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RESEARCH ARTICLE

The Treatment of Congenital Recto-vestibular Fistula and Recto-perineal Fistula, and the Effects of the Megarectum on Defecation

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Abstract:

Objective:

This study aims to discuss the treatment of congenital recto-vestibular fistula and recto-perineal fistula, and the effect of the megarectum on defecation

Background:

Congenital recto-vestibular fistula or recto-perineal fistula is the most common type of anorectal malformation, and surgical methods include posterior sagittal anorectoplasty, anterior sagittal anorectoplasty, and mid-sagittal anorectoplasty, which can be performed at stage one or stage two after the ostomy. In the later stages of a recto-vestibular fistula, constipation is a common complication. Rectal dilatation is frequently associated with constipation, and the effect of rectal dilatation on defecation should be discussed for patients with congenital recto-vestibular or recto-perineal fistula who had rectal dilatation prior to surgery. Rectal dilatation may be one of the causes of constipation for congenital recto-vestibular fistula and recto-perineal fistula.

Methods:

The patients in this study were 67 children with congenital recto-vestibular fistula or recto-perineal fistula treated in our hospital from March 2013 to February 2017. All patients underwent an MRI of the spine and a barium enema. Six patients with myelodysplasia and sacral agenesis were excluded from this study. There were 18 patients with rectal dilatation (ages: 4-month-old to 1 year old, male: 3, female: 15). Seven of them had anterior sagittal anorectoplasty (group A), and 11 had anorectoplasty with dilated rectum resection (group B). Forty-three patients (ages: 3- to 10 months old, male: 6; female: 37) without a dilated rectum underwent anterior sagittal anorectoplasty (group C).

Results:

All patients were followed up for 1 year to 5 years. Among the 50 patients who had undergone an anoplasty, 5 out of 7 patients with rectal dilatation developed post-operative constipation, and 3 of them had normal defectation after the second resection of the dilated rectum. Only two out of 43 patients without rectal dilatation developed post-operative constipation. One out of 11 patients with rectal dilatation who underwent anoplasty and resection of the dilated rectum developed post-operative constipation.

Conclusion:

Patients with congenital recto-vestibular fistula or recto-perineal fistula complicated by rectal dilatation are more susceptible to post-operative constipation. Resection of the dilated rectum at the same time can reduce the incidence rate of constipation. A barium enema should be performed pre-operatively for patients with congenital recto-vestibular fistula or recto-perineal fistula. If the dilated rectum is found, it can be resected at the same time.

Keywords: Anorectal, Malformation, Recto-vestibular fistula, Recto-perineal fistula, Megarectum, Constipation.

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1. INTRODUCTION

Anorectal malformations occur in 1 in 5000 births and recto-vestibular fistula or recto-perineal fistula is the common type of anorectal malformations, and the surgical methods include posterior sagittal anorectoplasty (PSARP) [1], anterior sagittal anorectoplasty (ASARP) [2 - 4], Neutral sagittal anorectoplasty (NSARP) [5], and laparoscopic-assisted anorectoplasty for rectovestibular Fistula [6], which can be performed at stage one [5 - 7], or can be performed at stage two after the ostomy [1]. PSARP provides excellent exposure and precise placement of the rectum within the striated muscle complex. However, PSARP involves the division of the external sphincter, the muscle complex, and part of the lower portion of the levator. ASARP has many advantages over PSARP, such as an easier mobilisation of the rectum from the vagina under direct vision, the avoidance of levator ani division, and accurate reconstruction of both the sphincter muscle and the perineal body. ASARP involves the division of the anterior fibres of the external sphincter. NSARP preserves the perineal skin bridge and the levator muscle and contributes to the improvement of the aesthetic appearance of the perineum. However, it has the disadvantages of poor reconstruction of the perineal body and blind tunnelling of the anal sphincter. Anterior sagittal anorectoplasty is a simple, onestage procedure. The most important step, rectovaginal separation, takes place in direct vision; reconstruction of the perineum and the perineal body is simple. Recently, more surgeons have tended to do a one-stage procedure. The reasons are multiple: avoidance of multi-stage operations, saving time and costs, and less stress and trauma for children and their parents. Furthermore, anorectoplasty without a colostomy means avoidance of colostomy-related complications, such as peristomal excoriations, prolapse, parastomal hernias, leakages, intraabdominal adhesions, or even bowel obstruction.

