19 cases showed a good anal appearance and function. The anal function of the other 19 cases was not satisfactory (scores of 0 to 4),

Table 1 Anal function scores before and after surgery[a]

Anal function score	Before surgery (n = 38)	After surgery (n = 35)
0–2	5	0
3–4	23	9
5–6	10	26

[a]For the comparison of poor, good, and excellent conditions before and after surgery, $\chi^2 = 18{,}143$; $P = 0.000$.

and 9 cases showed rectal retraction, including 5 cases of severe retraction. In 1 case, the anal opening was shifted forward and did not reach the external sphincter; the remaining 9 patients exhibited different degrees of uneven distribution of the external sphincter, anal stenosis, and or anal mucosa eversion. Urethral injury was intraoperatively found in 2 cases, including 1 case of complete rupture of the anterior urethra and 1 case of breakage in the posterior wall of the anterior urethra. Therefore, for pediatric patients with middle or high anorectal malformation who are considered to have rectourethral fistula, to reduce the occurrence of complications, perineal anoplasty should not be adopted.

For the treatment of postoperative residual rectourethral fistula following anoplasty, procedures with a posterior sagittal approach,[4] a perineal approach,[5] or an endorectal pull-through[6] are often applied. The 38 pediatric patients in the current study received urethral fistula repair using different surgical procedures based on the appearance and functionality of each patient's anus. For 6 pediatric patients (5 cases with severe anal retraction and 1 case of forward shifting of the anal opening) who had poor anal function, rectourethral fistula repair by posterior sagittal anorectoplasty could sufficiently release the rectum so that it could reach the perineal skin without tension, with redistribution of the sphincter complex, thus improving anal function. In our experience, although this surgical procedure allowed good exposure and could fully release the rectum, the operating scope was large, and the operative time was long. For the 32 pediatric patients with anal malformations that were less severe in appearance and who showed good anal function, rectourethral fistula repair via the perineal approach with anoplasty was performed. This surgical procedure included an incision between the posterior urethral wall and the anterior rectal wall so that the fistula opening could be accurately located. The fistula was gradually exposed in the rectourethral gap from the superficial to the deep layer, with a shallow surgical field and clear

exposure, and the urethra was not easily damaged. Additionally, the external sphincter could be partially repaired to improve anal function. The respective average surgical durations of the 2 surgical methods were approximately 105 and 74 minutes; posterior sagittal anorectoplasty thus required approximately 30 minutes more, suggesting that it is a more complex procedure.

Postoperative residual rectourethral fistula following anoplasty had an obvious tubular structure. The fistula was originally the end of the rectum, but because the posterior wall of the rectum was pulled and the posterior wall was cut to form the anus in the first surgical procedure, a fistula-like structure was formed at the rectal end between the urethra and the rectum. Furthermore, because the extent of pulling on the posterior wall of the rectum differed, the length and position of the resulting fistula differed. Therefore, in our experience, for most of the pediatric patients studied, the fistulas showed a horn-like structure, with a higher urethral end and a lower rectal end. This type of fistula should be explored from the rectal end during surgery, with separation toward the urethral end, to fully dissociate the distal and proximal urethral segments of the fistula opening through the urethral posterior wall. In the present study, after excision of the fistula, the urethral opening was transversely sutured in full thickness for a tension-free closure.

During urethral fistula repair, the appearance and function of the anus must be repaired. In our experience, anoplasty requires full disassociation of the rectum and rational allocation and repair of the sphincter complex. The perineal scar should be completely released, and the procedure needs to be carefully performed under the guidance of an electrical stimulator to prevent damage to the sphincter complex and to avoid postoperative rectal retraction, scar formation, and undesirable sphincter function. Because the anus is reshaped, the postoperative sphincter complex ring tends to be smaller, so daily anal dilatation and toilet training must be started within 2 weeks after the surgery to prevent anal stenosis. Among the 38 cases of pediatric patients in this study, preoperative anal function was unsatisfactory in 19 patients. After scar release and repair of the sphincter complex, the appearance of the anus was improved, and the function was improved significantly.

No matter which method was applied to treat the postoperative residual rectourethral fistula following anorectal surgery, complete resection of the fistula, repair of both ends of the fistula, and filling of the interval tissue between the urethra and the rectum during the surgery are key for a successful operation. A variety of surgical procedures for the treatment of rectourethral fistula all emphasize the importance of filling the interval tissue in between the rectum and the urethra. Considering the healing of the local scar tissue, the gracilis muscle or the rectal wall after removing the mucosa has often been used to separate the urethra and the rectum.[7–9] In the present study, the patients' first surgery was usually perineal anoplasty, and the scar tissue encountered in this surgery was restricted to the anal opening, with little residual scarring around the fistula, especially around the urethral opening of the fistula. Therefore, the filling tissue for the pediatric patients in this study was mostly the local deep transverse perineal muscle and the urethral sphincter near the fistula, along with the levator ani muscles (pubococcygeal muscle). The intact rectal wall was disassociated and dragged toward the proximal end so that the postoperative urethral incision and rectal incision were not on the same plane (the urethral incision was higher, whereas the rectal incision was lower), with no direct contact. This study included 38 pediatric patients, and 37 cases achieved success with the 1-stage surgery (1 case showed recurrence, with severe perineal scarring), indicating that the surgical efficacy was satisfactory and reliable with this filling tissue after fistula excision.

The complex operation on the perineal rectum and anus frequently requires proximal bowel fistulation and fecal bypass to ensure good perineal healing. However, for the 38 pediatric patients in this study, the surgery included posterior sagittal entry and perineal entry in front of the anus, and no intraoperative colostomy was performed; in this context, postoperative recovery was good. In our experience, apart from careful intraoperative work, minimizing operating field scarring, limiting the operation area to the terminus of the rectum, and performing careful postoperative local anal care are all important factors for preventing incision infection and dehiscence.

The choice of surgical procedure, that is, either posterior sagittal or anterior sagittal anorectoplasty, was dependent on the patients' anal appearance and functionality. We believe that it is better to choose posterior sagittal anorectoplasty in cases of a severely disordered perineal appearance, which is a relatively difficult and complicated operation that requires more operative time. In contrast, anterior sagittal anorectoplasty is best applied in patients

with minor alteration in perineal appearance; it has a clear surgical field and is easy to perform. In general, both surgical procedures achieved satisfactory results in the present study. In addition to the 2 surgical procedures discussed in this paper, laparoscopically assisted anorectal pull-through anoplasty[2] is also an option. According to the cited report, the advantages of this surgical procedure included excellent visualization of the rectal fistula and surrounding structures and minimally invasive abdominal and perineal wounds. However, we do not believe that this procedure is a good choice for postoperative residual rectourethral fistula following anoplasty because all our patients had experienced a botched operation, so the situation was relatively too complex to use this procedure.

Acknowledgments

This research did not receive any specific grants from funding agencies in the public, commercial, or not-for-profit sectors. The authors have no conflicts of interest to declare. Ethical approval was not required for this paper.

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