DOI: https://doi.org/10.54393/pjhs.v4i06.891

PAKISTAN JOURNAL OF HEALTH SCIENCES
https://thejas.com.pk/index.php/pjhs
Volume 4, Issue 6 (June 2023)

Original Article

Fecal Continence in Patients with Low Rectal Malformation After Anoplasty

Abdul Jabbar Baloch[1*], Shazia Perveen[2], Nitu Kumari[3] and Sana Aslam[4]

[1]Civil Hospital Turbat
[2]Sheffield Hospital England
[3]National Institute of Child Health, Karachi
[4]Bilawal Medical College, Liaquat University of Medical & Health Sciences, Jamshoro, Pakistan

ARTICLE INFO

Key Words:
Anoplasty, Fecal Continence, Incontinence, Morbidity, Rectal Malformation

How to Cite:
Baloch, A. J. ., Perveen, S. ., Kumari, N. ., & Aslam, S. . (2023). Fecal Continence in Patients with Low Rectal Malformation After Anoplasty: Fecal Continence with Low Rectal Malformation. Pakistan Journal of Health Sciences, 4(06). https://doi.org/10.54393/pjhs.v4i06.891

***Corresponding Author:**
Abdul Jabbar Baloch
Civil Hospital Turbat
jabbarbaloch327@gmail.com

Received Date: 1st June. 2023
Acceptance Date: 21st June, 2023
Published Date: 30th June, 2023

ABSTRACT

Anorectal malformations (ARMs) affect between 1:2000-2500 births comprising a heterogenous spectrum of malformations that vary considerably in their anatomical characteristics, complexity, and functional prognosis. Occasionally, the diagnosis is delayed to later infancy especially in cases where the bowel outlet is stenotic but at or near the proper anal position. **Objective:** To determine the frequency of fecal continence in patients with low rectal malformation after anoplasty. **Methods:** This was prospective Cross-Sectional Study conducted at Department Pediatric Surgery, National Institute of Child Health (NICH), Karachi, Pakistan from September, 2020 to March, 2021. All patients who fulfilled the inclusion criteria and visited to NICH, Karachi were included in the study. Informed consent was taken from parents / guardians after explaining the procedure, risks and benefits of the study. Parents/Guardian of patients were asked to fill the study questionnaire containing the Kelly Scoring System to assess the status of fecal continence of the patients. All the collected data were entered into the proforma and analyzed on SPSS. **Results:** Out of 189 patients, 63% were male and 37% were female. Fecal continence was found in 71.4% patients, statically significant with Body Mass Index. Mean ± SD of age was 3.7 ± 1.5 days. **Conclusions:** It is to be concluded that fecal continence is a frequent finding of low rectal malformation. Patients with low rectal malformation should be worked up for the fecal continence after anoplasty.

INTRODUCTION

Congenital abnormalities where the anus fails to open normally onto the perineum classified as low and high types based on the relationship of the terminal colon to the levator muscles of the pelvic floor. It is considered the most frequently encountered anomaly in neonatal pediatric surgery which occur approximately 1 in 5000 live births [1]. Despite recent advancement of surgical techniques, treatment of anorectal malformation is still a difficult challenge for pediatric surgeons [2]. There is a wide spectrum of presentation ranging from low anomalies with perineal fistula having simple management to high anomalies with complex management. Advances in the imaging techniques with improvement in knowledge of the embryology, anatomy and physiology of ARM cases have refined diagnosis and initial management [3]. There has been marked improvement in survival of such patient over the last century. The management of ARM has moved forward from classical procedures to PSARP to minimal invasive procedures. But still the fecal and urinary incontinence can occur even with an excellent anatomic repair, mainly due to associated problems [4]. There has been a paradigm shift in approach to these patients which involves holistic approach to the syndrome of Anorectal malformations with a long-term goal of achievement of

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complete fecal and urinary continence with excellent quality of life [5]. Low anorectal malformation comprises about half of all anorectal anomalies. Most of the literature concerning management of anorectal anomalies is centered around the treatment and outcome of high anomalies [6]. The management of low anomalies has been considered significantly less challenging than high anomalies. Also, the outcome of low anomalies has traditionally been considered good. However, recent more critical long-term follow-up reports show a different picture [7]. Many patients with low anomalies suffer from long-term anorectal functional problems, especially constipation and fecal incontinence but also soiling that occurs in a significant percentage of patients [8]. In a study conducted by Rintala, it is found that good fecal continence in 60% and normal bowel function in only 15% of adult patients with low malformations after anoplasty [9]. This study was designed to determine the frequency of fecal incontinence in patients with low rectal malformation after anoplasty. Since, no such study has ever been conducted in children. The emphasis on the surgical management of low anorectal anomalies is to use as minimally invasive operative methods as possible and preserve the native mechanisms of continence that usually are much better preserved than in more severe high anomalies.

