

Two-stage repair of low anorectal malformations in girls: is it truly a setback?

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Background/purpose Anorectal malformations (ARMs) affect 1 in 4000–5000 births. Low ARMs are nowadays treated in the first stage rather than at second or third stages. However, reports suggest problems with continence in these children because of wound dehiscence and infection; thus, protective colostomy may still be recommended. Colostomies do have complications, but the question is whether these disadvantages outweigh the protective effect on wound healing after anal reconstruction. The aim of this study was to define whether two-stage repair of low ARMs in girls is truly a setback or whether it is beneficial.

Patients and methods During the period of June 2008–June 2012, 30 female patients suffering from low ARMs were admitted to Mansoura University Children Hospital. Their ages at the time of surgery ranged from 3 to 11 months (mean age 6.2) and they were divided into two equal groups. The fistula location was defined either anocutaneous or anovestibular according to the Pena classification. The choice of management was totally randomized; thus, patients of group A underwent a two-stage posterior sagittal anorectoplasty and group B patients underwent a one-stage posterior sagittal anorectoplasty operation. Data recorded included age, fistula location, associated anomalies, operation performed, operative time, length of hospital stay, approximate cost, and postoperative complications.

Results A comparison of data showed that treatment of patients of group A involved more time and money and they had a longer duration of hospital stay than did patients of group B. Seven patients (47%) in group A and nine patients (60%) in group B showed postoperative

complications. Wound infection occurred in three patients (20%) of group A and in eight patients (53%) of group B. More importantly, two (13%) wound disruptions occurred among the three cases with wound infection in group A, whereas six (40%) disruptions occurred among the eight patients (53%) with wound infections in group B. The incidence of redo operation in group B was found to be significantly higher than in group A. Mucosal prolapse occurred in only one patient (7%) of group B. Complications related to colostomy occurred in group A only; five patients (33%) suffered skin excoriation around the stoma and one patient (7%) showed a prolapsed distal stoma loop. Constipation was noted during follow-up in five patients (33%) of group A and in six patients (40%) of group B.

Conclusion Two-stage repair of low ARM in girls is truly beneficial, as we could perform a successful operation and achieve continence in the child regardless of the complications of colostomy, which are temporary and tolerable. *Ann Pediatr Surg* 9:69–73 © 2013 Annals of Pediatric Surgery.

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Introduction

Anorectal malformations (ARMs) affect 1 in 4000–5000 births. The embryologic pathology is related to dysmorphogenesis of the cloaca and urorectum in early fetal life. The anomaly may occur in isolation but is commonly associated with other anomalies – urogenital and musculoskeletal abnormalities being the most common [1,2].

Consistent with incomplete separation of the cloaca, most patients have a fistula. In male and female patients, rectourethral fistula and rectovestibular fistula, respectively, are the most frequent variants. ARM without a fistula is uncommon and found only in 5% of cases [3].

The earliest classification of ARM was based on the position of the terminal rectum in relation to the levator ani or pelvic floor [4]. Termination of the rectum above the levator ani is termed 'high', and termination below the levator ani is termed 'low'. The relationship of the terminal rectum to the levator ani formed the basis of the

1984 Wingspread classification [5,6]. A classification based on the presence and position of the fistula was described by Pena in 1995 as a result of his experience with the posterior sagittal anorectoplasty (PSARP) [7,8].

Pena [3] introduced the PSARP for infants and children with ARM. They advocated performing a colostomy in all cases of ARM to prevent wound infection and thus prevent harm to the anal sphincter complex after reconstruction.

In general, colostomies performed during repair of ARM must be completely diverting [9]. Besides the extra operation that is needed for colostomy closure (as well as for creating one), there are complications such as prolapse, skin excoriation, and the burden for parents in dealing with an enterostomy. The question is whether these disadvantages outweigh the protective effect of a colostomy on wound healing after anal reconstruction [10].

It is reported that the majority of girls with an imperforate anus will have a lesion of the low variety. Treatment can be accomplished using a variety of techniques that can be safely performed without a diverting colostomy [11]. Although children with a low imperforate anus have been considered to show good results, several reports suggest problems with continence in these children [12]. Wound dehiscence and infection compromise the final functional result and make complications unacceptable. This is because the complexity of this defect is frequently underestimated; thus, every effort should be made to give these patients the best opportunity to have a successful reconstruction with a single operation. Protective colostomy is therefore strongly recommended [12,13].

Materials and methods

Thirty female patients admitted to Mansoura University Children Hospital during the period June 2008–June 2012 suffering from low ARM were included in this study. Their ages at the time of surgery ranged from 3 to 11 months (mean age 6.2). They were divided into two equal groups. Group A comprised 15 patients who were treated by two-stage PSARP with colostomy and group B comprised 15 patients treated by one-stage PSARP without colostomy.

