

Prognostic factors for constipation following primary surgery of anorectal malformations in Yogyakarta, Indonesia

Willy Hardy Marpaung^{1,2*}, Rochadi¹

¹Pediatric Surgery Division, Department of Surgery, Faculty of Medicine, Universitas Gadjah Mada/Dr. Sardjito General Hospital, Yogyakarta, ²Department of Surgery, Raden Matta Her Hospital, Jambi, Indonesia

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ABSTRACT

Anorectal Malformation (ARM) is a congenital malformation of the anorectal area, occur approximately once in every 5000 live births. There are several morbidities could occur after surgery in ARM. Constipation is the most common morbidity post-surgery in ARM. This study was conducted to evaluate the prognostic factors affecting constipation in postoperative anorectal malformation. This was a case control study involving 74 children who undergo surgery in Dr. Sardjito General Hospital, Yogyakarta from 2008 to 2012. The prognostic factors are sex, type of ARM, age, type of ostomi, type of anoplasty operation, dehiscence of anoplasty, and anal dilatation.

There were 41 (55.4%) male patients and 33 (44.6%) female patients in this study. The risk of constipation in the patients with high level ARM was lower than in low level ARM with an OR of 0.150 (95% CI=0.032 - 0.711; p=0.008). Abdominoperineal pullthrough had higher risk for constipation than non abdominoperineal pullthrough (PSARP and ASA) with p = 0.003. The constipation rate was also different between patients with and without dehiscence of anoplasty with an OR of 0.139 (95% CI=0.033 - 0.584; p=0.005). The gender, types of colostomy, types of colon for colostomy, age of primary operation, and frequency of anal dilatation did not affect the constipation rate. In conclusion, postoperative constipation is still a major problem in surgical for anorectal malformations in Yogyakarta. Types of anomaly, primary operative procedures, and dehiscence of anoplasty operation are the prognostic factors that affect the constipation in anorectal malformations.

ABSTRAK

Malformasi anorektal merupakan salah satu kelainan bawaan yang terjadi pada daerah anorektal dengan insiden 1 dalam 5000 kelahiran hidup. Beberapa morbiditas dapat terjadi setelah pembedahan pada malformasi anorektal. Konstipasi pascabedah merupakan salah satu morbiditas yang paling banyak terjadi setelah tindakan bedah malformasi anorektal. Penelitian ini merupakan penelitian observasional dengan rancangan kasus kontrol, dan dievaluasi 74 anak-anak yang dilakukan operasi di RSUP Dr. Sardjito Yogyakarta mulai 2008 sampai 2012. Faktor-faktor prognostik yang dievaluasi adalah jenis kelamin, tipe ARM, tipe ostomi, tipe operasi anoplasti, dehisensi pada anoplasti dan tindakan dilatasi anus.

Hasil penelitian menunjukkan terjadinya konstipasi pada 18,9% pasien, kebanyakan terjadi pada pada ARM tinggi dan pada dehisensi anoplasti (p<0,05). Konstipasi terjadi pada 40% anak-

* corresponding author: will_1810@yahoo.com

anak pasca operasi *abdominal pull-through*. Dilatasi anus pasca operasi anoplasti memberikan pengaruh baik pada fungsi usus. Dapat disimpulkan, konstipasi merupakan morbiditas yang umum terjadi pada pasacabedah ARM. ARM tinggi dan dehisensi anus pascabedah merupakan faktor-faktor yang berpengaruh terhadap terjadinya konstipasi pascabedah.

Keywords: prognostic factors - constipation - anorectal malformation - constipation - children

INTRODUCTION

Anorectal malformations comprise a wide spectrum of diseases, which can affect boys and girls, and involve several organ systems, such as urinary and genital tracts. Anorectal malformations are congenital anomalies that occur in approximately 1 in 5000 births and the anomalies range from minor to complex anomalies.¹⁻³ There are several classifications for anorectal malformations proposed by some authors. Every classification describes type, treatment, and also outcome for each anomaly.¹⁻³ The exact etiology for anorectal malformations remains unclear and is likely multifactorial. There are reasons to believe there is a genetic component. The embryology of anorectal formation consists of complex mechanism. In anorectal malformations, the rectal pouch that will descent normally to the anal canal stops in a certain level and some have connection with urinary and genital tracts. This is why anorectal malformations may have connection (fistula) with these tracts.^{1,4}

