

Surgical Complications and Functional Outcomes in Children with Anorectal Malformations with Vestibular Fistula

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Background: Vestibular fistula is one of the most common types of anorectal malformations in female infants. Colostomy prior to definitive anorectoplasty is still controversial.

Objective: To compare the complication rates and functional outcomes of the surgical treatments of vestibular anus between colostomy prior to anorectoplasty and treatment without colostomy.

Materials and Methods: The medical records of 40 patients with vestibular fistula that underwent definitive surgical treatments at Siriraj Hospital between January 2006 and December 2018 were reviewed. Demographic data, operative records, functional outcomes, and complications were analyzed.

Results: In the 40 vestibular fistula patients, colostomy (C) prior to vestibular fistula repair was performed in 18 patients, while vestibular fistula was repaired without colostomy (NC) in the other 22 patients. The C group had more associated anomalies than the NC group, including GI tract anomalies at 23.5% versus 9.1%, spinal anomalies at 47.1% versus 9.1%, and KUB anomalies at 52.9% versus 9.1%. The definitive anorectoplasty for vestibular fistula were anal transposition (31, 77.5%), PSARP (5, 12.5%), and other operations (7, 10%). Anal transpositions were performed in both the NC (90.9%) and C (61.1%) groups, respectively. NC had higher complications than C at 36.4% versus 22.2%. The recurrence of fistula occurred in 9.1% of NC cases versus 0% in C. Anal stricture was found in 18.2% of NC cases versus 5.6% in C. The wound infection rates of these two groups were the same. The complication rate with colostomy was 22.2% with prolapsed colostomy in 11.1% of cases and wound infection in 5.6%. The NC group had higher complication rates of anorectoplasty than the C group, but the overall complications, including colostomy complications, of these two groups were comparable. The Krickenbeck scoring system was used to evaluate the functional outcomes. In the median follow-up period of 1,336 days and 23 patients were evaluated. Even though the NC group had higher complication rates of anorectoplasty than the C group, the functional outcomes of these two groups were the same.

Conclusion: Vestibular fistula repair with prior colostomy had lower complication rates of anorectoplasty than primary vestibular fistula repair, but the functional outcomes of these two groups were the same.

Keywords: Vestibular; Rectovestibular; Anovestibular

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Vestibular fistula is the most common type of anorectal malformation in females. Vestibular fistula is diagnosed when the anal opening is located at the vulval vestibule, in an outer location to the hymen

but inner to the posterior labial minora commissure.

To clarify the classification of anorectal malformation, the International Conference for the Development of Standards for the Treatment of Anorectal Malformations at Krickenbeck developed a new classification called the “Krickenbeck classification”⁽¹⁾. In this classification, there is only “vestibular fistula”. The authors used this classification in the present study.

Although this defect has a good prognosis of functional outcomes, the surgical treatments are still debatable about which surgical technique would be the best choice for the management of vestibular fistula. Over the last decade, multistage surgery, such as primary colostomy followed by posterior sagittal anorectoplasty (PSARP), had been used for the

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treatment of this type of anorectal malformation⁽²⁻⁴⁾. The primary anorectoplasty techniques such as anal transposition, minimal PSARP, anterior sagittal anorectoplasty (ASARP), and other anoplasty offer benefits in terms of a reduction in the number of operations needed. The primary anorectoplasty techniques also avoid the complications that can arise with colostomy and have reported good safety, which led to good functional outcomes⁽⁴⁻¹⁰⁾. On the other hand, staged operations beginning with colostomy⁽²⁻⁴⁾ still has advantages such as 1) adequate decompression, 2) adequate bowel preparation before definitive anorectoplasty, and 3) decreased fecal contamination at the surgical site and able to carry out perfect anocutaneous anastomosis.

Colostomy can prevent fecal contamination⁽⁴⁾. If the perineal wound were to become infected, dehiscence of the anal anastomosis or perineal body and recurrent vestibular fistula might occur. Following an infected anorectoplasty wound, the anal sphincter would have poor function from intense fibrosis. Regarding this vestibular fistula, doing the first operation perfectly would lead to a better sphincter function than correcting any failed first operation with a second operation⁽¹¹⁾.