Wound infection, wound dehiscence, anorectal stenosis, rectal prolapse, retraction, and anterior migration of the anus are early postoperative complications. Constipation is a common complication of the recto-vestibular fistula in its later stages. A megarectum is an enlarged rectum defined by a rectopelvic ratio greater than 0.61. Increased rectum volume and decreased rectal sensory and peristaltic function are present in children with megarectum. Constipation can happen due to faeces retention in the rectum. Rectal dilatation is often related to the occurrence of constipation, and some patients with congenital recto-vestibular fistula or recto-perineal fistula, even some neonates, have rectal dilatation before surgery. Patients with congenital recto-vestibular or recto-perineal fistula who have undergone rectal dilatation prior to surgery should be counselled on the potential adverse effects of this procedure on defecation because rectal dilatation is commonly associated with constipation. Constipation is a common symptom of both congenital recto-vestibular fistula and recto-perineal fistula, and may be caused by rectal dilatation. From March 2013 to February 2017, a total of 67 patients with congenital 'rectovestibular fistula and recto-perineal fistula were admitted to the hospital, 18 of whom had a very dilated and hypertrophic rectum. The study observed and evaluated the correlation between constipation and rectal dilatation.

2. MATERIALS AND METHODS

2.1. Clinical Information

This is a retrospective study. From March 2013 to February 2017, 67 patients with anorectal malformations including congenital recto-vestibular fistula and recto-perineal fistula were followed. The age, sex, spine MRI, barium enema, surgical methods, and defecation were reviewed. All of the patients had a barium enema and a spine MRI. Six patients had myelodysplasia or sacral agenesis, and the remaining 61 patients were included in the scope of this study. Barium enema showed 18 cases with rectal dilatation (Fig. 1), aged 4month-old to 1 year old, 3 males and 15 females. Seven patients underwent stage one anterior sagittal anorectoplasty (Group A), and 11 patients underwent resection of dilated rectum at the same time as anoplasty (Group B). Forty-three patients without rectal dilatation, aged from 3-month-old to 9month-old, including 6 males and 37 females, underwent anterior sagittal anorectoplasty (Group C).



Fig. (1). Barium enema showing rectal dilatation.

2.2. Surgical Methods

During the operation, the lithotomy position was adopted, the urinary catheter was indwelled, the center of the striated muscle complex was stimulated by electricity, the traction line at the periphery of the fistula was sutured, a circular incision was made along the fistula and a mid-sagittal incision was made posteriorly from the fistula to the center of the striated muscle complex. The skin was incised subcutaneously to the posterior wall of the rectum and the anterior part of the anal sphincter to reveal the striated muscle complex (Fig. 2). The free rectum and surrounding tissues were about 3-4 cm,

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especially the posterior vaginal wall. In 11 cases of patients with rectal dilatation, the dilated bowel was removed (Fig. 3). After the bleeding was completely stopped, the free rectum was placed in the center of the striated muscle complex, and 5-0 absorbable sutures were used to reconstruct the perineal body and the anterior part of the striated muscle complex. A 5-0 absorbable suture was used to suture the end of the rectum and the surrounding skin to form the anus, and intermittently suture the anal sphincter skin.



Fig. (2). Suture the traction line around the fistula to make a circular incision along the fistula and mid-sagittal incision from the fistula to the back to the center of the sphincter-striated muscle complex.



Fig. (3). The dilated rectum and sigmoid colon were pulled out and resected

Prophylactic antibiotics were applied for 2 days after the operation. On the first day after the operation, Alfare enteral nutrition and Diosmectite were taken orally to reduce the frequency of defecation. Bilateral thigh abduction exposes the

perineum, and the perineum was washed with normal saline and iodophor every four hours or after defecation to keep the perineum dry. Anal dilation was started 2 weeks after the surgery. The chi-square test was used to compare the defecation. Constipation was defined according to the Rome IV criterion.

3. RESULTS

Among the 50 patients who underwent anoplasty alone, 5 out of 7 patients with rectal dilation developed post-operative constipation, and 3 of them had voluntary bowel movements after resection of the dilated rectum. Of the 43 patients without rectal dilatation, 2 had developed constipation and 2 had soiling after the operation. Of the 11 patients with rectal dilation who underwent anoplasty and resection of the dilated rectum, 1 had developed constipation and 1 had soiling after the operation. Among the 50 patients who underwent anoplasty alone, the incidence of post-operative constipation in patients with rectal dilatation was significantly higher than that in patients without rectal dilatation (p = 0.00). Meanwhile, of the 18 patients with rectal dilatation, the incidence of constipation in patients who underwent both anoplasty and resection of dilated rectum was lower than that in the 7 patients with anoplasty alone (p = 0.013), as shown in Table 1. Finally, there was no significant difference in the fecal incidence between the 47 patients who underwent anoplasty alone and the 14 patients who underwent both anoplasty and dilated rectal resection, as shown in Table 2.