The diagnostic for anorectal malformations consists of several examinations: physical, radiology, and laboratory examination.¹ The surgery treatments consist of neonatal surgery, primary repair (anoplasty), and the colostomy closure. The surgeon must decide the most appropriate surgical technique for every patient. In newborn period, the surgeon must decide whether the child require colostomy, or can undergo primary repair procedure. The

primary repair consists of several techniques such as posterior sagittal anorectoplasty, anterior sagittal anorectoplasty, anterior sagittal anoplasty, and abdominoperineal pullthrough. Each technique is used depends on the type of anomaly.^{1,2,3} Constipation is the most frequent disorder encountered after treatment for imperforate anus. It is also the most important problem to avoid after definitive repair for female patients with rectovestibular or rectoperineal fistula and for male patients with rectourethral fistula, imperforate anus without fistula, and rectoperineal fistula. Failure to avoid constipation can result in megarectum and megasigmoid, and can lead to fecal impaction and overflow incontinence.⁵ Constipation occurs usually due to the perirectal dissection causing a degree of denervation. The sacral anomaly also thought to be the cause of constipation, when the nerve for anorectal component is not well developed as well as the muscle complex. Constipation also appears due to hypomotility disorder secondary to chronic bowel dilatation that usually occurs in megarectum that common in transverse or loop colostomy when performed in newborn period. Post-operative constipation is also related to type of anorectal malformations and operative procedure of the anomaly in anorectal malformations.^{1,5} Types of the anorectal malformations are also known as prognostic factor for constipation. In perineal fistula and rectovestibular fistula, the constipation rate is 28.6-61.4%. In rectourethra

fistula, constipation occurs in 55.5%; while in rectovagina fistula constipation occurs in 25%.⁶The surgery techniques are also known as prognostic factor for constipation in anorectal malformations. Divided colostomy is recommended for the diverting technique than loop and double barrel colostomy. According to Pena⁷ large bowel that used as diverting colostomy for anorectal malformations is descendostomy. Transversostomy or ascendostomy diversion are not recommended due to lot of disused segment of the bowel, that related to constipation.^{1,6,7} Primary surgical technique is also a prognostic factor for constipation. Since Alberto Pena⁷ introduced posterior sagittal anorectoplasty in 1982, this technique is used all over the world for primary repair.⁶Constipation occurred in 78.5% high level anorectal malformations treated with PSARP, and 64.5% in low levels. PSARP was known superior than abdominoperineal pullthrough in defecation outcome.^{1,6}

Age at primary operation is also known to have impact in defecation function. Dehiscence on primary operation and daily anal dilatation are also known significant in bowel function on anorectal malformations.⁶ In this study, we evaluated these prognostic factors that influence constipation in anorectal malformations.

MATERIALS AND METHODS

This was a retrospective study which evaluates the prognostic factors that influence constipation in anorectal malformations in Yogyakarta, Indonesia. We evaluated anorectal malformations patients that had been operated for primary repair in Dr. Sardjito General Hospital, Yogyakarta, Indonesia from January 2008 to December 2012. The inclusion

criteria were patients that underwent primary operation for 1 stage procedure, or underwent colostomy closure for 3 stages procedure, had a complete operation. They were evaluated for constipation by telephone, mail, or direct examination. The prognostic factors were sex, age at primary operation, types of anorectal malformations, types of primary operation, types of colostomy, types of colon for colostomy, dehiscence of anoplasty, and anal dilatation frequency.

We defined constipation as the incapacity to empty the rectum spontaneously without help everyday, and needs changes in diet, laxatives, or enemas.^{6,7} Statistical analysis univariate, multivariate logistic regression analysis was performed with a p value < 0.05 considered significant. .

RESULTS

A total of 74 patients fulfilled the criteria for inclusion in this study. There were 41 (55.4%) male patients and 33 (44.6%) female patients. Forty five (60.8%) patients underwent primary operation more than 6 months of age, while 29 patients (39.2%) were operated less than 6 months of age.

Most of the anomalies were high level anomaly without fistula, that were 26 (35.1%) patients, while others were rectovestibular fistula in 14 (18.9%) patients, perineal fistula in 9 (12.2%) patients, rectourethral fistula in 7 (9.5%) patients, low level anomaly without fistula in 6 (8.1%) patients, midraphe fistula in 5 (6.8%) patients, rectovesica fistula in 4 (5.4%) patient, and cloaca in 3 (4.1%) patients. We divided these types of anomalies into high level anomaly in 54 (73%) patients, and low level anomaly in 20 (27%) patients (TABLE 1).

TABLE 1. Types of anorectal malformation-associated with constipation

Types of Anorectal Malformations	N (%)	Constipation	
		(+)	(-)
High level without fistula	26 (35.1%)	11 (42.3%)	15 (57.7%)
Low level without fistula	6 (8.1%)	1 (16.6%)	5 (83.4%)
Perineal fistula	9 (12.2%)	1 (11.1%)	8 (88.2%)
Midraphe fistula	5 (6.8%)	0 (0.0%)	5 (100%)
Rectourethral fistula	7 (9.5%)	4 (57.1%)	3 (42.9%)
Rectovesica fistula	4 (5.4%)	4 (100%)	0 (0.0%)
Rectovestibuler fistula	14 (18.9%)	3 (21.4%)	11 (78.6%)
Cloaca	3 (4.1%)	1 (33.3%)	2 (66.7%)

In types of colostomy procedure, the most frequent colostomy technique used in Dr Sardjito Hospital was loop colostomy. It is used in 53 (96.4%) patient, while divided colostomy in 2 (3.6%) patients (TABLE 2).