At the Division of Pediatric Surgery, Department of Surgery, Faculty of Medicine Siriraj Hospital, Mahidol University, patients with vestibular fistula are operated on one of the two pathways, 1) primary anorectoplasty from birth or 2) staged operations beginning with colostomy. The decision about which pathway should be done is made on a case-by-case basis depending on the patient's status and surgeon's experience. The purpose of the present study was to compare the complication rates and functional outcomes of the surgical treatments for vestibular fistula between multistage operations beginning with colostomy prior to anorectoplasty (C) and anorectoplasty without colostomy (NC). The functional outcomes following the operations were evaluated by the Krickenbeck scoring system⁽¹²⁾.

Materials and Methods

The present study was approved by the Siriraj Institutional Review Board (COA No. Si 324/2019). A retrospective review was performed of the medical records of all patients with anorectal malformations with vestibular fistula that underwent surgical management at the Division of Pediatric Surgery, Department of Surgery, Faculty of Medicine Siriraj Hospital, Mahidol University, Thailand, between January 2006 and December 2018.

The exclusion criteria were 1) exstrophy bladder, 2) inadequate follow-up period, 3) major chromosomal anomaly with an incapability to survive, 4) Currarino triad with presacral tumor, anorectal malformation, or spine anomaly, or 5) incomplete medical records.

The present study used Krickenbeck's classification for anorectal malformation. At the Division of Pediatric Surgery, Department of Surgery, Faculty of Medicine Siriraj Hospital, Mahidol University, patients with vestibular fistula were operated in one of the two pathways, 1) multistage operations beginning with colostomy prior to anorectoplasty (C) or 2) primary anorectoplasty without colostomy (NC). The decision about which pathway should be done was made on a case-by-case basis depending on the patient's status and the surgeon's experience.

Data included the patients' basic demographic data, associated anomalies, location of colostomy, complications following colostomy, types of definite anorectoplasty, age at definite anorectoplasty, complications following definite anorectoplasty, treatments of complications following anorectoplasty such as colostomy or re-anoplasty, and complications following colostomy closure. All data were processed through the SPSS Statistics software and statistical analyses were carried out.

Functional outcomes following the operations were evaluated by the Krickenbeck scoring system⁽¹²⁾. The evaluation was conducted in patients older than three years of age and all operations were performed in full in at least six months' intervals. This scoring system evaluated the functional outcomes based on three modules, voluntary bowel movements, soiling, and constipation. The functional outcomes were assessed by direct face-to-face interviews as well as interviews by telephone.

Results

Forty patients with vestibular fistula were treated at the Division of Pediatric Surgery, Department of Surgery, Faculty of Medicine Siriraj Hospital, Mahidol University, Thailand, between January 2006 and December 2018. Of those, 18 patients underwent multistage operations beginning with colostomy prior to anorectoplasty (C), while 22 patients underwent primary anorectoplasty without colostomy (NC).

The demographic data of all the patients are shown in Table 1. The average gestational age was 37.34 weeks. The average gestational ages in both groups were the same at 37.33 and 37.35 weeks.

Table 1. Demographic data of all patients

Data	With colostomy (C) (n=18)	Without colostomy (NC) (n=22)	Total (n=40)	p-value
Average GA (weeks); mean±SD	37.33±2.49	37.35±1.59	37.34±1.94	0.473
Associated anomalies; n (%)	17 (94.4)	11 (50.0)	28 (70.0)	0.002
Cardiac	9 (52.9)	8 (72.7)	17 (60.7)	0.255
GI	4 (23.5)	1 (9.1)	5 (17.9)	0.330
Spine	8 (47.1)	1 (9.1)	9 (32.1)	0.036
GU	9 (52.9)	1 (9.1)	10 (35.7)	0.018
Ventilator support; n (%)	2 (11.1)	0 (0.0)	2 (6.3)	0.044
Age at definite surgery (days); median (P ₂₅ , P ₇₅)	290 (181, 420)	2 (2, 3)	43 (2, 257)	0.021
LOS for definite surgery (days); median (P ₂₅ , P ₇₅)	12 (10, 24)	12 (10, 22)	12 (10, 22.5)	0.328

SD=standard deviation; GA=gestational age; GI=gastrointestinal; GU=genitourinary; LOS=length of stay

Table 2. Types of definitive anorectoplasty surgery

Type of operations	With colostomy (C) (n=18); n (%)	Without colostomy (NC) (n=22); n (%)	Total (n=40); n (%)
PSARP	5 (27.8)	0 (0.0)	5 (12.5)
Anal transposition	11 (61.1)	20 (90.9)	31 (77.5)
Anoplasty	1 (5.6)	2 (9.1)	3 (7.5)
Explor lap with rectal pull-through for both vagina and anus repair	1 (5.6)	0 (0.0)	1 (2.5)