METHODS

The cross-sectional study was conducted at Department of Pediatric Surgery, National Institute of Child Health (NICH), Karachi from September, 2020 to March, 2021. 189 newborns up to 28 days old of either gender, who had received anoplasty 2 to 4 years ago, were included in the study via non-probability, consecutive method while children who had undergone post sagittal anorectoplasty and parents or guardians who were unwilling to provide informed consent were excluded from the study. The sample size was calculated via W.H.O sample size calculator with frequency of fecal continence with low rectal malformation after anoplasty, taken as 60% with confidence level of 95% and margin of error as 7%. The Non-Probability, Consecutive Sampling technique used for data collection through Performa by all those who came in inclusion criteria. Low Rectal Malformation was defined as having presence of the anal opening was missing or in the wrong place or the anus or rectum was too small to allow stool to pass or the rectum was connected to the perineum, an area of skin between the anus and genitals while fecal continence was measured by using Kelly protocol with Kelly Score > 3.00, was labeled as positive.

Table 1: Kelly Protocol

Kelly Protocol	
Accidents	
Never	2
Occasional (At least three times per week)	1
Daily	0
Staining	
Never	2
Occasional (At least three times per week)	1
Daily	0
Sphincter squeeze	
Strong	2
Weak	1
Absent	0

The data were analyzed with the help of SPSS version-21.0. Mean ± Standard deviation was calculated for quantitative variables while frequencies and percentages were calculated for qualitative variables. Chi-square test was applied to check for association between different variables while p-value < 0.05 was considered statistically significant.

RESULTS

Out of 189 patients 63% were male and 37% were female as, 41.8% population of our study were belonging to rural areas and 58.2% living in urban areas. The Mean ± SD of age, weight, height and BMI were 3.7 ± 1.5, 4.1 ± 0.8, 0.46 ± 0.25, 8.2 ± 2.4 respectively. The frequency of fecal continence was found to be in 71.4% patients shown in Table 2. The table depicts the description of the sample in frequencies (percentages).

Table 2: Sample Description (N=189)

Sample Description		
Age		3.7 ± 1.5 days
Weight		4.1 ± 0.8 kg
Height		0.46 ± 0.25 meter
BMI		8.2 ± 2.4 (kg/m2)
Gender	Male	119 (63%)
	Female	70 (37%)
Residency	Rural	79 (41.8%)
	Urban	110 (58.2%)
Fecal Continence	Yes	135 (71.4%)
	No	54 (28.6%)

Educational status of mother showed that 58.2% were illiterate, 3.2% had primary education, and 17.5% had secondary while 21.2% were intermediate. Educational status of father showed that 29.6% were illiterate, 3.7% had primary education, 5.3% had secondary, 31.2% were intermediate while 30.2% were graduate and above as shown in Figure 1. Figure represents the educational status of mothers and fathers of patients.

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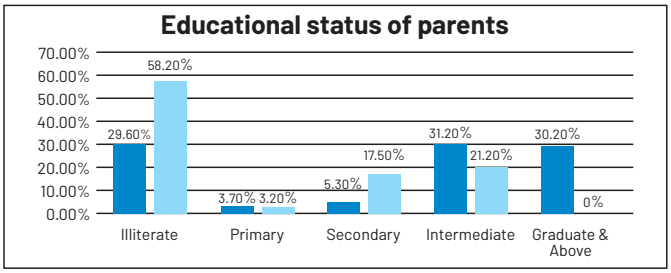

Figure 1: Educational Status of Parents (N=189)

Stratification of age group, gender, educational status of mother & father and body mass index with respect to fecal continence was done and it shows association with Body Mass Index *p* -value 0.0001 rest of variable had no significance as shown Table 3. The table represents the relation of fecal continence with Gender, BMI, Age Group and Maternal and Paternal Educational Status.

Table 3: Stratification of Age Group with Fecal Continence (N=189)

Variables		Fecal Continence		p-value
		Yes	No	
Gender	Male	88 (46.6%)	31 (16.4%)	0.317
	Female	47 (24.9%)	23 (12.2%)	
Mother's Educational Status	Illiterate	79 (41.8%)	31 (16.4%)	0.647
	Primary	5 (2.6%)	1 (0.5%)	
	Secondary	21 (11.1%)	12 (6.3%)	
	Intermediate	30 (15.9%)	10 (5.3%)	
Father's Educational Status	Illiterate	41 (21.7%)	15 (7.9%)	0.467
	Intermediate	37 (19.6%)	22 (11.6%)	
	Primary	5 (2.6%)	2 (1.1%)	
	Secondary	8 (4.2%)	2 (1.1%)	
	Graduate & Above	44 (23.3%)	13 (6.9%)	
BMI In Kg/M2	4 – 7	90 (47.6%)	15 (7.9%)	0.0001
	>7	45 (23.8%)	39 (20.6%)	
Age Group In Days	1 – 3	78 (41.3%)	24 (12.7%)	0.097
	>3	57 (30.2%)	30 (15.9%)	

