



Rectovestibular fistula with normal anus: a simple resection or an extensive perineal dissection?

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Abstract

Purpose: The purpose of the study was to review a 10-year experience of operative procedures for rectovestibular fistula with normal anus (RVFNA) at one institution.

Materials and Methods: From January 1999 to December 2008, 182 female patients of RVFNA were treated surgically in the Department of Surgery, Beijing Children's Hospital, Capital Medical University, China. The patients' age ranged from 4 months to 15 years, with a mean age of 3.4 years. Fourteen children had a failed previous operation in other institutions. One hundred fifty-six patients (85.71%) had a definite history of vulvar inflammation within 3 months after birth and fecal leakage thereafter. In 61 of the 156 patients, a perineal abscess appeared after an episode of diarrhea. Four cases (2.20%) had associated anomalies of the VACTERL type. One hundred seventy-three patients (95.05%) had 1 external opening, whereas 9 others (4.95%) had 2 external openings separated by a skin bridge. In all cases, the internal orifice opened above the dentate line. The fistula in 135 cases (74.18%) presented as an opening with a small diameter (<5 mm). In our series, vestibular-rectal pull-through procedure was performed in 98 cases, transanal procedure in 69 cases, and anterior perineal anorectoplasty in 15 cases. None had a diverting colostomy.

Results: Recurrence of fistula occurred in 5 to 10 days after operation in 21 cases (11.54%). Eight of them healed spontaneously after daily sitz bath with 3% boric acid, whereas other 13 patients (7.14%) required reoperation. Follow-up was obtained by telephone or by personal visit in the outpatient department, from 3 months to 10 years (median, 5.7 years). All the patients were continent and had regular bowel movement.

Conclusions: Most RVFNA was acquired after infection. Procedure requiring extensive perineal dissection and diverting colostomy are unnecessary in most RVFNA cases. We performed simple resection in most patients with satisfactory results. In the cases with a large external opening and abnormal perineal appearance, a more extensive perineal dissection might be necessary.

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Rectovestibular fistula with normal anus (RVFNA) is an abnormal communication between the normal rectum and vestibule in girls, sometimes associated with fecal leakage.

The etiology of this condition may be either congenital or acquired. Various surgical procedures, including simple fistula resection and extensive perineal dissection, have been recommended for this condition, and postoperative fistula recurrence is not infrequent. From January 1999 to December 2008, 182 patients with RVFNA were treated

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surgically in the Department of Surgery, Beijing Children's Hospital, Capital Medical University, China, and the different operative procedures performed were reviewed.

1. Materials and methods

1.1. Clinical characteristics

The patients' age ranged from 4 months to 15 years, with a mean age of 3.4 years. All patients were admitted with the complaint of passing stool or flatus through the vestibule. Fourteen children had a failed previous operation in other institutions. One hundred fifty-six patients had a definite history of vulvar inflammation within 3 months after birth and fecal leakage from vestibule thereafter. The other patients had a vague history of inflammation, but none had a history of the fistula discovered at the time of birth. In 61 of the 156 patients, a perineal abscess appeared after an episode of diarrhea. Four cases had associated anomalies of the VACTERL type. One hundred seventy-three patients had 1 external opening, whereas 9 others had 2 external openings separated by a skin bridge. Most of the external openings were located at navicular fossa (Fig. 1). No opening was found in the vagina. The internal orifices in all cases were seen in the anterior rectal wall 0 to 2.0 cm above the dentate line (Fig. 2). The fistula in 135 cases presented as an opening with a small diameter (<5 mm; Table 1).

1.2. Perioperative management

Preoperative bowel preparation consisted of oral metronidazole (10 mg/kg) for 3 days, liquid diet for 1 day, and saline enemas the night before and on the morning of operation. In the operating theater, a sterile bandage soaked in iodophor solution was packed deeply into the rectum to sterilize the anorectal canal and prevent fecal soiling during the operation.



Fig. 1 A probe in the fistula between rectum and vestibular fossa.



Fig. 2 The internal opening located at the anterior rectal wall.

1.3. Operative procedure

Three different operative procedures were used for the 182 cases, but no diverting colostomy was performed either before or after the procedure.