PSARP=posterior sagittal anorectoplasty

The major difference between these two groups, the C group and the NC group, was the prevalence of associated anomalies. Multistage operations beginning with colostomy (C) had associated anomalies in about 94.4% of the cases, while primary anorectoplasty without colostomy (NC) had a rate of anomalies of about 50%. This statistically significant difference could affect the surgeon's decision to choose whether colostomy should be done. For the four patients who had gastrointestinal anomalies, three patients with esophageal atresia, and the patient with duodenal atresia, the decision to perform colostomy was justified. Three patients with esophageal atresia received thoracotomy to correct esophageal atresia, and then colostomy was created to reduce the operative time. Colostomy was also done for vestibular fistula associated with spinal and genitourinary anomalies. Although these associated anomalies could affect the decision whether to do colostomy. These anomalies themselves could not affect the incidence of wound infection from anorectoplasty. The median lengths of admission for the definitive surgery of multistage operations beginning with colostomy and for anorectoplasty without colostomy were also the same at 12 days.

In the 18 patients that underwent colostomy, 14 sigmoid colostomy operations were performed, and

one transverse colostomy was done. An unknown site of colostomy was noted in three patients referred to Siriraj Hospital from the other hospitals. The median age of colostomy formation was two days (P₂₅=1.5, P₇₅=3).

Three of the 18 patients (16.7%) that underwent colostomy had complications. Two patients developed a prolapsed colostomy, with colostomy revision done in one of them. Wound infection at the colostomy site was found in one patient. The median length of stay for a colostomy operation was 16 days (P₂₅=11, P₇₅=21).

The types of definitive anorectoplasty surgery are listed in Table 2. Anal transposition, performed in 31 patients, was the most common operation to correct the vestibular fistula. Among the 18 patients who underwent multistage operations beginning with colostomy, 11 (61.1%) patients had anal transposition performed and five (27.8%) patients underwent PSARP. Twenty (90.9%) patients with primary anorectoplasty without colostomy, representing all except two of the anoplasty cases, had anal transposition performed. One patient who had vestibular fistula associated with vaginal atresia, was performed colostomy, followed by exploratory laparotomy with rectal pull-through for both the vagina and anus repair.

Table 3. Early complications following definitive anorectoplasty

	With colostomy (C) (n=18); n (%)	Without colostomy (NC) (n=22); n (%)	Total (n=40); n (%)	p-value
Early complications	4 (22.2)	8 (36.4)	12 (30.0)	0.332
Wound infection	1 (5.6)	1 (4.5)	2 (5.0)	0.884
Anal stricture	1 (5.6)	4 (18.2)	5 (12.5)	0.230
Recurrence of fistula	0 (0.0)	2 (9.1)	2 (5.0)	0.189
Prolapsed anal mucosa	2 (11.1)	1 (4.5)	3 (7.5)	0.093
Bleeding	0 (0.0)	1 (4.5)	1 (2.5)	0.360
Re-operation	3 (16.7)	7 (31.8)	10 (25.0)	0.135

Table 4. Details of the re-operations

Complications requiring re-operation	With colostomy (C)	Without colostomy (NC)
Anal stricture	0/1 - No re-operation	3/4 - Anoplasty
Recurrence of fistula	0/0 - No re-operation	2/2 - Colostomy then PSARP
Prolapsed anal mucosa	2/2 - Excision of the mucosal prolapse	1/1 - Anoplasty
Severe constipation	1/1 - PSARP	1/1 - Anoplasty

PSARP=posterior sagittal anorectoplasty

Table 5. Functional outcomes following the operations evaluated by Krickenbeck scoring system

