A comparative study between early and late correction of cases of anorectal malformation assessed by fecal and urinary functional outcome

# Original Article

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#### **ABSTRACT**

**Background:** Anorectal malformation (ARM) includes a wide spectrum of anomalies of the anorectal and urogenital region. They are not rarely associated with orthopedic, spinal, and neurologic abnormalities effecting the short- and long-term outcome. anorectoplasty at an early age is important for obtaining a better life quality in ARM patients.

**Aim:** The work aims to study defecation and urinary functional outcome according to the timing of correction of ARM. **Patients and Methods:** This was a cross—sectional study conducted at patients with anorectal malformations (perineal fistula, rectourethral fistula, rectovesical fistula, vestibular fistula, no fistula, anal stenosis), at the Ain Shams pediatric surgery department and Qena pediatric surgery unit.

**Results:** Bowel function and quality of life were significantly higher in the early correction group compared with late correction group(P < 0.001). The mean of the quality of life in the early and late correction groups was  $12.83 \pm 0.59$  and  $11.13 \pm 2.73$ , respectively.

**Conclusion:** Early anorectoplasty and early appropriate bowel management are advisable, as that is essential for less fecal and urinary functional complications, and for proper life quality.

Key Words: Anorectal malformation, anorectoplasty, bowel movement, fistula.

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## INTRODUCTION

Anorectal malformation (ARM) encompasses a broad range of abnormalities in the anorectal and urogenital regions. To determine the 'continence predictor index' in patients with ARM, factors such as the type of ARM, the quality of sacrum, and the status of the spine were considered<sup>[1,2]</sup>. The quality of life of a child with ARM is influenced by various factors, including fecal continence, constipation, and associated genitourinary anomalies, which occur in ~30–50% of cases<sup>[3,4]</sup>.

While it is easy to identify ARM in the neonatal period, many countries face a prevalent issue of delayed diagnosis. This delay in diagnosis can result in increased morbidity, emphasizing the importance of timely anorectoplasty for optimal postoperative bowel function. Therefore, performing anorectoplasty at an earlier age may be beneficial for bowel function<sup>[5,6]</sup>. However, few studies have explored the relationship between the optimal timing of anorectoplasty and long-term postoperative bowel function<sup>[5]</sup>. Therefore, our study aimed to investigate the

functional outcomes of defecation and urine based on the timing of ARM correction.

## PATIENTS AND METHODS:

This was a cross–sectional study conducted at The Ain Shams Pediatric Surgery Department and Qena Pediatric Surgery Unit.

A study carried out on pediatric patients who present to the pediatric surgery clinic of Ain Shams University hospitals and Qena University hospitals with postoperative corrected ARM Before inclusion in the study, all participants gave informed consent, and the study protocol was approved by the Local Ethics Committee.

Inclusion criteria: patients with anorectal malformations (perineal fistula, rectourethral fistula, rectovesical fistula, vestibular fistula, no fistula, anal stenosis).

Exclusion criteria: patients with cloaca, pouch colon, rectal atresia/stenosis, rectovaginal fistula, or H-type fistula.

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## Research strategy and methodology

## The participants were divided into two groups:

- (a) Group 1 (early correction): patients who end all procedures for correction of ARM before the age of 6 months.
- (b) Group 2 (late correction): patients who end all procedures for correction of ARM after the age of 6 months (Table 1).
- (i) Then we compared the two groups according to: (constipation, soiling, voluntary bowel movements, dysfunctional voiding scoring, and quality of life) to study the results and the efficacy of the early correction of ARM.

**Table 1:** Age when patients completed their interventions (in the late group)

	Number (%)
Cases completed 6 months-1 year old	14 (46.7)
Cases completed 1-2 years old	11 (36.7)
Cases completed 2-4 years old	5 (16.6)

A detailed history and examination were taken from all participants.

Including type of ARM, time of presentation, timing of interventions, associated genitourinary anomalies, quality of the sacrum, and status of the spine.

Evaluating colorectal functions in the postoperative period was done using Krickenbeck classification for postoperative results (age >3 years)<sup>[6]</sup>.

Assessment of urinary functions was done using the dysfunctional Voiding Scoring System questionnaire over the last month (in toilet-trained children, >3 years).

Quality of Life for corrected ARM children (3–4 years old) was assessed according to the questionnaire Wigander *et al.*<sup>[7]</sup>.

## Statistical analysis

Simple descriptive statistical analysis for the variables, we compared the two groups using the proper statistical tests (parametric or nonparametric) according to the distribution of variations and the types of variables involved.

Statistical analysis was done using IBM SPSS statistics for windows, Version 23.0. Armonk, NY: IBM Corp.

## **RESULTS:**

Figure 1 shows comparison among the two studied groups regarding sex.

The mean age in the early and late correction groups was  $6.78\pm2.93$  years and  $6.30\pm3.31$  years, respectively. There was no statistically significant difference between the two groups regarding sex and age (P>0.05).