- (1) Vestibular-rectal pull-through procedure was performed in 98 cases (Fig. 3). The fistula tract was

Table 1 Clinical characteristic of 182 girls with RVFNA

| | n = 182 | % |
|------------------------------|---------|-------|
| Vulvar inflammation | | |
| Definite history | 156 | 85.71 |
| Vague history | 26 | 14.29 |
| After diarrhea | 61 | 33.52 |
| Previous operation | 14 | 7.69 |
| Associated anomalies | 4 | 2.20 |
| Spina bifida | 3 | 1.65 |
| Atrioseptal defect | 1 | 0.55 |
| Fistula external opening | | |
| Single | 173 | 95.05 |
| Double with skin bridge | 9 | 4.95 |
| External orifice sites | | |
| Navicular fossa | 142 | 78.02 |
| Right labia majora | 12 | 6.59 |
| Left labia majora | 17 | 9.34 |
| Other place of vestibule | 11 | 6.04 |
| Internal orifice sites | | |
| Above the dentate line | 182 | 100 |
| Below the dentate line | 0 | 0 |
| Fistula diameter (mm) | | |
| <5 | 135 | 74.18 |
| >5 | 47 | 25.82 |
| Abnormal perineal appearance | 15 | 8.24 |
| Operation age (y) | | |
| <1 | 4 | 2.20 |
| 1-2 | 21 | 11.54 |
| 2-4 | 133 | 73.08 |
| >4 | 24 | 13.19 |



Fig. 3 The fistula tract was freed and excised through vestibulum.



Fig. 4 The fistula tract was freed and excised through rectum.

freed and excised from the surrounding muscles through the vagina. The rectal wound was sutured transversely with a continuous absorbable suture (without tension), whereas the transverse perineal muscle was sutured longitudinally with interrupted sutures to eliminate a dead space anterior to the rectum.

- (2) Transanal procedure was performed in 69 cases (Fig. 4). The fistula was dissected to include the inner opening in the rectum and the opening in the vestibule. The transverse perineal muscle was sutured through the inner opening on the anorectal wall. A tension-free suturing of the rectal wall and longitudinal suture of the transverse perineal muscle are also essential points of this procedure.
- (3) Anterior perineal anorectoplasty was used in 15 cases with a large external opening and abnormal perineal appearance (Fig. 5). An incision was made around the orifice and extended posteriorly along the midline leaving an intact 8- to 10-mm skin bridge anterior to anus. (The perineal body was kept intact). The external

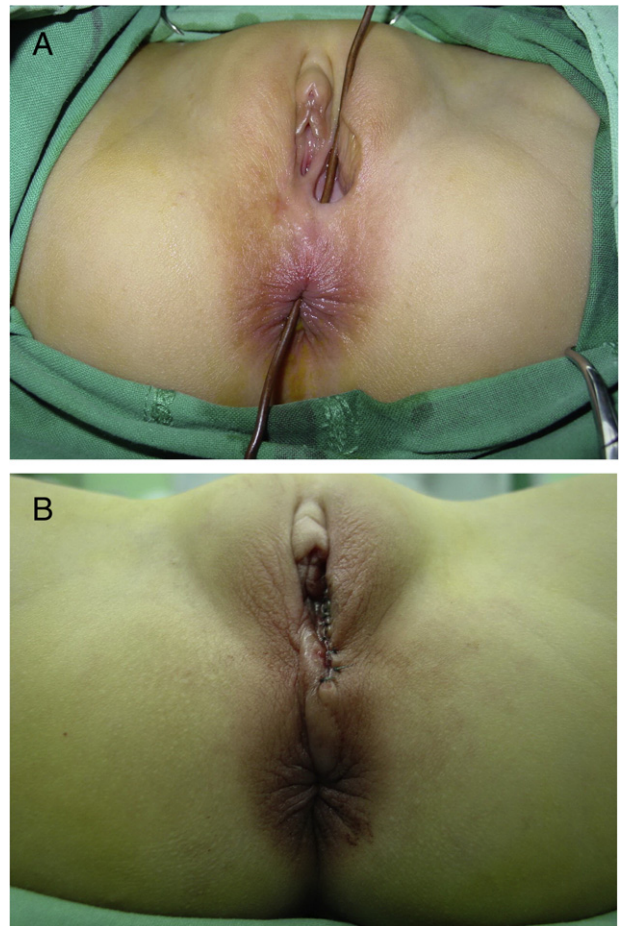


Fig. 5 A, A large external opening located at left labia majora appearing abnormal perineum. B, The perineum was reconstructed leaving an intact skin 8 to 10 mm anterior to anus.

sphincter muscle was incised to expose the anterior wall of the rectum. After the fistula tract was dissected out, the perineum was reconstructed layer by layer with interrupted 5/0 polyglactin (Dexon) sutures.