	With colostomy (C) (n=9)	Without colostomy (NC) (n=14)	Total (n=23)	p-value
Follow-up period; median (P ₂₅ , P ₇₅)			1,336 days (532, 2,407)	
Age of evaluation; median (P ₂₅ , P ₇₅)			1,818 days (956, 2,919)	
Voluntary bowel movement; n (%)	9 (100)	14 (100)	23(100)	
Feeling of urge	8 (88.9)	14 (100)	22 (95.7)	0.202
Capacity to verbalize	8 (88.9)	14 (100)	22 (95.7)	0.742
Hold the bowel movement	8 (88.9)	13 (92.9)	21 (91.3)	0.202
Soiling; n (%)	3 (33.3)	5 (35.7)	8 (34.7)	0.660
Grade 1: occasionally (once or twice per week)	1	2	3	
Grade 2: every day, no social problem	2	3	5	
Grade 3: constant, social problem	-	-	-	
Constipation; n (%)	6 (66.7)	9 (64.3)	15 (65.2)	0.598
Grade 1: manageable by changes in diet	-	1	1	
Grade 2: requires laxatives	5	8	13	
Grade 3: resistant to diet and laxative	1	-	1	

Early complications following definitive anorectoplasty

Early complications following definitive anorectoplasty are listed in Table 3. Multistage operations beginning with colostomy had a lower incidence of complications than the primary anorectoplasty at 22.2% versus 36.4%. The most common complication following multistage operations beginning with colostomy was a prolapsed anal mucosa in two patients (11.1%), requiring re-operation to excise the prolapsed anal mucosa. This re-operation was a minor operation. However, the complications following the primary anorectoplasty

were more serious complications, such as recurrent vestibular fistula in two patients (9.1%) and anal stricture in four patients (18.2%). These two recurrent vestibular fistulae required multistage repairs, which colostomies were first done and PSARP was performed later. Three of four patients with primary anorectoplasty required re-anoplasty due to severe anal stricture. Both groups had one case each of intractable constipation requiring another operation to correct this long-term complication. Details of the re-operations are given in Table 4.

The functional outcomes following the operations were evaluated by the Krickenbeck scoring system⁽¹²⁾

and the results are reported in Table 5. The evaluation was conducted in patients older than three years of age and all operations were performed in full at least in six months' intervals. This scoring system evaluated the functional outcomes based on three modules, voluntary bowel movements, soiling, and constipation. The functional outcomes were assessed by direct face-to-face interviews as well as by interviews by telephone. In the present study, 23 patients were evaluated by their Krickenbeck score at a median age of 1,818 days.

Twenty-three patients were evaluated for functional outcomes using the Krickenbeck score at a median age of 1,818 days, or 4.98 years old. There were no significant differences in voluntary bowel movement, frequency of soiling, and constipation between patients who underwent multistage operations beginning with colostomy and those who underwent primary anorectoplasty. The median follow-up period was 1,336 days or 3.66 years.

Discussion

Vestibular fistula is the most common type of anorectal malformation in female infants. Although this defect has good prognosis in functional outcomes, the surgical treatments are still debated about which surgical technique is the best choice for the management of vestibular fistula. The algorithm to treat this type of anorectal malformation has changed over the last three decades. The Wingspread classification of anorectal malformations classifies vestibular fistula into two types⁽¹³⁾ anovestibular fistula, which is a low type of anorectal malformation, and rectovestibular fistula, which is an intermediate type of anorectal malformation. In anovestibular fistula, anoplasty without prior colostomy can be done from birth. The most preferred operation is anal transposition, as derived by Potts⁽¹⁴⁾. This single stage repair has benefits in terms of reducing the number of operations needed and avoids complications from performing colostomy. In rectovestibular fistula, multistage operations are performed. In this rectovestibular fistula, difficulty can arise from the long common wall existing between the vagina and the rectum, and because two walls must be created out of one. The dissection continues cephalad until the rectal and vaginal walls are fully separated. If the rectum and the vagina were not completely separated, a tense anal anastomosis predisposes the wound to dehiscence, retraction, and stricture⁽¹¹⁾. Therefore, in rectovestibular fistula, multistage operations are performed, where the first operation is colostomy,

while the second operation is anorectoplasty followed by a third operation for closure of the colostomy.

A revolution in the operation for anorectal malformation was developed by deVries and Peña⁽²⁾, first published details of "posterior sagittal anorectoplasty" in 1980⁽²⁾ and emphasized this again in 1982⁽³⁾. Peña developed a new classification and first coined the term "rectovestibular fistula". Anovestibular fistula is no longer used. To clarify the classification of anorectal malformation, the International Conference for the Development of Standards for the Treatment of Anorectal Malformations at Krickenbeck developed a new classification called "Krickenbeck classification"⁽¹⁾. In this classification, there is only "vestibular fistula". Also in the present study, the authors used this classification. Because patients with vestibular fistula can be operated on with either of these two options, primary anorectoplasty since birth or staged operations beginning with colostomy, the present study wanted to compare these two options of the treatment for vestibular fistula.