DISCUSSION

Anorectal malformations (ARMs) are complex congenital malformations that result from the abnormal formation of the developing hindgut [10, 11]. The care of this condition was revolutionized in 1982 with the description of the posterior sagittal anorectoplasty (PSARP) developed for the first time by Pena and Devries for a precise anatomic reconstruction as well as stratification of the spectrum of anomalies [12]. The ultimate goals of the surgical repair are to disconnect the rectum from the urinary or genital tract if a fistula is present and create an adequately sized anal opening centered within the sphincter complex. These surgical principles maximize the chance the child will be continent of stool later in life [12, 13]. Although the functional outcomes in ARM are generally good, particularly if the sacrum and spine are normal, a proportion of patients remain fecal incontinent, and their optimal management provides a challenge to the surgeon [14-18]. Fecal incontinence after PSARP is usually due to poor pelvic muscular and sensory development which can impair the continence mechanisms [10, 19, 20]. For a child with good continence potential, incontinence can result from a technical complication related to their original reconstruction. Such complications include mis location of the anoplasty, stricture, remnant of the original fistula (ROOF), or rectal prolapse, which might not be discovered until years later when the child attempts to toilet train as found by Pena *et al.*, in 2003 and 2007 [21, 22]. Most children with low malformations are referred to surgical care as newborns. Occasionally, the diagnosis is delayed to later infancy especially in cases where the bowel outlet is stenotic but at or near the proper anal position. We have adopted a management policy that does not include routine diagnostic imaging in relation to the level of the anomaly. Although the distance between the rectal blind pouch and the perineum may be roughly estimated by invertogram, lateral pelvic radiograph or perineal ultrasound examination, the results obtained from these studies are more or less inaccurate [23-25]. Newborns with anal stenosis or perineal fistula usually pass meconium in the first 48 h which together with careful clinical examination enables correct diagnosis in vast majority of cases. Instead, if a baby fails to pass meconium after the first 24–48 h and there is no evidence of perineal fistula after detailed clinical examination including gentle perineal probing a double-barreled diverting sigmoid ostomy is performed. Soiling after ARM repair is a source of major morbidity in children born with an ARM [26-29]. These symptoms often do not present until years after the initial PSARP when it is discovered that the child is unable to successfully toilet train. The clinician is then tasked to identify the cause of the patient's incontinence and determine whether the cause is ideally addressed with a medical (e.g. bowel management) or surgical solution. The goal of the assessment is to identify whether an anatomic source for soiling is present, as a medical solution is less likely to be successful, for example in a patient with a strictured or mis located anoplasty. Furthermore, attempts to treat a child with laxatives or enemas without a detailed anorectal examination may delay diagnosis of an anatomic cause of the incontinence. Thus, any provider caring for such patients must first make an anatomic assessment of the surgical repair before treating the functional problem of soiling [30]. Rectal prolapse is seen following PSARPs, and is more common in more complex malformations. Poor muscle tone and constipation are believed to be factors that predispose the patient to this complication. The optimal management is dictated by the

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degree of prolapse. If relatively minor (<5 mm), a perineal-only resection of the prolapsing mucosa is adequate. However, in cases of more severe prolapse, a formal reoperation is needed to adequately secure the rectum to the posterior limit of the muscle complex. The mean age in our study was found to be 3.7 ± 1.5 days. Wood *et al.*, noted to have a mean age of 3.7 days. Out of 189 patients, 119 (63%) were male while 70 (37%) were female similarly Wood *et al.*, found 109 (71%) male patients. In this study, 79 (41.8%) were resident of rural areas while 110 (58.2%) were resident of urban areas. In present study, fecal continence was found to be in 71.4% patients. The study done by Wood *et al.*, noted the prevalence of fecal continence as 511 (75%) that is similar to our study [31]. Another study by Kyrklund *et al.*, reported the same in 76% cases [32]. In present study, stratification of confounders / effect modifiers with respect to fecal continence, insignificant difference was reported in age group (p=0.097), gender (p=0.317), educational status of mother (p=0.647), educational status of father (p=0.467), socioeconomic status (p=0.898), residential status (p=0.889) while significant difference was noted in body mass index (p=0.0001).

CONCLUSIONS

It is concluded that fecal continence is a frequent finding among patients with low rectal malformation.

Authors Contribution

Conceptualization: AJB
Methodology: SP, NK
Formal analysis: SA
Writing-review and editing: SP, NK, SA, AJB

All authors have read and agreed to the published version of the manuscript.

Conflicts of Interest

The authors declare no conflict of interest.

Source of Funding

The authors received no financial support for the research, authorship and/or publication of this article.

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