

# Functional Assessment of the Patients with Perineal and Vestibular Fistula Treated by Anterior Sagittal Anorectoplasty

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## Abstract

**Background:** Anterior sagittal anorectoplasty (ASARP) has been a standardised operative treatment for anorectal malformation (ARM). This retrospective study was undertaken to evaluate patients with perineal fistula (PF) and vestibular fistula (VF) treated by ASARP in our institution. **Patients and Methods:** Twenty patients (PF,  $n = 14$ ; male,  $n = 8$  and female,  $n = 6$  and VF,  $n = 6$ ) were evaluated. Eighteen patients underwent primary ASARP without protective colostomy. Two patients underwent colostomy because of intestinal atresia and suspected of other type ARM. The age range of operation was from 4 months to 5.0 years. Sixteen patients (PF,  $n = 13$ ; male,  $n = 7$  and female  $n = 6$  and VF,  $n = 3$ ) over 3 years of age were evaluated according to the Krickbeck classification. **Results:** Operative complications occurred in one patient. Minor wound dehiscence occurred in six patients. Mucosal prolapse occurred in two patients. According to the Krickbeck classification, amongst male patients with PF, all patients had voluntary bowel movements (VBMs) and two patients had Grade 1 soiling, while four patients had Grade 2 constipation. Amongst female patients with PF, all patients had VBM and no soiling, one patient had Grade 1 and two patients had Grade 2 constipation. In patients with VF, one patient was continent with Grade 1 soiling. One patient had Grade 2 and two patients had Grade 3 constipation. **Conclusions:** ASARP without colostomy carried a risk of wound dehiscence. The ASARP technique provided normal or moderate outcomes for VBM and soiling. However, in about half of patients, defecation management with laxative therapy was required to achieve a normal condition.

**Keywords:** Anorectal malformation, anterior sagittal anorectoplasty perineal fistula, vestibular fistula

## INTRODUCTION

Perineal fistula (PF) and vestibular fistula (VF) are common forms of anorectal malformations (ARMs).<sup>[1,2]</sup> The rectum in PF and VF is anterior and outside the voluntary external sphincter complex. The fistula is outside of the hymenal ring and separate from the urethra and vagina. Anterior sagittal anorectoplasty (ASARP) was introduced by Okada *et al.*<sup>[3]</sup> based on the principle of posterior sagittal anorectoplasty (PSARP).<sup>[4]</sup> The sphincter muscle is cut from the anterior aspect longitudinally through a median perineal skin incision, and the rectum is covered through the central portion of the sphincter muscle with the patient in the lithotomy position.<sup>[3]</sup> The position of the external sphincter complex in relation to the fistula is delineated by electrical stimulation. In contrast to full PSARP, only the anterior aspect of the external sphincter is divided. Dissection of the rectum from the adjacent

structures is performed to achieve tension-free positioning in the centre of the sphincter complex. This operation allows for the placement and anchoring of the mobilised rectum within the muscle complex, and the sphincter muscle and the perineal body are reconstituted. ASARP has become the standard operative technique for PF and VF.

Several studies have reported on the bowel outcomes after ASARP with a focus on patients with PF and VF.<sup>[5-9]</sup> Furthermore, although several techniques have been practised by many centres in the treatment of ARM, there is still variation in the follow-up criteria used. Long-term outcomes of ARM

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have been reported using several different scoring methods. The clinical scoring criteria for outcome assessment have been described by Kelly,<sup>[10]</sup> Templeton and Ditesheim,<sup>[11]</sup> Holschneider<sup>[12]</sup> and Rintala *et al.*<sup>[13]</sup> In an attempt to resolve this issue, the category documenting functional outcome criteria were published by the Krickenbeck group.<sup>[14]</sup> The resulting classification, devised after an international workshop attended by experts in the field, incorporated criteria from the Wingspread and Pena classification.<sup>[4]</sup> However, few studies have examined the post-operative outcomes for PF and VF treated through other procedures using the Krickenbeck classification.<sup>[6,9,15-17]</sup>

The present retrospective study aimed to evaluate the results and long-term follow-up of ASARP in the treatment of PF and VF as defined by the Krickenbeck classification and other assessments in our institution.

## PATIENTS AND METHODS

Twenty patients (male  $n = 8$ ; female  $n = 12$ ) with ARMs were treated in our institution from January 2000 to August 2017. Amongst these, 14 patients (male,  $n = 8$  and female,  $n = 6$ ) were diagnosed with PF (including 2 male patients with an incompletely covered anus) and 6 patients were diagnosed as VF. Eighteen patients underwent primary ASARP without protective colostomy. Two patients received a colostomy due to intestinal atresia and the suspicion of high- or intermediate-type ARMs. Two patients who underwent surgery at over 24 months of age had suffered from chronic constipation at birth, and the diagnosis of ARM was delayed. Preoperatively, Hegar dilatation and a glycerine enema were used for the defecation management.

