

Outcomes of modified posterior sagittal anorectoplasty with internal sphincter preservation in the treatment of intermediate anorectal malformations in children

Tran Anh Quynh^{1*}, Le Quang Du¹, Pham Duy Hien¹, Nguyen Thi Minh Huyen¹, Tran Thi Thuy¹

ABSTRACT

Objective: This study aimed to evaluate the long-term outcomes of modified posterior sagittal anorectoplasty (PSARP) with internal sphincter preservation for the treatment of intermediate anorectal malformations (ARM) in children. **Methods:** A total of 41 medical records of pediatric patients who were diagnosed with intermediate ARMs according to the Wingspread classification at the National Children's Hospital from February 2014 to December 2015 were reviewed. The collected data included general information, intraoperative and postoperative outcomes, and early and long-term complications. **Results:** The study showed that 28 (68.3%) achieved good bowel function, 11 (26.8%) had moderate bowel function, and only two (4.8%) had poor bowel function. Constipation was observed in 6 patients (14.6%), ranging from grade I to grade II, with all cases responding to dietary changes and/or laxatives. Most patients did not experience fecal incontinence or only had occasional incontinence (78.1%), with only 4.8% experiencing frequent incontinence. All children had normal urinary function, and all boys had morning erections. **Conclusion:** Modified PSARP with preservation of the sphincter in the treatment of intermediate-type ARM in children provides favorable long-term outcomes with good bowel function, excellent bowel control, and minimal complications.

Keywords: Anorectal malformations, Modified posterior sagittal anorectoplasty, Sphincter

¹ Viet Tiep Hospital, Hai Phong, Viet Nam

² Hai Phong University of Medicine and Pharmacy, Viet Nam

* Corresponding author

Tran Anh Quynh

Email:

tranquynh.nhp@gmail.com

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INTRODUCTION

Anorectal malformations (ARM) are among the most common congenital anomalies encountered during pediatric surgery. The history of ARM treatment spans centuries and has been extensively documented in surgical literature. In the 7th century, Aegineta introduced a simple technique to treat these anomalies [1]. Since then, numerous techniques have been developed and introduced, but no single

method has been deemed ideal because of the high incidence of postoperative fecal incontinence, which significantly affects the mental and psychological well-being of children. The primary aim of ARM surgery is not only to correct anatomical defects but also to restore critical functions, particularly defecation.

In 1982, De Vries P.A. and Peña A. described the posterior sagittal anorectoplasty (PSARP) technique, which has become the global standard for treating

ARM, including intermediate types. However, this method is limited by the division of the entire sphincter complex, which affects defecation function and the risk of infection due to the opening of the rectal pouch [2], [3].

Since 1984, improved PSARP techniques for preserving the sphincter complex have been performed at the National Children's Hospital by surgeons Nguyen Xuan Thu and Nguyen Thanh Liem in Vietnam, showing promising results [4]. Long-term outcome studies by Nguyen Thanh Liem and Bui Duc Hau in 2001 indicated excellent defecation function in children post-surgery, with very good and good outcomes in 58.3% of cases, average outcomes in 37.5%, and poor outcomes in 4.2% [5]. Furthermore, Dao Trung Hieu et al. (2013) described single-stage ARM surgeries in 101 patients at Children's Hospital 1, demonstrating feasibility, safety, reduced hospital stay, and lower treatment costs [6].

At the National Pediatric Hospital, improved PSARP preserving the sphincter complex has been performed for intermediate ARM since 1988. However, there is a lack of studies evaluating the long-term outcomes of this surgical technique. This study aimed to assess the long-term results of improved PSARP for sphincter preservation in children with intermediate ARM.

PATIENTS AND METHODS

Patients

This study included pediatric patients aged two weeks old who were diagnosed with intermediate ARM according to the Wingspread classification. Contrast-enhanced imaging of the distal bowel showed that the rectal pouch was located below the pubococcygeal line with or

without a fistula in the urethra. The patient underwent improved PSARP, preserving the sphincter, at the National Pediatric Hospital between February 2014 and December 2015. Patients with insufficient medical records or those who underwent different surgical approaches were excluded from the study. The study was approved by the Ethics Committee of the National Pediatric Hospital under decision number 842/BVNTU-HĐĐĐ.

Methods

This study was conducted between March and June 2022 at the Department of Pediatric Surgery of the National Pediatric Hospital. This descriptive case series study used a retrospective medical record analysis. A convenient sample of 41 medical records meeting the inclusion criteria was selected.