Table 1. Comparison of post-operative defecation function in 61 patients

Group	Number of cases	Constipation	No constipation
Group A	7	5	2
Group B	11	1	10
Group C	43	2	41

Note: Comparison of A and C, P=0.00, Comparison of A and B, P=0.013 (Fisher exact probability method)

Table 2. Comparison of post-operative bowel control function in 61 patients

Surgical method	Number of cases	Feces incidence	No feces incidence
Anoplasty	50	2	48
Anoplasty + Rectal dilatation resection	11	1	10

Note: $X^2 = 0.00$, (continuously corrected by chi-square test)

4. DISCUSSION

Congenital recto-vestibular fistula and recto-perineal fistula are common congenital anorectal malformations. In 1982, de Vries and Pena [1] proposed a posterior sagittal anorectoplasty, with a mid-sagittal incision from the sacrum to the fistula. The advantages are that the exposure is good, the pelvic floor muscle structure can be reconstructed under direct vision, and the incision is located in the middle to avoid damage to the pelvic floor nerves and blood vessels. In 1992, Okada *et al.*. [2] proposed anterior sagittal anorectoplasty, that the anterior part of the sphincter complex was incised. The

anterior sagittal approach is less invasive and will provide better exposure for rectal and vaginal dissociation. In addition, Dave and Shi proposed an anal displacement technique that preserves the skin bridge between the anus and the fistula, named the mid-sagittal anorectoplasty [5], with the main advantage of not damaging the skin and tissue between the anus and vagina. The laparoscopic-assisted anorectoplasty technique has shown several unique strengths, including lower risks of complications, and minimal muscular injury, with a comparable bowel function [6]. It was originally advocated that the operation should be done in three stages [1]. The purpose of this was to avoid the occurrence of infection during anal reconstruction, and the consequent anal retraction, anal stenosis, and fistula recurrence. In recent years, more and more people advocate stage one surgery despite the fact the risk of incision infection is higher after stage one surgery. However, it can reduce the number of operations, thus reducing the cost of treatment and avoiding skin erosion, wound infection, prolapsed stoma or stenosis, water and electrolyte disturbances, and other complications related to ostomy [7 - 9]. Furthermore, adequate bowel preparation prior to surgery, as well as antibiotic prescriptions before and after surgery, results in no significant difference in complication rate or treatment effect from staged surgery [8 - 10]. Regarding the timing of surgery, many people believe that it can be completed in the neonatal period [8, 9]. Wakhlu et al.. [8] performed stage one anterior sagittal anorectoplasty in 1206 children, 267 of which were completed in the neonatal period and there was no surge in complication rate. On the other hand, anal reconstruction in the neonatal period contributes to the early establishment of the defecation reflex, pelvic floor muscle training, and the formation of synapses and neural networks, resulting in normal or near-normal anal function [11]. Some surgeons thank delayed single-stage repair of rectoperineal and rectovestibular fistulae can be performed safely in infants beyond the newborn period [12, 13].

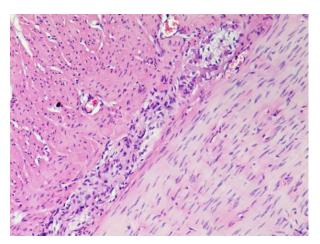


Fig. (4). The nerve fibers in the myenteric plexus of the dilated bowel were proliferated and a few small ganglion cells can be seen (×40).

Constipation is the most common late post-operative complication of congenital recto-vestibular and recto-perineal fistulas [14 - 17]. Levitt *et al.* [15] reported that the incidence of constipation was 55% while Zhang Yanan *et al.* [16]

reported that the incidence of constipation after anterior sagittal anorectoplasty was 57.7%. Defecation control involves several different neural pathways and the synergy and interaction of the pelvic floor muscles and the anorectum. The factors of defecation control include stool volume and viscosity, colonic transit function, rectal compliance, tone and volume, rectal movement and emptying ability, anorectal angle, anorectal sensation, and reflex mechanism, movement of the anal canal, and integrity of pelvic floor muscles. Anal stenosis, rectal and sigmoid colon dilatation [18], pelvic diaphragm dysplasia [19], pelvic floor muscle incoordination [20, 21], and myelodysplasia may cause post-operative constipation [22].