TABLE 2. Types of colostomy associated with constipation

Types of Colostomy Procedure	N (%)	Constipation	
		(+)	(-)
Divided	2 (3.6%)	1 (50.0%)	1 (50.0%)
Loop	53 (96.4%)	23 (43.3%)	30 (56.7%)

In types of colon for colostomy, right transversocolostomy was used in 22 (40%) patients, left transversocolostomy in 17 (30.9%) patients, sigmoidostomy in 15 (27.3%) patients, and descendostomy in 1 (1.8%) patient (TABLE 3).

TABLE 3. Types of colostomy associated with constipation

Types of colon for colostomy	N (%)	Constipation	
		(+)	(-)
Sigmoidostomy	15 (27.3%)	5 (33.3%)	10 (67.7%)
Right Transversocolostomy	22 (40%)	11 (50%)	11 (50%)
Left Transversocolostomy	17 (30.9%)	7 (41.1%)	10 (58.9%)
Descendostomy	1 (1.8%)	1 (100.0%)	0 (0.0%)

PSARP was used in 50 (67.6%) patients, while ASA in 19 (25.7%) patients, and abdomino perineal pullthrough in 5 (6.8%) patients (TABLE 4). Sixty three (85.1%) patients were having no dehiscence, while 11 (14.9%) patients were having dehiscence. Most of the patients, 70 (94.6%) patients, were routinely did anal dilatation, while 4 (5.4%) patients were not routinely dilate the anus after primary operation.

TABLE 4. Types of primary operation associated with constipation

Types of primary operation	N (%)	Constipation	
		(+)	(-)
Anterior sagittal anoplasty (ASA)	19 (25.7%)	1 (5.2%)	18 (94.8%)
Posterior sagittal anorectoplasty (PSARP)	50 (67.6%)	19 (38.0%)	31 (62.0%)
Abdominoperineal pullthrough	5 (6.8%)	5 (100.0%)	0 (0.0%)

TABLE 5. Correlation between prognostic factors with constipation

Variables		N (%)	Constipation		P	OR (95%CI)
			(+)	(-)		
Sex	Male	41 (55.4%)	13 (17.6%)	28 (37.8%)	0.674	0.813 (0.309 - 2.138)
	Female	33 (44.6%)	12 (16.2%)	21 (28.4%)		
Age at primary operation	≤ 6 months	29 (39.2%)	6 (8.1%)	23 (31.1%)	0.056	0.357 (0.122 - 1.047)
	> 6 months	45 (60.8%)	19 (25.7%)	26 (35.1%)		
Types of ARM	High level	20 (27.0%)	2 (2.7%)	18 (24.3%)	0.008*	0.150 (0.032 - 0.711)
	Low level	54 (73.0%)	23 (31.1%)	31 (41.9%)		
Types of colostomy procedure	Divided	2 (3.6%)	1 (1.8%)	1 (1.8%)	0.853	1.304 (0.077 - 21.982)
	Loop	53 (96.4%)	23 (41.8%)	30 (54.5%)		
Types of colon for colostomy	Descendostomy	1 (1.8%)	1 (1.8%)	0 (0.0%)	0.251	
	Non descendostomy	54 (98.2%)	23 (41.8%)	31 (56.4%)		
Types of primary operation	Abdominoperineal pullthrough	5 (6.8%)	5 (6.8%)	0 (0.0%)	0.003*	
	Non Abdominoperineal pullthrough	69 (93.2%)	20 (27%)	49 (66.2%)		
Dehiscence of anoplasty	Dehiscence	63 (85.1%)	17 (23%)	46 (62.2%)	0.005*	0.139 (0.033 - 0.584)
	Non dehiscence	11 (14.9%)	8 (10.8%)	3 (4.1%)		
Anal dilatation	Routine	70 (94.6%)	25 (33.8%)	45 (60.8%)	0.293	
	Non routine	4 (5.4%)	0 (0.0%)	4 (5.4%)		

Most patients were male (55.4%), with no statistically difference with female in correlation with constipation (p=0.674). Age at primary operation revealed no difference between patients that operated on 6 months of age, and over 6 months of age (p=0.056). There was a significantly different between patients with high level and low level (p=0.008) with

an OR of 0.150 (95% CI=0.032 - 0.711), while types of colostomy, and types of colon for colostomy did not show any significant difference with risk for constipation. Based on type of operation, in primary operation procedure, abdominoperineal pullthrough was significantly different with non abdominoperineal pullthrough (PSARP and ASA) with

$p = 0.003$. Dehiscence of anoplasty was also significantly different with non-dehiscence patients in risk from constipation in anorectal malformations with $p = 0.005$ with an OR of 0.139 (95% CI=0.033 - 0.584). Frequency of anal dilatation was not statistically significant in risk from constipation in anorectal malformations ($p= 0.293$).