Data collection

A list of patients meeting the selection criteria was compiled from department records. Data, including general information, intraoperative and postoperative outcomes, and early- and long-term complications, were collected using a standardized medical record form. Defecation function was evaluated based on the number of bowel movements per day. In patients older than 36 months, defecation control was assessed according to the Krickenbeck classification, including the criteria for continence, constipation, and soiling [21]. The defecation function classification (adapted from Julia's Krickenbeck criteria) included the following:

Good: Voluntary defecation, no or rare soiling, mild constipation (resolved with dietary changes or laxatives).

Average: Voluntary defecation, occasional soiling grade 1 or 2, and constipation grade

1 or 2 (grade 3 constipation not responding to dietary changes or laxatives).

Poor: Involuntary defecation, soiling grade 2 or 3, and constipation of all grades.

Additionally, urinary function and penile erection capability were evaluated in male patients.

Statistical analysis

Data were entered and managed using the SPSS software (version 25.0). Descriptive statistics were applied, and chi-square tests were used for comparisons. Statistical significance was set at $P < 0.05$.

RESULTS

The mean age at the time of surgery was 77.4 ± 70.3 days, with a male-to-female ratio of 1:5.8. All patients underwent planned surgeries upon hospital admission, and none required emergency surgery. PSARP was performed in all patients with preservation of the sphincter muscles. The majority of patients (80.5%, $n=33$) underwent simple anal reconstruction, whereas 19.5% ($n=8$) underwent combined procedures, including colostomy closure and anal reconstruction.

The mean surgery duration was 73.7 ± 24.2 minutes, ranging from 40 to 180 minutes. The mean postoperative hospital stay was 3.5 ± 1.2 days, range, 2–6 days). The mean duration of postoperative antibiotic use was 5.7 ± 1.1 days, with one case extending to 10 days due to postoperative infection. The mean total hospital stay was 7.5 ± 4.3 days, ranging from 3 to 23 days. (Table 1)

Table 1. Intraoperative and Postoperative Characteristics ($n = 41$)

Variable	Mean \pm SD (Min-Max)
Surgery duration (minutes)	73.7 ± 24.2 (40–180)
Postoperative antibiotic use (days)	5.7 ± 1.1 (4–10)
Postoperative hospital stays (days)	3.5 ± 1.2 (2–6)
Total hospital stays (days)	7.5 ± 4.3 (3–23)

No intraoperative complications occurred in any of the patients. Postoperative complications were noted in two cases (4.8%): one case of wound bleeding managed with compression and hemostatic medication, and one case of wound infection on postoperative day 3, which was managed with intensive wound care and resolved before discharge.

Regarding bowel function, 24 patients (58.5%) had 1-2 bowel movements per day, 11 (26.8%) had 3-4 bowel movements per day, and 6 (14.6%) experienced constipation. Bowel movement frequency across age groups and malformation types. The frequency of 1-2 bowel movements per day was 56.6% in patients younger than 3 months, 60.0% in patients aged 3-6 months, and 66.7% in patients older than 6 months. Constipation was absent in the 3-6 months age group. There were no statistically significant differences between the groups ($p > 0.05$). The highest frequency of to 1-2 bowel movements per day was observed in the vestibular fistula group (73.7%). No constipation was observed in the non-fistula or rectourethral fistula group. There was no significant correlation between bowel movement frequency and the type of malformation ($p > 0.05$). (Table 2).

Table 2. The relationship between Bowel Movement Frequency and Various Factors

Characteristics	Bowel Movements/Day			p
	1 – 2 n (%)	3 – 4 n (%)	Constipation n (%)	
Age	< 3 months (n = 23)	13 (56.6)	5 (21.7)	0.172
	3 – 6 months (n = 15)	9 (60.0)	6 (40.0)	
	> 6 months (n = 3)	2 (66.7)	0 (0)	
Malformation	No fistula (n = 4)	2 (50.0)	2 (50.0)	0.094
	Rectourethral (n = 6)	2 (33.3)	4 (66.7)	
	Rectovestibular (n = 19)	14 (73.7)	3 (15.8)	
	Rectovaginal (n = 12)	6 (50.0)	2 (16.7)	

Most patients (85.4%) did not experience constipation. Grade 1 constipation accounted for 4.9% and grade 2 constipation accounted for 9.7% of the patients. No case of grade 3 constipation was observed.