Megarectum or megarectal colon is a common cause of intractable constipation and fecal incontinence after anoplasty [23]. Patients with rectal and sigmoid colon dilatation have increased rectal volume, decreased rectal sensation and peristalsis function, and feces are easily stored in the rectum, leading to constipation. Therefore, it is advocated that the resection of the dilated bowel can obtain satisfactory results [24]. In the group of 18 patients with rectal dilatation, 7 patients only underwent anoplasty in the early stage, and 5 patients had difficulty in defecating post-operation. Among them, 3 patients no longer had constipation after the second operation to remove the dilated bowel. Of the 11 cases at the later stage, the dilated rectum was excised at the same time as anoplasty, and only 1 case developed constipation postoperation. In addition, among the 50 patients who underwent anoplasty alone, the incidence of post-operative constipation in 7 patients with rectal dilatation was significantly higher than that in 43 patients without rectal dilation, indicating that rectal dilatation is the main cause of constipation after the surgeries of recto-vestibular and recto-perineal fistulas. Removal of the dilated bowel at the same time of anoplasty can prevent the occurrence of post-operative constipation. In addition, there was no significant difference in the incidence of soiling between the 14 patients who underwent dilated rectal resection and the 47 patients who underwent only anoplasty, indicating that resection of the dilated rectum did not increase the incidence of fecal incontinence. The cause of rectal dilatation in patients with recto-vestibular or recto-perineal fistula is unclear. The group of patients with pre-operative rectal dilatation was treated late, and no treatment such as enema or fistula dilatation was given before the operation, and rectal dilatation may be the consequential change due to poor bowel movements. De la Torre et al.. reported that some patients had rectal dilatation during the neonatal period [25]. The pathological results of this group showed that nerve fibers in the myenteric plexus of the rectum and distal colon were proliferated, and a small number of ganglion cells were seen (Fig. 4). Therefore, rectal dilatation may be the consequence of enteric nerve dysplasia. For patients who are diagnosed with recto-vestibular fistula and recto-perineal fistula after birth, the operation can be completed in one stage in the neonatal period. If the operation is delayed, the fistula expansion and enema should be given before the operation to avoid a secondary megarectum. De la Torre et al. [25] reported that 60% of the cases were complicated by rectal dilatation. Barium enema was an important method to evaluate the anatomy of the rectum and sigmoid colon. Therefore, patients with recto-vestibular fistula

and recto-perineal fistula should undergo barium enema before surgery to know whether rectal and sigmoid colon dilatation exists in order to perform dilated rectal resection while undergoing anoplasty for patients with rectal dilatation. Bowel management was the common and effective method to treat constipation, but it was a long-term treatment. Our study indicates resection of the dilated rectum at the time of anoplasty can avoid constipation.

CONCLUSION

Congenital recto-vestibular fistula and recto-perineal fistula are the most common congenital anorectal malformations. Constipation is the most common late post-operative complication of congenital recto-vestibular and recto-perineal fistulas. After anal atresia surgery, a common cause of intractable constipation and faecal incontinence is the megarectum or megarectal colon. Patients with congenital recto-vestibular fistula or recto-perineal fistula complicated by rectal dilatation are more likely to experience post-operative constipation, according to our findings. Concurrent resection of the dilated rectum can reduce the frequency of constipation. For patients with congenital recto-vestibular or recto-perineal fistulas, a barium enema should be performed prior to surgery. If a dilated rectum is discovered, it can be removed at the same time.

LIST OF ABBREVIATIONS

PSARP = Posterior Sagittal Anorectoplasty

ASARP = Anterior Sagittal Anorectoplasty

NSARP = Neutral Sagittal Anorectoplasty

ETHICS APPROVAL AND CONSENT TO PARTICIPATE

This study was approved by the Ethics Committee of the Xuanwu Hospital, Capital Medical University, Xicheng District, Beijing, China China (Reference no. 2016037).

HUMAN AND ANIMAL RIGHTS

No animals were used in this research. All procedures performed in studies involving human participants were in accordance with the ethical standards of institutional and/or research committees and with the 1975 Declaration of Helsinki, as revised in 2013.

CONSENT FOR PUBLICATION

Written informed consent was obtained from the patient for the publication of this report and any accompanying images.

STANDARDS OF REPORTING

STROBE guidelines have been followed for this study.

AVAILABILITY OF DATA AND MATERIALS

The dataset used and/or analyzed during the present study are available from the corresponding authors [X.S].

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CONFLICT OF INTEREST

The authors declare no conflicts of interest, financial or otherwise.

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Declared none.

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