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Management and Complications of Staged Approach in Children with Anorectal Malformations; Experience at Tertiary Care Facility

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ABSTRACT

Objective: To present the data of children with anorectal malformations treated with a staged approach, including the complications of each stage.

Study Design: Prospective longitudinal Study

Place and Duration of Study: Paediatric Surgery Departments, Combined Military Hospital, Malir and Multan, Pakistan from Jan 2017 to Jun 2019 and Jul 2019 to Jul 2022, respectively.

Methodology: Twenty-four children with anorectal malformations were included. All were treated with a staged approach. The complications encountered during each approach were documented.

Results: A total of 24 patients were operated on. Fourteen (58.3%) were female, whereas 10 (41.7%) were male patients. The age range was 1-4 years, with a mean of 2.00 ± 0.97 years. Female patients had an ARM with a rectovestibular fistula. Two (20%) of the male patients had rectal atresia, a recto-urethral bulbar fistula, a recto-urethral prostatic fistula, a recto-bladder neck fistula, or a perineal fistula. Two (8.3%), three (12.5%), two (8.3%), and one (4.1%) patients who had stage 1 had stomal stenosis or retraction, wound dehiscence, wound infection, and stoma prolapse respectively. In the second stage, two patients (8.3% of all patients) had an infection, a wound dehiscence, a dehiscence of the perineal body, and anal stenosis. In the other patient, one (4.1%) had an anal mucosal prolapse and severe urethral injury. After stage 3, only one patient (4.1%) had a wound infection.

Conclusion: Patients with ARM are generally treated with a multistaged approach, and each stage of treatment has its own complications.

Keywords: Anoplasty, Anorectal malformations, Rectovestibular fistula.

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INTRODUCTION

Anorectal malformation (ARM) is a spectrum of congenital anomalies that is one of the most common presentations in pediatric surgical clinics. The incidence ranges between 1 in 2000 and 1 in 5000 live births. The exact cause of such malformations is unclear, and it is assumed to be multifactorial.[1,2]

Treatment for ARM is a real challenge, both for the clinician and for the parents. Even after the complete treatment, children still keep on visiting the clinic because of the effects of the primary disease and associated anomalies, which may affect as much as 50% of the children with ARM.[3] After thorough investigations, treatment of ARM includes initial resuscitation with intravenous fluid and broad-spectrum antibiotics, followed by surgical intervention, which includes primary or staged anesthesia.[4] While posterior sagittal anorectoplasty (PSARP) is thought to be the best surgery for ARM. The long-term results in terms of faeces incontinence or faeces soiling are very different between PSARP and other common surgeries.[5,6]

In a staged approach, procedures can be completed in either two or three stages. We adopted a three-stage approach in which the child is first subjected to a protective loop or divided pelvic colostomy, followed by PSARP, followed by stoma closure.[7] This approach carries the morbidity of multiple surgeries, complications from stoma formation such as prolapse, retraction, and stenosis, and complications from stoma closure such as anastomotic leak, wound dehiscence, and infection.[8]

On the other hand, in a primary procedure, anoplasty is performed without a diverting colostomy. Both approaches have their own merits and demerits.

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In terms of primary anoplasty, there are some absolute contraindications, such as severe life-threatening associated congenital malformations, sepsis, necrotizing enterocolitis with pneumoperitoneum, extreme prematurity, and common cloaca. There is considerable risk to the urinary tract because the surgeon is unaware of a precise anorectal defect.[9]

With the advent of minimal access surgery, laparoscopic repair of high-type ARM became a popular procedure. With laparoscopically assisted anorectal pull-through, the surgeon can avoid a lot of dissection, find a fistulous communication between the rectum and the renal tract, and place the rectum correctly in the muscle complex.[10] We aimed to present the data of children with ARM treated with a staged approach, including the complications of each stage. The rationale of the study was that a staged approach carries more morbidity in the treatment of anorectal malformations.

METHODOLOGY

The prospective longitudinal study was carried out at the Departments of Paediatric Surgery, Combined Military Hospital, Malir, and Multan, from January 2017 to June 2019 and July 2019 to July 2022, respectively after seeking approval from the Hospital Ethical Review Committee (Number 13/Trg-64/2023 dated February 2023).

Inclusion Criteria: Children of either gender with anorectal malformations were included.