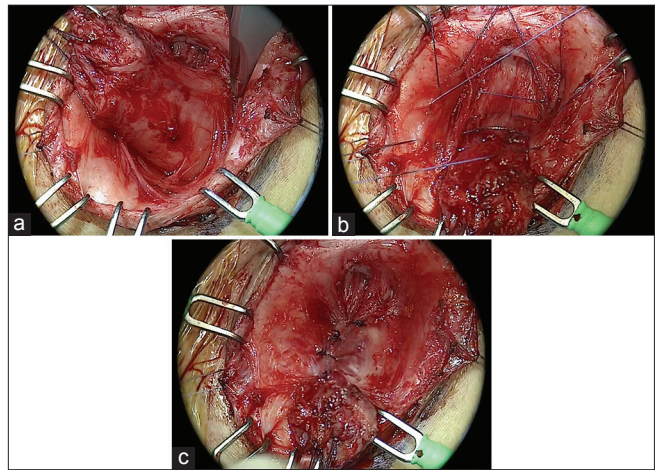
### Operative technique

The proposed anal sites were found using an electric neurostimulator. The sphincter muscle cut from the anterior aspect through a median perineal skin incision. Lone Star® Retractor Ring (Cooper surgical, INC., Trumbull, CT, USA) was used for dilation for the skin incision (please check this). The rectum was placed within the muscle complex with the help of a muscle stimulator [Figure 1a]. The sphincter muscle complex was sutured and wrapped with the anal canal using 4-0 VICRYL® (ETHICON, INC., Somerville, NJ, USA) [Figure 1b and c]. Dissection of the rectum from the adjacent structures was performed to achieve tension-free positioning in the centre of the sphincter complex. The anal canal and skin were sutured using 4-0 VICRYL®, and the perineal skin incision was sutured using 4-0 ETHILON® (ETHICON, INC.).

Oral intake was withheld for 24 h before surgery, and oral feeding was started based on the surgeons' decision. Depending on the condition of the perineal wound, the urinary catheter was removed after the 7<sup>th</sup> post-operative day.

### Post-operative period

Local wound care consisted of washing the perineum with water after defecation. With regard to wound care, the wound



**Figure 1:** (a) The figure shows that the rectum and the sphincter muscle divide anterior aspect which stimulate with the help of a muscle stimulator. (b) The figure shows that external sphincter muscle is sutured between the anal canal. (c) The figure shows that the anal canal is wrapped by external sphincter muscle

was kept clean by water and the application of an antibiotic ointment 3–4 times a day. Parenteral broad-spectrum antibiotics were administered from the 5<sup>th</sup> to 7<sup>th</sup> day. Before discharge on the 14<sup>th</sup> post-operative day, the methods of performing anal dilatation using a Hegar dilator and/or the parent's finger were routinely explained. Glycerine enema and laxatives, such as sodium picosulfate hydrate and magnesium oxide, were commonly administered over the long term. Anal dilatation, enema and laxatives were adjusted according to the post-operative bowel function.

Amongst the 20 patients, the 16 with ARMs who were older than 3 years of age were retrospectively evaluated the functional outcomes according to three assessment methods: Kelly's clinical score, the bowel functional score (BFS) and the Krickenbeck classification.

### Functional outcomes

#### Kelly's clinical score

This system of scoring awards points for three basic parameters: continence, staining and sphincter. An overall score of 5–6 is considered good, 3–4 fair and 0–2 poor. It is by far the simplest of all of the scoring systems and the easiest to apply, even in the office setting [Table 1].<sup>[10]</sup>

#### Bowel functional score

The BFS is a seven-item qualitative scoring system devised by Rintala *et al.*<sup>[13]</sup> for the assessment of the bowel function in benign anorectal disorders [Table 2].<sup>[13]</sup>

#### Krickenbeck classification

The Krickenbeck classification is composed of three elements concerning the post-operative results: voluntary bowel movements (VBMs), soiling and constipation. The Krickenbeck classifications were determined to assess the patients' clinical status [Table 3].<sup>[14]</sup>

**Table 1: Kelly's clinical score**

	Score
Continence	
Normal, no soiling	2
Occasional accidents, faeces/flatus escape	1
No control, frequent accident	0
Staining	
Always clean	2
Occasional staining	1
Always stained	0
Sphincter	
Strong and effective squeeze	2
Weak and partial squeeze	1
No contraction	0