All the patients were female with low ARMs. The fistula location was defined as being either anocutaneous (Fig. 1) or anovestibular (Fig. 2) according to the Pena classification. They were screened for associated anomalies such as VACTERL (Vertebral, Cardiac, Tracheo-Esophageal, Renal, Limb malformations). Routine preoperative laboratory investigations were performed for all patients.

Total bowel irrigation was performed for all the patients using saline instead of polyethylene glycol as it is not available at our institute. Preoperative bowel sterilization was performed using oral neomycin and metronidazole 48 h before the operation. Intraoperative use of third-generation cephalosporins and metronidazole infusion was continued for 3 days postoperatively for all patients. Nothing per oral and parenteral nutrition were started 1 day preoperatively and continued for 2 days postoperatively for all patients.

The choice of management was totally randomized. Thus, patients of group A underwent a two-stage PSARP: the first stage was a PSARP operation with a right transverse divided loop completely diverting colostomy (Fig. 3) and the second stage was a closure of colostomy 3–4 weeks later. Group B patients underwent a one-stage operation (Fig. 4). Only three patients showed a dilated colon during PSARP, which needed tapering.

All children had postoperative anal dilatations as per the Peña scheme. Data recorded included age, fistula location, associated anomalies, operation performed, operative time, length of hospital stay, approximate cost, and postoperative complications.

Statistical analysis was performed using the Pearson χ^2 and Fisher exact tests for nonparametric data and the *t*-test for equality for parametric data.

Fig. 1



Anocutaneous fistula.

Fig. 2



Anovestibular fistula.

Results

Group A included 15 female patients with low ARM: seven patients (47%) had ARM with anocutaneous fistula and eight (53%) showed anovestibular fistula. Their ages at the time of first-stage surgery ranged from 3 months to 11 months (mean age 6.4). Two patients (13%) had associated anomalies: one showed an absent kidney and the other had a minor cardiac anomaly. The operative time of this group was calculated for each patient by adding the operative times of both stages; it ranged from 140 to 180 min (mean time 160). The total length of hospital stay for both stages together ranged from 8 to 10 days (mean 8.5). The total approximate cost for both stages for each patient ranged from 1600 to 1800 LE (mean cost 1700).

Group B included 15 female patients with low ARMs: eight (53%) were ARMs with anocutaneous fistula and seven (47%) showed anovestibular fistula. Their ages

Fig. 3



Right transverse divided loop colostomy.

Fig. 4



Posterior sagittal anorectoplasty operation.

at the time of surgery ranged from 3 months to 11 months (mean age 6). Two patients (13%) had a minor cardiac anomaly. The operative time of this group was calculated for each patient and ranged from 60 to 120 min (mean

time 87). The total length of hospital stay ranged from 5 to 8 days (mean 5.5). The total approximate cost for both stages for each patient ranged from 800 to 1000 LE (mean cost 870).

On comparing the data of both groups, a noticeable variation was found between the two groups: operative time was 160 and 87 min, hospital stay was 8.5 and 5.5 days, and approximate total cost was 1700 and 870 LE, respectively. These data suggest that treatment of patients of group A involved more time and money and they experienced a longer hospital stay compared with group B patients. A statistical significance was found for all three items (Table 1).

Sixteen (53%) patients suffered from 41 postoperative complications: seven (47%) of group A had 17 postoperative complications, whereas nine (60%) of group B had 24 postoperative complications, which indicates that more complications occurred in patients undergoing one-stage PSARP (Table 2).

Wound infection occurred in three patients (20%) of group A and in eight patients (53%) of group B, showing a marked increase in the latter. More importantly, two (13%) patients among the three with wound infections in group A developed wound disruptions; however, they healed conservatively and did not need a redo. The third patient turned out to have anal stenosis, which may not be related to the infection and was managed by regular dilatation. However, six (40%) patients among the eight with wound infections in group B developed wound disruptions (Fig. 5). All of them resulted in a short perineum and three (20%) of these patients developed anal stenosis. The wound disruption in these patients occurred 7–10 days postoperatively and a completely diverting right transverse divided loop colostomy was performed 10–15 days postoperatively when disruption was found to be progressive. They all needed a redo PSARP.

The incidence of redo operation in group B was found to be significantly higher than that in group A in which no redo was needed. Mucosal prolapse occurred in only one patient (7%) of group B and it needed a minimal operation to remedy the condition. Complications related to colostomy occurred in group A only; five patients (33%) suffered skin excoriation around the stoma and one patient (7%) showed a prolapsed distal stoma loop. They were managed conservatively until the stomas were closed.