DISCUSSION

Constipation still becomes a problem in post-operative anorectal malformations patients. In this study, the rate of constipation was 33.78%. This rate was not different from other studies. In 1995, Pena described his result, among 285 children in his study, 61.4% patients with vestibular fistula, 55% patients with bulbar fistula, and 41.4% patients with prostatic fistula constipated.⁷ Huang *et al.*,⁸ showed 8 constipation rate was 64.5% in low anomaly, and 78.5% in high anomaly. Male patients were more frequent than female in this study, where 41 (55.4%) patients were male. This finding was similar from the former study, when Smith and Stephens initially reviewed that 55-70% patients were male.⁶ A study in Singapore also showed that 63% anorectal malformations were male.⁹ There was no difference between sex according to constipation in our study. Age at primary operation has been suggested to influence the long-term functional outcome. In this study, there were no difference between patients that were operated at ≤ 6 months of age and > 6 months of age. A higher risk of organ injury will occur if we do not well identify the organ, even though there are some advantages in performing the definitive reconstruction earlier. The critical anal dilatation is easier to perform in an infant. Moreover, early reconstruction may theoretically allow the early development of neural pathways between

the anal canal and the brain, facilitating better anorectal sensation and sphincteric function. Eventhough, there is no evidence that early repair would provide better functional outcome.⁶ Based on the levels of anomaly, there was a statistically difference between the high level and low level anomaly, $p=0.008$, with OR=0.150 (95% CI: 0.032-0.711), meaning that the high level anomaly is lower in risk for constipation than the low anomaly. This result was different with other studies. Huang *et al.*⁸ showed that the constipation rate in high level anomaly was higher than the low anomaly (78.6% vs. 64.5%), similar to a study by Rintala, showing that the surgical management in low anorectal anomalies much better in continence than severe high anomalies.¹⁰ The reasons for high level anomaly have a higher risk for constipation are the hypoplasia of the voluntary sphincter muscles, and the severe sacral abnormalities. Other study showed different from these studies, where constipation particularly occurred in the low type. Constipation in low level anomaly usually related to dilatation of distal colon and rectum, causing hypomotility of the bowel. Diet and early management of constipation in low level usually give good result.⁵ Types of colostomy showed no difference between the divided colostomy and loop. Also the bowel for colostomy had no difference between descendostomy and non descendostomy with $p=0.251$. A divided descending colostomy is ideal for the management of anorectal malformations. The completely diverting colostomy provides bowel decompression as well as protection for the final repair of the malformations. A descending colostomy has several advantages over transversocolostomy because there is a relatively short segment of defunctionalized distal colon, and easier to cleanse the distal part.^{1,6,11} Our study did not show any

difference in this variable. We usually perform loop colostomy in our institution. Types for primary operation differed significantly between abdominoperineal pullthrough and non abdominoperineal pullthrough (ASA and PSARP). This finding is similar with other study. Surgical method of anorectal reconstruction is known as significant prognostic factor for constipation. Holschneider *et al.*,⁶ reported significantly better continence outcome in PSARP than abdominoperineal pullthrough. Abdominoperineal pullthrough usually breaks many nerves and muscle that usually related to defecation process.⁶ Evaluation of PSARP in our study showed that 19 of 50 patients (37.2%) treated with PSARP was constipated. This finding was not different from other study by Hassett, showing that 21% - 42% patients treated with PSARP was constipated.¹² Dehiscence of anoplasty operation significantly affected constipation rate. Wound dehiscence usually causing scar on the anus, and could make an anal stricture which related directly to constipation. Anal dilatation is important to prevent constipation. However, our study showed that anal dilatation does not influence the constipation rate. The anal dilatation might prevent anal stricture.⁶ The quality of life of anorectal malformations needs a long term follow up. Study about long term functional outcome and quality of life in high imperforate anus found that stooling patterns were perceived to worsen with age.¹³ Associated anomalies that usually found in anorectal malformations patients also influence the quality of life. Neurogenic bladder dysfunction, spinal cord malformation, sacral malformation, and prostatic fistula all are negative predictive factors for bowel score at five years.¹⁴

CONCLUSION

Postoperative constipation is still a major problem in surgical for anorectal malformations in Yogyakarta, Indonesia. Types of anomaly, primary operative procedures, and dehiscence of anoplasty operation were the prognostic factors that affect the constipation in anorectal malformations.

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