**Table 2: Bowel functional score**

	Score
Ability to hold back defecation	
Always	3
Problem <1/week	2
No voluntary control	1
Feels, reports urge to defecation	
Always	3
Most of time	2
Uncertain	1
Absent	0
Frequency of defecation	
Every other day-2/day	2
More often	1
Less often	0
Soiling	
Never	3
Staining, <1/week, no underwear charge	2
Frequent, often underwear charge	1
Daily, protective aids	0
Accident	
Never	3
<1/week	2
Weekly, often protective aids	1
Daily, protective aids day and night	0
Constipation	
Nil	3
Manage with diet	2
With laxatives	1
With enemas	0
Social problems	
Nil	3
Sometimes	2
Deterioration in social life	1
Severe social/psychological problems	0

## RESULTS

ASARP was successfully performed in all patients [Table 4]. The mean age of the patients at the operation was 13.3 months (median: 10.5 months, 25<sup>th</sup>–75<sup>th</sup> percentile:

8.25–12 months). The mean operative time was 128 min (median: 130 min, 25<sup>th</sup>–75<sup>th</sup> percentile: 111–145 min). The intra-operative blood loss was under 20 ml in each patient. No blood transfusion was required. One patient (5.0%) experienced an operative complication (urethral injury) that was successfully repaired. In the post-operative period, minor wound dehiscence occurred in 6 (33.3%) patients who underwent primary ASARP without protective colostomy.

Amongst all patients, mucosal prolapse occurred in 2 (10.0%) who required mucosal plications with the Gant-Miwa procedure. There were no recurrences of fistula, anal stenosis or anterior displacement of the neorectum. The mean period of post-operative anal dilation was 6.2 months (median: 5.5 months, 25<sup>th</sup>–75<sup>th</sup> percentile: 2.3–8.0 months). The mean duration of post-operative follow-up was 48.2 months (median: 45.0 months, 25<sup>th</sup>–75<sup>th</sup> percentile: 18.5–65.0 months).

The mean age of the 16 patients who were evaluated for their functional outcomes was 56.5 months (median: 52.0 months, 25<sup>th</sup>–75<sup>th</sup> percentile: 31.8–70.5 months). Thirteen patients (male  $n = 7$  and female  $n = 6$ ) were diagnosed with PF and three patients were diagnosed with VF.

Amongst the total patients, the mean overall Kelly's clinical score was 5.5 (median: 6, 25<sup>th</sup>–75<sup>th</sup> percentile: 5–6). Fourteen patients (87.5%; PF [ $n = 13$ ] and VF [ $n = 1$ ]) showed a good outcome and 2 (12.5% VF [ $n = 2$ ]) showed a fair outcome. Eleven patients (68.8% PF; male  $n = 4$  and female  $n = 7$ ) had an optimum Kelly's clinical score.

Amongst the total patients, the mean overall BFS was 18.3 (median: 18, 25<sup>th</sup>–75<sup>th</sup> percentile: 18–20). Fourteen patients (87.5%; PF [ $n = 13$ ] and VF [ $n = 1$ ]) had a BFS in the normal range (BFS  $\geq 17$ ) consistent with a good outcome and two patients (12.5% VF [ $n = 2$ ]) had a moderate outcome (BFS: 14–16). Of the two patients with a moderate score, constipation reduced the BFS by 0 points and was associated with minor soiling. Five patients (31.3% PF; male  $n = 2$  and female  $n = 3$ ) had an optimum BFS.

According to the Krickbein classification, amongst male patients with PF, all patients had VBM and two patients had Grade 1 soiling, while four patients had Grade 2 constipation. Amongst female patients with PF, all patients had VBM and no soiling and one patient had Grade 1, and two patients had Grade 2 constipation. Amongst patients with VF, one patient was continent with Grade 1 soiling, one patient had Grade 2 constipation and two patients had Grade 3 constipation. All patients with Grade 2 constipation were administered magnesium oxide as a laxative. Grade 3 constipation in two patients was controlled with a glycerine enema twice a week.