There was no statistically significant difference between the two groups in quality of the sacrum or status of the spine.

The comparison between the two studied groups regarding type of ARM shows no statistically significant difference (P>0.05) as illustrated in (Fig. 2).

Comparison between the studied groups regarding constipation is illustrated in (Fig. 3).

A statistically significant difference was observed between the two groups regarding soiling (P=0.011) (Fig. 4).

Voluntary bowel movements were found in 100% of children in the early correction group and in 80% of children in the late correction group. Voluntary bowel movements were significantly higher in the early correction group compared with late correction group (P=0.010).

Urinary incontinence was found in one case in both the early correction group and late correction groups. No statistically significant difference was found between the two groups regarding urinary incontinence (P > 0.05).

Associated genitourinary anomalies were found in 13.3% of children in the early correction group and in 16.7% of children in late correction group (Fig. 5).

## In the early group:

- (a) Two cases had VUR (vesicoureteric reflux), one developed recurrent UTI (urinary tract infection) and needed deflux injection, the other improved on follow-up.
- (b) One case had hydronephrosis improved on follow-up.
- (c) One case had left renal stones at the age of 1.5 years old.

## In the late group:

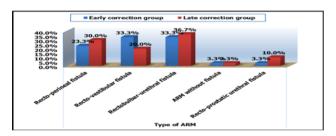
- (a) A case of bilateral VUR, deflux injection was done but failed and the patient needs ureteric reimplantation, another case had VUR and improved on follow up.
- (b) The case of recto-prostatic fistula was complaining of drippling of urine till age of 4 years, then cystoscopy was done and revealed diverticulum at site of ligated fistula, relegation was done, the pt after that completely continent.

- (c) Two cases of hydronephrosis relieved on follow-up.
- (d) A case of urachal cysts.
- (e) A case with history of rectourethral fistula, now the pt is 23 years with drippling of urine once or twice per month, history of sigmoidectomy for resistant constipation at the age of 16 years old (Table 2).

The mean quality of life in the early and late correction groups was  $12.83\pm0.59$  and  $11.13\pm2.73$ , respectively. Quality of life was significantly higher in early correction group compared with late correction group (P<0.001) (Fig. 6).



Fig. 1: Comparison among the two studied groups regarding sex.



**Fig. 2:** Comparison between the two studied groups regarding the type of anorectal malformation.

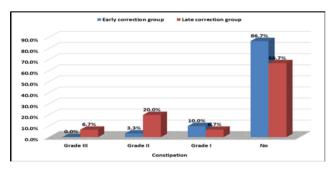


Fig. 3: Comparison between the groups studied regarding constipation.

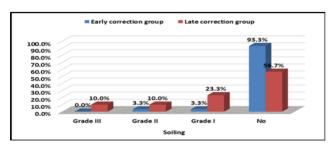


Fig. 4: Comparison between the groups studied regarding soiling.

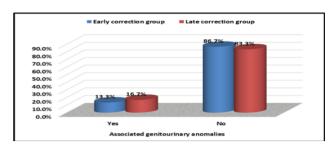


Fig. 5: Associated genitourinary anomalies in early and late groups.

Table 2: Associated genitourinary anomalies

	Number (%)
VUR	4 (6.67)
Hydronephrosis	3 (5)
Renal stones	1 (1.67)
Urachal cyst	1 (1.67)
Urethral diverticulum	1 (1.67)

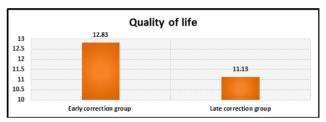


Fig. 6: Comparison between the groups studied regarding quality of life.

#### **DISCUSSION**

Our study revealed that rectovestibular fistula and rectobulbar-urethral fistula were the most prevalent malformations in the early correction group, accounting for 33.3% of cases, followed by rectoperineal fistula in 23.3% of cases. In the late correction group, rectobulbar-urethral fistula was the most common malformation, accounting for 36.7% of cases, followed by rectoperineal fistula in 30% of cases. This finding is consistent with the results of Harris and Wilcox's study<sup>[8]</sup>. In the study of Harumatsu and colleagues, the recto-urethral fistula was the most common malformation among their studied cases (80%). And the recto-prostatic urethral fistula was the least common malformation (3.8%)<sup>[5]</sup>.

In our study, the quality of sacrum and status of spine was good in both groups and consequently no affection the outcomes.

In our work constipation after the operation was evaluated, we found that in the early correction group, constipation grade I that managed by change in diet was found in 10% of the children, and grade II that managed by laxatives in 3.3% children. In the late

correction group, constipation grade I managed by a change in diet was found in 6.7% of the children, grade II that managed by laxatives in 20% of children, and grade III was resistant to laxatives and diet in 6.7% children. There were 26 children (86.7%) with no constipation in the early correction group. There was no statistically significant difference between the two groups regarding constipation.