Constipation was noted during follow-up in five patients (33%) of group A and in six patients (40%) of group B and they were all managed conservatively and responded well.

Discussion

We believe that the most important decision to be taken for a baby with an ARM is the creation of a colostomy as part of the treatment plan. This is an easy decision if the ARM is intermediate or high; however, the performance of colostomy in low anomalies is a decision disputed by many [14].

Table 1 Demographic data

	Group A	Group B	Significance
Age	6.4 months	6 months	0.599
Operative time	160 min	87 min	0.000
Hospital stay	8.5 days	5.5 days	0.000
Approximate cost	1700 LE	870 LE	0.000
Associated anomalies	2 cases	2 cases	1.000

Table 2 Postoperative complications

	N (%)		Significance
	Group A	Group B	
Wound infection	3 (20)	8 (53)	0.128
Wound disruption	2 (13)	6 (40)	0.215
Anal stenosis	1 (7)	3 (20)	0.598
Constipation	5 (33)	6 (40)	1.000
Skin excoriation	5 (33)	–	–
Prolapsed stoma	1 (7)	–	1.000
Mucosal prolapse	–	1 (7)	–
Total number of complications	17	24	–
Total number of cases	7 (47)	9 (60)	–

Fig. 5

Wound disruption.

Pena [15] insisted at performing a covering colostomy in cases of vestibular fistula. Then, in 1993 he proposed avoiding colostomy in these patients by giving the infants a low-residue diet for 1 week preoperatively and keeping the bowel as empty as possible. In the immediate preoperative period, the colon is thoroughly washed through the fistula to keep the wound as clear as possible in the postoperative period [8].

Low ARMs in girls appear simple but are actually complex anomalies and often underestimated by the treating physician, thus corrected without a covering colostomy or proper preoperative preparation that may result in disruption of the whole repair. The main aim of

managing these anomalies is to achieve continence in the child, which remains the real challenge in pediatric surgical practice [1].

In our study we compared two-stage PSARP with colostomy to one stage in girls with low ARMs, aiming to define whether colostomy is of benefit or a setback. Total bowel irrigation was performed for all the patients; nothing per oral and parenteral nutrition were started 1 day preoperatively and continued 2 days postoperatively for all patients. We used this strategy so that group B patients would suffer no disadvantage if patients were prepared by rectal irrigation only.

Our results show that fewer complications occurred in group A than in group B, especially with regard to wound infection and wound disruption; however, no statistical significance was noted. This is because the number of patients is not enough for sufficient statistics. Redo PSARP with a covering colostomy was needed in six patients (40%) of group B who had suffered wound disruptions; however, none of the two patients (13%) with wound disruptions in group A needed a redo as they healed conservatively. This was found to be statistically significant.

Treatment of group B patients involved less time and money and they experienced a shorter hospital stay compared with group A and a statistical significance was found for all three items. They were also spared the complications of colostomy. However, if we consider the fact that six patients (40%) of group B needed a redo two-stage PSARP, we feel that the advantages of one-stage operation are over-rated. We also believe that the complications of colostomy are temporary and tolerable if we can achieve a sound operation and a continent child.

Performing an operation without a colostomy most of the time works as demonstrated by most, but sometimes it does not. If a patient with a perineal fistula has a dehiscence, it is not so relevant; however, for a patient with a vestibular fistula, it is a serious problem, and sometimes it compromises bowel control [16].

We know that the tendency is to perform operations without a colostomy, and we believe that the decision is based on personal experience.

Bowel control in perineal and vestibular fistula patients should be 100%, but they may soil once in a while; most of that soiling is caused by constipation. When you treat the constipation adequately, the soiling disappears, suggesting that they suffer from overflow pseudocontinence. The patients may experience what is called hidden constipation [17]. In our study we had 11 patients (37%) suffering from constipation: five in group A and six in group B, all of whom were managed conservatively and responded well. However, toilet training is a tedious procedure in cases of ARM, even in low types.

Conclusion

Low ARMs in girls are actually complex anomalies that, when corrected without a covering colostomy, may result in disruption of the whole repair. The main aim of managing these anomalies is to achieve continence in the

child, which remains the real challenge in pediatric surgical practice. The performance of colostomy in low anomalies is a decision disputed by many but we believe that two-stage repair of low ARMs in girls is truly beneficial, as we could achieve a sound operation and a continent child regardless of complications of colostomy, which are temporary and tolerable.

Acknowledgements

Conflicts of interest

There are no conflicts of interest.

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