Exclusion Criteria: Patients who failed to follow up were excluded.

All of the patients underwent staged repair of the ARM with a sequence of surgeries, such as pelvic divided colostomy, PSARP after high-pressure distal colostogram in male babies for delineation of fistulous communication with the urinary tract, and finally stoma closure after achieving the target anal dilatation for age. All patients underwent these surgical procedures performed by a single consultant surgeon. Out of 24 patients, 16 were booked in the same hospitals, whereas 8 were referred from other medical facilities. On presentation, patients were assessed in clinical detail for ARM and associated anomalies. The results were confirmed, and they showed that there was no anal opening, perineal fistula, vestibular fistula, no fistulous opening, buttock development, natal cleft, and development of the spine, coccyx, and sacrum. We requested appropriate investigations, including a lateral cross-table X-ray 24 hours after birth, renal system ultrasound, echocardiography, spine ultrasound, blood chemistry, and urine analysis. Parents were counseled in detail about the disease, treatment plan, and timeline. For male babies, a pelvic divided colostomy was performed once prepared for surgery 24 hours after birth. For female babies, as all had rectovestibular fistulas, parents were counselled for home care with regular followup, and pelvic-ddivided colostomy was performed once the baby started getting constipated at the age of around 3-4 months. Subsequently, we subjected both male and female babies to anoplasty when deemed appropriate, taking into account their weight and comorbidities, followed by stoma closure once we achieved the target anal dilatation for their age. All babies were operated on under general anesthesia. A specially designed proforma was prepared in which complications associated with each stage of treatment were documented, such as stoma prolapse, retraction, stenosis, wound dehiscence, and infection, as well as functional outcomes like soiling, incontinence, or constipation. The patients were discharged from the hospital once they were stable and started to tolerate oral feeding. After every stage of treatment, parents were counselled in detail, both verbally and in the form of written instructions regarding the home care of a stoma, a perineal wound, and the sequence of anal dilatation using Hegar`s dilator. All the patients were followed up initially at two weeks and then at regular intervals to evaluate for complications and progress.

The data was analysed using SPSS (Statistical Package for Social Sciences) version 20.0. For quantitative variables the mean and SD (standard deviation) were calculated. For qualitative variables like gender and procedure, frequency and percentage were calculated.

RESULTS

During the study period, a total of 24 patients were operated on. Out of 24 patients, 14(58.3%) were female, whereas 10(41.7%) were male patients. The patients' ages ranged from 1-4 years. All female patients had ARM and a rectovestibular fistula. Out of 10 male patients, two (20%) had rectal atresia, one had a recto-urethral bulbar fistula, one had a recto-urethral prostatic fistula, one had a recto-bladder neck fistula, and one had a perineal fistula.

After Stage I (Pelvic Divided Colostomy), 3 patients (12.5%) had a wound dehiscence, 2 patients (8.3%) had retraction or stenosis, and 2 patients (8.3%) had an infection (Table-I). One patient (4.1%) had a

prolapsed stoma. Four patients (8.3% of the total) who went through Stage II (PSARP) had wound dehiscence, two (8.3% of the total) had dehiscence of the perineal body, two (8.3% of the total) had wound infection, one (4.1%) had anal mucosal prolapse, two (8.3% of the total) developed anal stenosis, and one (4.1%) had severe urethral injury (Table-II). After Stage III (stoma closure), only 1(4.1%) patient developed a wound infection that was treated conservatively; however, no patient developed an anastomotic leak. Regarding the long-term follow-up, 2(8.3%) patients developed constipation, which was managed with a bowel management program(Table-III).

Table-I: Complications after Stage-1 (Pelvic Divided Colostomy) (n=24)

Complications	n(%)
Stoma stenosis / retraction	2(8.3)
Wound dehiscence	3(12.5)
Wound infection	2(8.3)
Stoma prolapsed	1(4.1)

Table-II: Complications After Stage-2 (PSARP) (n=24)

Complications	n(%)
Wound infection	2(8.3)
Wound dehiscence	2(8.3)
Dehiscence of perineal body	2(8.3)
Anal mucosal prolapsed	1(4.1)
Anal Stenosis	2(8.3)
Severe urethral injury	1(4.1)

Table-III: Complications After Stage-3 (Stoma Closure) (n=24)