In consistent with our results, the study of Harumatsu *et al.*<sup>[5]</sup> and Koga *et al.*<sup>[9]</sup> reported that the early correction group showed a better trend toward improvement of constipation score than the late correction group.

We found that continent children were high in early corrected groups with good improvement in voluntary bowel movements, which also was illustrated in the study of Statovci *et al.*<sup>[10]</sup> and the study of Harumatsu *et al.*<sup>[5]</sup>.

This difference between the two groups may be because early stool passage leads to the early establishment of the brain–defecation reflexes<sup>[5]</sup>.

We found that urinary anomalies were the most common associated anomalies with ARM, that was in conjugation with the study of Shenoy and colleagues.

We did good assessment of urological functions and anomalies as the study of Harris and Wilcox proved that chronic kidney disease and end-stage renal disease are the greatest causes of decreased life expectancy in patients with ARM<sup>[8]</sup>.

Urinary incontinence for less than half the time was found in just one case in the early correction group and late correction group. So, there was no statistically significant difference was found between the two groups regarding urinary incontinence.

In contrast to our study, the study of Bjoersum-Meyer *et al.*<sup>[11]</sup> found that there were many cases of patients with anorectal malformations who underwent surgery, Complaining of long-term outcomes of urinary incontinence.

This difference may be owing to heterogeneity, as we were not able to delineate outcomes by type of ARM, type of surgical repair, or associated anomalies. The methods for reporting outcomes were also largely heterogeneous because of the different questionnaires used in the studies and poor or absent definitions of reported outcomes<sup>[11]</sup>.

We found that the quality of life was significantly higher in early correction group compared with late correction group, that was proved also in the study of Turowskietal *et al.*<sup>[12]</sup> where they said that the delay in detection and correction of ARM is usually associated with more morbidity than early presentation.

And the study of Harumatsu *et al.*<sup>[5]</sup> showed that anorectoplasty at an early age was important for obtaining a better life quality in ARM patients.

We documented that our study has limitations, including the small number of cases and limited follow-up

### CONCLUSION

- (a) The early performance of anorectoplasty achieved a better bowel function, especially in terms of continence, improved soiling, and good bowel movement. In addition to that, the early performance of anorectoplasty has a great effect in improving the quality of life.
- (b) Evaluation of the urological system is very important.

Therefore, we advocated early anorectoplasty and early appropriate bowel management as that is essential for less fecal and urinary functional complications, and for proper life quality.

## **CONFLICT OF INTEREST**

There are no conflicts of interest.

#### REFERENCES

- 1. Divarci E, Ergun O. General complications after surgery for anorectal malformations. Pediat surg int 2020; 36:431–45.
- 2. Stephens F, Smith E, Santulli T, Falcone JrR, Levitt M, Peña A, *et al.* Anorectal malformations. Clin colon rectal surg 2018; 31:061–70.
- 3. Peña A, Bischoff A, Peña A, Bischoff A. Urologic problems in anorectal malformations. Surg Treat Colorectal Problems in Children 2015:371–96.
- 4. Senel E, Akbiyik F, Atayurt H, Tiryaki HT. Urological problems or fecal continence during long-term follow-up of patients with anorectal malformation. Pediat surg int 2010; 26:683–9.
- Harumatsu T, Kaji T, Nagano A, Matsui M, Yano K, Onishi S, et al. Early definitive operation for patients with anorectal malformation was associated with a better long-term postoperative bowel function. Pediat Surg Int 2021; 37:445–50.

- 6. Levitt MA, Peña A. Anorectal malformations. Orphanet j rare dis 2007; 2:1–3.
- 7. Wigander H, Nisell M, Frenckner B, Wester T, Brodin U, Öjmyr-Joelsson M. Quality of life and functional outcome in Swedish children with low anorectal malformations: a follow-up study. Pediat Surg Int 2019; 35:583–90.
- 8. Harris KT, Wilcox DT. Management of the urological tract in children with anorectal malformations—a contemporary review. Therap Adv Urol 2023; 15:17562872231161468.
- 9. Koga H, Miyake Y, Yazaki Y, Ochi T, Seo S, Lane GJ, Yamataka A. Long-term outcomes of male imperforate anus with recto-urethral fistula: Laparoscopy-assisted anorectoplasty versus posterior sagittal anorectoplasty. Pediat surg int 2022; 38:761–8.

- Statovci S, Heta H, Hyseni N, Grajçevci S, Berisha M. Çeku G. Treatment of Children with Anorectal Malformations-10-Year Review. Open Access Library Journal 2015; 2:1–8.
- 11. Bjoersum-Meyer T, Kaalby L, Lund L, Christensen P, Jakobsen MS, Baatrup G, *et al.* Long-term Functional Urinary and Sexual Outcomes in Patients with Anorectal Malformations—A Systematic Review. Eur Urol Open Sci 2021; 25:29–38.
- 12. Turowski C, Dingemann J, Gillick J. Delayed diagnosis of imperforate anus: an unacceptable morbidity. Pediatric surgery international 2010; 26:10836.