Most patients (78.1%) either did not experience or rarely experienced fecal soiling, with only 4.8% frequently experiencing soiling. Table 4 shows that fecal soiling was not significantly correlated with age at surgery ($P > 0.05$). The frequency of no or rare soiling was highest in the 1-2 bowel movements per day group (91.7%). Frequent soiling was more common in patients with constipation (16.7%), indicating a significant correlation between soiling and bowel movement frequency ($P < 0.05$). (Table 3)

Table 3. Correlation Between Soiling and Various Factors (n = 41)

Characteristic	Soiling Frequency				p
	None n (%)	Rarely n (%)	Sometimes n (%)	Often n (%)	
Age	< 3 months (n=23)	13 (56.5)	4 (17.4)	4 (17.4)	0.9
	3 – 6 months (n=15)	8 (53.3)	4 (26.7)	3 (20.0)	
	> 6 months (n=3)	2 (66.7)	1 (33.3)	0 (0)	
Malformation	No fistula (n = 4)	2 (50.0)	0 (0)	2 (50.0)	0.5
	Rectourethral (n = 6)	3 (50.0)	2 (33.3)	1 (16.7)	
	Rectovestibular (n = 19)	12 (63.2)	4 (21.1)	3 (15.8)	
	Rectovaginal (n = 12)	6 (50.0)	3 (25.0)	1 (8.3)	
Bowel Movements/Day	Constipation (n = 6)	2 (33.3)	2 (33.3)	1 (16.7)	0.004
	1 – 2 (n = 24)	19 (79.2)	3 (12.5)	2 (8.3)	
	3 – 4 (n = 11)	2 (18.1)	4 (36.4)	4 (36.4)	

Nearly all patients were aware of their need to defecate, with 97.6% able to sense their urge, 97.6% able to communicate verbally, and 95.1% able to control it. One patient with Down syndrome lacked these abilities (2.4%). (Table 4)

Table 4. Monitoring of Sensory Perception During Defecation (n=41)

Sensory Perception During Defecation	Yes		No	
	n	%	n	%
Urge to defecate	40	97,6	1	2,4

Ability to express needs	40	97,6	1	2,4
Ability to withhold	39	95,1	2	4,9

Overall, 28 patients (68.3%) had good bowel function, 11 patients (26.8%) had average bowel function, and 2 patients (4.8%) had poor bowel function. There were 31 patients (75.6%) who had normal anal status, nine (22.0%) had anal stenosis, and one (2.4%) had rectal mucosa prolapse. All the patients had normal urinary function (100%). All boys (100%) had morning erections upon waking before urination.

DISCUSSIONS

In this study, the STT approach for anorectal malformation repair was used in all the patients. Most surgeries were performed in a single stage, with 33 cases (80.5%), whereas 8 cases (19.5%) underwent a two-stage procedure. The first stage involved creating a colostomy and the second stage involved combining colostomy closure with anorectal reconstruction.

None of the patients in our study experienced intraoperative complications. There were two cases (4.8%) of postoperative complications: one patient (2.4%) had bleeding at the surgical site, managed with compression and hemostatic medication, and one patient (2.4%) had an infection at the surgical site on the 3rd postoperative day, which was treated with intensive care and resulted in stable discharge. Nguyen Thanh Liem and Tran Anh Quynh (2015) also reported no intraoperative complications and early postoperative complications [7]. In contrast, Dao Trung Hieu (2013) reported three deaths (2.97%), primarily due to sepsis, and eight cases of wound infection (7.92%), including four cases of wound erythema and four cases of partial wound dehiscence [6]. Wound infection is a common complication, potentially due to the young age of children with weaker immune systems, the anatomical and functional characteristics of the anal region, and postoperative wound care. With improvements in sterile conditions in surgical facilities, preoperative

bowel cleansing, and prophylactic antibiotics, the rate of wound infection has significantly decreased.

Postoperative anal function was assessed on the basis of the number of bowel movements performed per day. Initially, patients often had frequent bowel movements, which gradually decreased. Results showed that 24 patients had 1-2 bowel movements per day, representing the highest proportion at 58.5%; 11 patients had 3-4 bowel movements per day, representing 26.8%, and only 6 patients had constipation, accounting for 14.6%. According to Nguyen Thanh Liem and Tran Anh Quynh (2015), 46 children (88.5%) had 1-2 bowel movements daily, 2 children (3.85%) had 3-4 bowel movements daily, 1 child (1.9%) had more than 4 bowel movements, and 3 children (5.8%) had bowel movements every 2-3 days [7]. Clinical observations suggest that in addition to surgical principles, the role of parents in establishing a regular bowel habit and adjusting the diet significantly affects bowel function. Long-term studies also emphasize the importance of daily bowel training for improving bowel function [8], [9].