Primary anorectoplasty or anal transposition, minimal PSARP, ASARP, other anoplasty, which offers benefits in terms of a reduction in the number of operations needed and avoids complications from colostomy, has been reported to have good safety and functional outcomes⁽⁴⁻¹⁰⁾. On the other hand, staged operations beginning with colostomy⁽²⁻⁴⁾ still have advantages such as 1) adequate decompression, 2) adequate bowel preparation before definite anorectoplasty, and 3) decreased fecal contamination at the surgical site. Colostomy can prevent fecal contamination⁽⁴⁾. If the perineal wound was infected, dehiscence of anal anastomosis or the perineal body and recurrent vestibular fistula might occur.

In the present study, the significant difference between these two groups, the C group and the NC group, was the prevalence of associated anomalies. Multistage operations beginning with colostomy had associated anomalies in about 94.4% of cases, while primary anorectoplasty without colostomy had a rate of about 50%. This statistically significant difference could affect the surgeon's decision to choose whether colostomy should be first done. In vestibular fistula associated with spinal and genitourinary anomalies, colostomy was likely to be performed. Although, these associated anomalies could affect the surgeon's decision to do colostomy, these anomalies would not affect the incidence of wound infection following anorectoplasty.

Multistage operations beginning with colostomy

had a lower incidence of complications than primary anorectoplasty at 22.2% versus 36.4%. The most common complication following multistage operations beginning with colostomy was a prolapse of the anal mucosa, which all required re-operation to excise the prolapsed anal mucosa. This re-operation was a minor operation. However, the complications following primary anorectoplasty were serious complications, such as 9.1% recurrent vestibular fistula and 18.2% anal stricture. These two recurrent vestibular fistula required multistage repairs. Colostomy was first done and then PSARP was performed later. There were few recurrences of fistula, but this was one of the most serious complications following operations for vestibular fistula. In the follow-up period, the incidence of post-operative anal stricture in primary anorectoplasty repair was greater than in multistage operations beginning with colostomy. Three out of four cases required re-anoplasty due to severe stricture.

Vestibular fistula repair with prior colostomy done first had a lower incidence of complications than the primary vestibular fistula repair. Following complications of anorectoplasty, the anal sphincter would have a poor function from intense fibrosis. Regarding this vestibular fistula, doing the first operation perfectly would lead to a better sphincter function than correcting any failed first operation with a second operation⁽¹¹⁾.

Functional outcomes following the operations were evaluated by the Krickenbeck scoring system⁽¹²⁾. Surprisingly, there were no significant differences in voluntary bowel movement, frequency of soiling, and constipation between the multistage operations beginning with colostomy and the primary anorectoplasty without colostomy.

However, the present study has limitations to note due to the small sample size and selection bias. The patients with severe associated anomalies were likely to have undergone colostomy prior to anorectoplasty.

Conclusion

Vestibular fistula repair with prior colostomy done first had a lower incidence of complications than the primary vestibular fistula repair. The serious complications following primary anorectoplasty were recurrent vestibular fistula and anal stricture. These recurrent vestibular fistulae required multistage repairs. Colostomies were first done and then PSARP was performed later. The severe anal strictures required re-anoplasty.

Surprisingly, the functional outcomes using

the Krickenbeck score revealed no significant differences in voluntary bowel movement, frequency of soiling, and constipation between the multistage operations beginning with colostomy and the primary anorectoplasty without colostomy.

However, the present study had limitations due to the small sample size and the selection bias.

What is already known on this topic?

Vestibular fistula is one of the most common types of anorectal malformations in female infants. Colostomy prior to definitive anorectoplasty is still controversial.

What this study adds?

Vestibular fistula repair with prior colostomy done first had a lower incidence of complications than the primary vestibular fistula repair.

The serious complications following primary anorectoplasty were recurrent vestibular fistula and anal stricture. These recurrent vestibular fistulae required multistage repairs. Colostomies were first done and then PSARP was performed later. The severe anal strictures required re-anoplasty.

The functional outcomes using the Krickenbeck score revealed no significant differences between the two techniques.

Conflicts of interest

The authors declare no conflict of interest.

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