Complications	n(%)
Wound infection	1(4.1%)
Anastomotic leak	Nil

DISCUSSION

ARM is a very common anomaly, appearing frequently in pediatric surgery clinics. Treatment of ARM has social, psychological, financial, and ethical implications. There has always been a debate about how to treat such patients, which includes primary versus staged repair. A study looked at how a one-stage approach worked for kids with ARM who had a lot of problems. They found that primary repair is better than multiple surgeries and psychological effects, but it needs experience and careful patient selection to avoid the bad results that come with poor functional outcomes because the sphincter muscle complex isn't fully developed.[11]

Similarly, in another study, the data of 12 male patients who underwent primary surgery showed promising results, with only two patients having surgical site infections and one having pseudo-incontinence, but at the same time concluded that the primary approach is suitable with careful selection of patients in experienced hands.[12] In this study, we adopted a staged approach with all patients and assessed the complications and outcomes associated with each procedure.

These complications are also reported in the literature, such as a study that analyzed the complications of stoma in 138 patients, out of which 38 (27.5%) had complications like prolapse, retraction, stenosis, and skin excoriation.[13] The results are comparable to our study. Another study published the data of 116 children with colostomies, out of which 62 (53%) suffered complications like wound infection, dermatitis, and prolapse.[14] Our results surpass those of this study, particularly in the area of parastomal skin excoriation, likely due to improved parental education through the provision of written handouts for stoma care at home. PSARP is the second procedure in the ARM management process. In our study, we found that 10 patients (41%) experienced complications such as superficial wound infection, wound dehiscence, superficial dehiscence of the perineal body, rectal mucosal prolapse, anal stenosis, and urethral injury. Studies have mentioned such complications after PSARP, like when Tofft et al. analysed the results of 90 children who underwent PSARP in a primary and secondary approach. Multiple factors led to complications, such as wound infection and wound dehiscence, as well as the absence of a stoma in primary repair and associated anomalies.[15] Similarly, another study conducted by Elekiabi *et al*. analysed the outcome of 49 patients who underwent PSARP in a staged approach and found that 20.5% of the patients had complications like wound dehiscence.[16] In our study, 6(25%) patients developed wound infections, leading to wound dehiscence, so our results are comparable to the study mentioned.

Another study by Uzair *et al*. shared the results of PSARP in 40 female patients, in which 5 patients had wound infection, 5 sustained injury to the posterior vaginal wall, and 1 had wound dehiscence.[17] Our findings are superior in terms of injury to the vaginal wall, as out of a total 14 female patients, none sustained this injury. Another study conducted by Nkoworo *et al.* documented the results of 60 patients with ARM and concluded that 16% of patients suffered complications associated with procedures at different stages, whereas 10 patients died mainly due to associated anomalies.[18]

In our study, no patient had an anastomotic leak, and only 1(4.1%) patient had a superficial wound infection that was managed conservatively. The literature documents complications following stoma closure. A study had a large number of patients, i.e., 2110, who underwent stoma closure, and 7.6% developed surgical site infections that were attributed to longer operation times.[19] Our results are superior to that, as only 1 out of 24 patients developed a wound infection, but obviously the difference in sample size is not comparable. Another study examined the results of 56 children who underwent stoma closure. Out of these, 7.1% had an anastomotic leak and 12.6% had a wound infection, whereas 1 child died.[20] Our results are better in terms of mortality, as none died, and also in terms of anastomotic leak, as no patient suffered this complication.

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CONCLUSION

Anorectal malformations management is a real challenge, and it demands continuous refinement in surgical techniques to minimize the complications associated with different procedures in a staged approach. On the other hand, with better facilities and experience, we should gradually shift the mode of management from a staged approach to the primary approach in selected cases, with better post-operative care and the availability of parenteral nutrition.

Conflict of Interest: None.

Authors' Contribution

Following authors have made substantial contributions to the

HR & GA: Data acquisition, data analysis, data interpretation, critical review, approval of the final version to be published.

AM & JR: Study design, data interpretation, drafting the manuscript, critical review, approval of the final version to be published.

NA & GF: Conception, data acquisition, drafting the manuscript, approval of the final version to be published.

Authors agree to be accountable for all aspects of the work in ensuring that questions related to the accuracy or integrity of any part of the work are appropriately investigated and resolved.

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