Postoperative fasting and intravenous antibiotics were continued for 3 to 5 days. The perianal area was kept clean and dry by frequent local cleansing as well as TDP-Lamps radiation twice per day.

2. Result

Recurrence of fistula occurred in 5 to 10 days after operation in 21 cases (11.54%). This was treated with sitz baths containing 3% boric acid thrice per day for 6 to 8 weeks. Eight cases healed spontaneously after daily sitz baths alone, whereas the other 13 patients (7.14%) required reoperation. (Table 2) There were no instances of perineal body dehiscence after the operation. Reoperations were performed at least 6 months after the initial fistula recurrence. Reoperations included vestibule-rectal pull-through procedure in 9 cases, transanal procedure in 3, and anterior perineal anorectoplasty in 1 patient. There were no second recurrences.

Follow-up was obtained by telephone or by personal visit in the outpatient department, from 3 months to 10 years (median, 5.7 years). All of the patients were continent and had regular bowel movements.

3. Discussion

Rectovestibular fistula with normal anus is a relatively common condition. The reported incidence is 2.24% to 7.1% of all anorectal malformations (ARFs) [1-4]. In this study, we identified 182 cases of RVFNA of the 1274 cases with ARF treated surgically in our hospital during the past 10 years. The incidence rate of RVFNA in our facility was 14.29%.

Rectovestibular fistula with normal anus has been considered a congenital ARF in some reports and has also been referred to as H-type anorectal fistula [1,5,6], N-type fistula [7], or double termination of the alimentary tract [2,3,8]. Other investigators believe that it is an acquired condition [9-12]. Sun et al [13] compared the histologic findings in a group of 19 patients with fistula and a normal anus to another group of 10 having a fistula associated with imperforate anus and suggested that most RVFNA originated from a vulvar abscess. In our series, microscopic examination of the tissue specimens showed granulation tissue and epithelial lining of the excised fistula tract in some cases, but none showed a surrounding smooth muscle coat or anal glands. Furthermore, we noted that 156 (85.71%) of our patients had a definite history of vulvar inflammation before fecal discharge from the vestibule was observed. The others had a vague memory of vulvar inflammation, but none of them had a definite history of fistula at birth. Another interesting observation was that an episode of diarrhea occurred before the development of a perineal abscess in 33.52% of the patients. These findings were more consistent with an inflammatory origin. Moreover, associated anomalies were common in the report of Rintala et al [1] (60%) but rare in our series. Therefore, we believe that most RVFNA that we encountered were acquired after the infection. Rectovestibular fistula with normal anus seems to be more common in Asia. We think that the method of caring for babies and sanitation in this region may be implicated in the development of acquired fistula. Too frequent changing of diapers and wiping the anus forcefully with cloth diapers may potentially irritate the anal mucosa, particularly at the anal crypts. However, we recognize that this is conjectural and have no definite proof of this causality.

Various procedures have been performed for this condition, but there is still no consensus on the operation of choice. (Table 3) We divided the procedures for RVFNA into 2 groups: simple resections and extensive perineal dissections. The simple resections, such as vestibule-rectal pull-through and transanal procedure, excised the fistula with no perineal incision. In the anterior perineal anorectoplasty,

Table 2 Postoperative results in 182 girls with RVFNA

| | Operative procedures | | | | | |
|-----------------------------|--|-------|---------------------|-------|----------------------------------|-------|
| | Vestibular-rectal pull-through procedure | | Transanal procedure | | Anterior perineal anorectoplasty | |
| | n = 98 | % | n = 69 | % | n = 15 | % |
| Fistula recurrence | 8 | 8.16 | 11 | 15.94 | 2 | 13.33 |
| Healed by sitz bath | 4 | 4.08 | 4 | 5.80 | | |
| Reoperation | 4 | 4.08 | 7 | 10.14 | 2 | 13.33 |
| Histopathologic examination | | | | | | |
| Granulation tissue | 64 | 65.31 | 42 | 60.87 | 13 | 86.67 |
| Epithelial lining | 27 | 27.55 | 18 | 26.09 | 9 | 60.00 |

In the total of 182 girls, recurrence occurred in 21 cases (11.54%), and reoperation was performed in 13 cases (7.14%); histopathologic examination of postoperative specimens showed granulation tissue in 119 cases (65.38%) and epithelial lining in 54 cases (29.67%).