Constipation is a common complication following anorectal reconstruction. In our study, constipation was observed in only six patients (14.6%), with 4.9% having grade 1 constipation and 9.7% having grade 2 constipation. Our results are consistent with those of Dao Trung Hieu's (2013) report of 15.22% cases of constipation. Nguyen

Thanh Liêm and Tran Anh Quynh (2015) reported a lower rate of 5.8% [7]. Constipation rates are also related to the type of surgical approach: Peña's posterior sacral approach has a 10.4% rate of constipation, the abdominal combined with Peña's posterior sacral approach has a 9.5% rate, and only 3.6% of children with low-type malformations experience frequent and severe constipation [3].

Postoperative fecal incontinence is a serious complication that affects the quality of life and social integration. In our study, the majority of patients (78.1%) had no or rare fecal incontinence, with only 4.8% experiencing frequent incontinence (see Chart 3.3). There was no correlation between fecal incontinence and age at surgery or type of malformation ($p > 0.05$), but a significant relationship with the number of bowel movements per day ($p < 0.05$) was observed. Specifically, in the group with 1-2 bowel movements per day, most had no or rare fecal incontinence (91.7%), in the 3-4 bowel movements per day group, incontinence was occasional and frequent at 45.3%, and in the constipation group, frequent incontinence was 16.7%. Thus, fecal incontinence was more common in the constipation and frequent bowel movement groups and less common in the 1-2 bowel movements per day group.

In our follow-up, fecal incontinence was influenced by parental care and training. Children whose parents actively managed bowel habits and ensured timely bowel movements had shorter durations and less severity of fecal incontinence. Conversely, children who did not complete bowel movements during one session experienced incontinence more frequently because of retained feces in the rectum. Parental education in bowel training significantly

improved incontinence. Postsurgical long-term care is crucial to achieve good bowel function.

Active bowel control is a critical criterion for assessing bowel function and the long-term effectiveness of surgical treatment for ARM. In our study, 95.1% of the patients achieved complete bowel control, while only 4.9% did not fully control their bowel function (Table 3.24). According to Krickenbeck's criteria, most patients had sensory perception during defecation: urge to defecate, 97.6%; ability to express their needs, 97.6%; and ability to withhold, 95.1%. Notably, one patient with Down syndrome lacked sensory perception during defecation (2.4%).

Persistent fecal incontinence and poor bowel control can lead to psychological issues, absence of school, and social limitations. Early intervention in patients with poor bowel control can improve bowel function and quality of life. Holschneider suggested teaching children self-control over bowel movements and establishing bowel habits as key to reducing chronic post-surgery issues [10].

Various scoring systems for bowel function assessments are available worldwide. Our study used the Krickenbeck scoring system to evaluate bowel function in patients aged > 36 months using three criteria: bowel control, constipation, and fecal incontinence. The results showed that 28 patients (68.3%) had good bowel function, 11 patients (26.8%) had average function, and only 2 patients (4.8%) had poor bowel function.

Our study and previous reports indicate that children with ARM treated with improved posterior sacral surgery while preserving the sphincter muscle generally have good or satisfactory bowel function,

with only a small proportion having poor function. This is a significant advantage of this method as it preserves sphincter integrity, leading to improved bowel function.

In our study, 75.6% of the patients had normal anal status, 22.0% had anal stenosis, and 2.4% had rectal mucosal prolapse. Anal stenosis is a common late complication, potentially due to scar tissue from hypertrophic scarring or noncompliance with postoperative dilation protocols. Our study found that 22.0% of patients had anal stenosis, often due to non-compliance with dilation procedures.

Urinary function and penile erection are also important considerations in boys. In our study, all patients had normal urinary function, and 100% of the boys had morning erections after waking and before urinating. These results are consistent with those of Nguyen Thanh Liem and Bui Duc Hau's (2001) findings of normal urinary function and penile erections in all boys [5].

CONCLUSIONS

The improved posterior sacral approach for preserving the sphincter in the treatment of intermediate-type ARM in children provides favorable long-term outcomes with good bowel function, excellent bowel control, and minimal complications. This method can be implemented in pediatric surgical facilities with appropriate equipment, anesthesia, and experienced surgeons.

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