## DISCUSSION

PF and VF are the most common types of ARMs. Many procedures have been described for the treatment of these anomalies, ranging from the cutback operation<sup>[18]</sup> to Pott's technique,<sup>[19]</sup> and more recently, PSARP<sup>[4]</sup> and ASARP.<sup>[3]</sup> With

the ASARP technique, the incision in the anterior approach was much smaller than with the posterior approach of PSARP. An anterior approach requires the division of only the anterior fibres of the external sphincter, and separation of the urethra or vagina and rectum takes place under direct vision, while with the posterior approach, dissection is performed blindly.<sup>[20]</sup>

Wound infection is a common early complication with the ASARP technique. Okada *et al.*<sup>[3]</sup> reported that prolonged fasting for 2 weeks was necessary for proper wound healing. Kulshreshtha *et al.*<sup>[20]</sup> also reported that VF patients undergoing single-stage repair through ASARP with the oral intake started

early through a liquid diet from the 2<sup>nd</sup> day and continued for 8–10 days showed a higher incidence of complications than in patients with prolonged fasting and TPN. Gupta *et al.*<sup>[16]</sup> reported the findings of their randomised controlled trial comparing a primary definitive procedure with a conventional three-stage procedure for the management of PF and VF treated with PSARP, mini PSARP or ASARP in women. Superficial and deep wound dehiscence occurred in 13 of 33 (39.4%) children with the primary definitive procedure and in 6 of 31 (18.2%) children with the three-stage procedure, a significant difference ( $P = 0.038$ ). A primary definitive procedure involves a single operation and carries a risk of wound dehiscence.<sup>[17]</sup> However, Aziz *et al.*<sup>[21]</sup> reported wound disruption in 13% (3/23) of patients undergoing ASARP despite the initiation of oral feeding on the 4<sup>th</sup> post-operative day. Wakhlu *et al.* reported that the overall incidence of complications in their patients was 5% that seen in other procedures used for VF. Oral intake was allowed upon recovery from anaesthesia.<sup>[22]</sup> They also reported that traumatic dissection, haematoma or inadequate mobilisation of the rectum with anoplasty under tension caused wound disruption.<sup>[16]</sup> In our series, the rate of minor wound dehiscence was 30.0%. As the pre-operative defecation management was not troubled, the operative age was adjusted to between 8 and 12 months so that the patients could undergo the operation safely. As the operative age was much older than in other reports,<sup>[8,21,22]</sup> the area of tissue

**Table 3: Krickenbeck classification**

Voluntary bowel movements	Yes/no
Feeling of urge, capacity to verbalize, hold the bowel movement	
Soiling	Yes/no
Grade 1	Occasionally (once/twice per week)
Grade 2	Every day, no social problem
Grade 3	Constant, social problem
Constipation	Yes/no
Grade 1	Manageable by change in diet
Grade 2	Require laxative
Grade 3	Resistant to laxatives and diet

**Table 4: Patient characteristics**

Patients	Gender	Type of fistula	Age at operation (month)	Received colostomy	Follow up (month)	Kelly's clinical score	BFS	Krickenbeck classification	Associated anomalies
1	Male	PF	11		62	2/1/2	3/3/2/2/3/3/3	Yes/1/no	
2	Male	PF	60		15	2/2/2	3/3/2/3/3/3/3	Yes/no/no	
3	Female	PF	10		52	2/2/2	3/3/2/3/3/3/3	Yes/no/no	
4	Female	PF	10		66	2/2/2	3/3/2/3/3/3/3	Yes/no/1	
5	Female	VF	9	+	156	1/1/2	2/3/2/2/2/0/3	No/1/3	CIA, spinal AVM, neurogenic bladder
6	Female	VF	12		54	2/2/2	3/3/2/3/3/1/3	Yes/no/2	
7	Male	PF (CAI)	12	+	108	2/2/1	3/3/2/3/3/3/3	Yes/no/no	Ventricular septal defect
8	Female	PF	6		42	2/2/2	3/3/2/3/3/3/3	Yes/no/no	
9	Female	PF	7		31	2/2/2	3/3/2/3/3/1/3	Yes/no/2	
10	Female	PF	11		72	2/2/2	3/3/2/3/3/3/3	Yes/no/no	
11	Male	PF	12		72	2/2/2	3/3/2/3/3/1/3	Yes/no/2	
12	Female	VF	8		52	1/1/1	3/3/2/2/3/0/3	Yes/1/3	
13	Male	PF (CAI)	12		48	2/1/2	3/3/2/2/3/1/3	Yes/1/2	
14	Male	PF	24		26	2/2/2	3/3/2/3/3/1/3	Yes/no/2	
15	Male	PF	12		34	2/2/2	3/3/2/3/3/1/3	Yes/no/2	
16	Female	PF	22		14	2/2/2	3/3/2/3/3/1/3	Yes/no/2	
17	Female	VF	9		23				Double vagina
18	Female	VF	26		17				Perineal lipoma
19	Male	PF	20		14				
20	Female	VF	9		5				Perineal lipoma