Table 3 Literature review: various procedures been performed for RVFNA

| Procedures | Year | Author | Cases | Colostomy | Recurrence |
|--|------|-------------------------------|-------|-----------|------------|
| Vestibulorectal pull-through | 1969 | Chatterjee [2] and Chatterjee | 9 | 0 | 2 |
| | 1980 | Talukder [8] | | | |
| | 1984 | Tsuchida et al [3] | 3 | 0 | 2 |
| | 2008 | Banu et al [4] | 2 | 2 | 0 |
| Anterior perineal resection | 1978 | White et al [7] | 2 | | |
| Anterior wall of rectum pull-through | 1984 | Tsuchida et al [3] | 7 | 4 | 0 |
| | 2008 | Banu et al [4] | 22 | 0 | 5 |
| Posterior midsagittal approach | 1997 | Keramidas et al [6] | 1 | 1 | 0 |
| Anterior sagittal anorectovaginoplasty | 1998 | Kulshrestha et al [11] | 13 | 1 | 0 |
| | 2003 | Yazlcl et al [5] | 1 | 1 | 0 |
| | 1999 | Tsugawa et al [14] | 19 | 4 | 1 |

excision of the fistula with total or partial perineal dissection (layer by layer) is required.

Most of the fistulae had a small diameter (1~5 mm). Leakage at the vestibular opening was considerable in the early months of life but improved later on. In some cases, fecal leakage only occurred in the presence of diarrhea. Incontinence was negligible, and the parents were encouraged to pursue operative treatment mostly for psychologic reasons. Therefore, when discussing the risks and benefits to the families, performance of a colostomy and its subsequent closure or the risk of a perineal wound dehiscence in cases of extensive perineal dissection procedures would be unacceptable. For those patients with a small diameter fistula, a simple resection was perceived as a better choice.

In our series, we preferred to perform simple resection procedures (transanal or vestibular-rectal pull-through procedure), which provided satisfactory results. The timing for the operation is when the patient is about 3 years old when the perineal body is better developed, leakage is minimal, and vulvar inflammation is negligible. The main advantage of the simple resection procedure was a complete excision of the fistula under direct vision without incision of perineal body. For cases with a deep external opening, which is difficult to expose, we choose the transanal repair of the fistula to avoid a fourchette incision. Anterior perineal anorectoplasty was reserved for cases with a large external opening and abnormal perineal appearance, which requires reconstruction. One should avoid opening the perineal body when possible. We have never done a diverting colostomy in any of these patients. An initial diverting colostomy results in the need for at least 3 operations, which seems to be excessive in these cases. In our series, even those patients with fistula recurrence only had 2 operations.

The fistula recurred in 21 cases, most of which had a previous operation, or abundant inflammatory tissue around the internal opening. Recurrence was more frequently encountered in cases managed by anterior perineal anorectoplasty. Those cases usually had a very large fistula surrounded by abundant hyperplastic tissue. Local ischemia and inflammation may be the main factors associated with

fistula recurrence. Hence, based on our experience, the fistula should be resected completely along with any surrounding inflammatory hyperplastic tissue, leaving a fresh wound with clean edges. Besides, the intrarectal sterile pack to prevent soiling during the procedure, electrocautery provided hemostasis, and use of absorbable suture and perioperative wound care treatments are helpful in avoiding infection and recurrence. Recurrence healed spontaneously in some cases (n = 8) that had less extensive procedures and a small fistula. Sitz baths seemed to be beneficial in promoting spontaneous healing.

Procedures requiring extensive perineal dissection and diverting colostomy are unnecessary in most RVFNA cases. We performed simple resection in most patients with satisfactory results. In the cases with a large external opening and abnormal perineal appearance, which requires reconstruction, a more extensive perineal dissection might be necessary.

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