Kelly's clinical score (continence/staining/sphincter). BFS (ability to back hold/feel, report urge to defecation/frequency of defecation/soiling/accident/constipation/social problems). Krickenbeck classification (voluntary bowel movement/soiling/constipation). PF: Perineal fistula, VF: Vestibular fistula, CAI: Covered anus incomplete, CIA: Congenital intestinal atresia, AVM: Arteriovenous malformation, BFS: Bowel functional score, +: Received colostomy

dissected was consequently much larger, creating greater risk in patients if infection occurred.<sup>[9,23]</sup>

The ability to pass a bowel movement voluntarily implies the presence of the adequate innervation and function of the pelvic floor, rectum and internal and external anal sphincter.<sup>[17]</sup> Soiling occurs because of defects in the sphincter mechanism or overflow from chronic constipation.<sup>[16]</sup> In our series, most patients were in a good condition with regard to soiling. ASARP allows for the placement and anchoring of the mobilised rectum within the muscle complex covered through the central portion of the sphincter muscle. With this technique, the quality of the sphincter muscle was good. Furthermore, management with magnesium oxide to soften the stool did not induce overflow soiling.

All patients with Grade 2 or 3 constipation according to the Krickenbeck classification were administered magnesium oxide. Constipation can occur due to the failure to evacuate the stool adequately. It is often seen in association with PF and VF in other reports.<sup>[17,21]</sup> However, most patients administered magnesium oxide in the present study were in a good condition with regard to defecation management. As laxative therapy guided the decision about the grade of constipation, Krickenbeck classification is difficult to reveal other outcomes such as frequency of defecation and social problems.<sup>[14,15]</sup> Demirogullari *et al.*<sup>[15]</sup> reported that, after the appropriate treatment, the grade of soiling significantly decreased, but improvement in the grade of constipation was not very apparent.

The long-term outcomes of ARM have been reported using several different scoring methods. However, few studies have examined the post-operative outcomes for PF and VF treated with other procedures using the Krickenbeck classification.<sup>[6,9,15-17]</sup> Hassett *et al.*<sup>[17]</sup> evaluated patients with PSARP according to the Krickenbeck classification. In 19 children with PF, 90% had VBM with no soiling and 21% had Grade 2 constipation. One child in this group underwent a MACE procedure for resistant constipation. In 14 children with VF, 57% had VBM with no soiling and Grade 3 constipation was noted in 28%. One child had Grade 1 soiling and one child had Grade 2 soiling. In this group, two children had a MACE stoma fashioned because of intractable constipation.<sup>[17]</sup> These functional outcomes for PF and VF treated with PSARP were comparable between that study and the present study. Wang *et al.*<sup>[9]</sup> evaluated 26 patients with VF treated with single-stage modified ASARP, in which a potential tunnel was created through the centre of the external sphincter complex under endoscopic guidance according to the Krickenbeck classification. None of the patients was classified as having a poor outcome. Three patients (11.5%) had soiling once or twice per week. Four patients (15.4%) had constipation that was managed by changes in the diet. In contrast to PSARP, precise siting of the neorectum within the sphincter complex and adequate separation of rectum up to the level of the pelvic peritoneal

reflection from vagina was able to be performed with direct visualisation using ASARP.<sup>[9]</sup>

An important feature of the Krickenbeck classification is the precise description of the outcome criteria for continence parameters. These criteria are based on a simple grading classification with variables for constipation and soiling.<sup>[14,17]</sup>

In our study, most patients had normal-range findings, which were consistent with a good outcome according to Kelly's clinical score and BFS. However, defecation management through laxative therapy was evaluated severe grading of constipation according to BFS and the Krickenbeck classification.

Several limitations associated with the present study warrant mention. This study was retrospective, and a relatively small number of cases were included. Most of the functional evaluation was performed based on the clinical symptoms and physical examination findings.

## CONCLUSIONS

ASARP without colostomy carried a risk of wound dehiscence. According to the functional outcomes, our results indicated that the ASARP technique provided normal or moderate outcomes for VBM and soiling. However, in about half of patients, defecation management with laxative therapy was required to achieve a normal condition. Long-term follow-up is necessary to confirm our findings.

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N. H. drafted the manuscript. Y. T. and M. Y. critically reviewed the manuscript and supervised the whole study process. K. A., S. F. and S. I. underwent operation. N. S., N. H., M. Y., D. M., S. S. and T. S gave conceptual advice. All authors read and approved the final manuscript.

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## Conflicts of interest

There are no conflicts of interest.

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