



One-stage vs. three-stage repair in anorectal malformation with rectovestibular fistula

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ABSTRACT

Background: Anorectal malformations (ARMs) disease is one of the congenital anomalies with an incidence of about 1 in 5000 neonate births, and treatment requires surgical intervention. Selecting the one- or three-step surgical procedure to treat the disease, especially in female neonates with rectovestibular fistula, is a subject of debate. This study aims to compare the advantages and disadvantages of these two methods. **Materials and Methods:** Forty female neonates with ARM and rectovestibular fistula between March 2011 and March 2013 were included in the study, and they were divided into two equal groups. Allocation of the first case was random, and all cases were then allocated alternatively (every other subject was assigned to a treatment group) until each group received 20 cases equally patients of study group underwent a one-stage posterior sagittal anorectoplasty (PSARP) and in control group patients underwent a three-stage operation (colostomy, PSARP, and closure of colostomy). The complications during and after the surgery were recorded in both groups, and the results were compared. **Results:** In the control group, only one case (5%) of wound infection and dehiscence was seen, whereas in the one-stage study group, six cases (30%) of wound infection and dehiscence were seen (P value = 0.046). However, regarding the incidence of other complications, such as iatrogenic vaginal injury as well as final recovery, no considerable differences were seen between the two groups. **Conclusions:** Despite more surgical site infections and dehiscence in the one-stage repair, but due to the numerous advantages compared to the three-stage method, which is more time-consuming, more costly, and causes more adverse effect on parents and children, performing the one-stage repair is recommended for this anomaly.

Key words: Anorectal malformation, female neonate, one-stage repair, rectovestibular fistula, three-stage repair

INTRODUCTION

Anorectal malformations (ARMs) are birth defects in which the anus is absent or malformed. Its incidence occurs in 1 in 5000 births and affects boys and girls equally. ARMs are a spectrum of different congenital anomalies that vary from fairly minor lesions to a complex anomaly.^[1] Different surgeons use different terminologies when referring to types of ARMs. The clearest fact is that there is a spectrum of defects, so every attempt to classify them is arbitrary and somewhat inaccurate. Consequently, the traditional classification of 'high', 'intermediate' and 'low' defects renders the results ambiguous or uncertain.^[2] Results have however shown that it appears to be present as a low version 90% of the time in females and 50% of the time in males. ARM usually requires immediate surgery to open a passage for feces, unless a fistula can be relied on, or until corrective surgery takes place. Depending on the severity of the anomaly, it is treated either with perineal anoplasty alone or with colostomy in the first stage and a definite repair later.^[3]

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The most common anomaly in newborn girls is a rectovestibular fistula. Perineal inspection reveals a typical urethra, typical vagina, and another orifice, which is the rectal fistula in the vestibule. In newborns with clinical evidence of a rectovestibular fistula, a diverting colostomy is the safest option for surgeons who do not have extensive experience in anorectal anomalies. Although colostomy prior to the main repair avoids infection complications and dehiscence,^[4] colostomy creation in neonates is an invasive procedure. Apart from the challenges of post-operative care, a colostomy is associated with many complications, such as skin excoriations, wound infection, bleeding, sepsis, prolapse, stricture, fluid and electrolytes losses, which are poorly tolerated by young children. These challenges contribute to the poor acceptance by parents/caregivers, especially in developing countries. One-stage posterior sagittal anorectoplasty (PSARP) is a definitive repair that can be carried out in neonates without prior colostomy creation. The virtually sterile meconium during the 1st week of life reduces the risk of infection from fecal contamination. Many centres in developed countries have recorded multiple successes with primary PSARP in neonates.^[5] Another option reported is two-stage repair, PSARP plus colostomy that is performed simultaneously as the first stage and a colostomy closure as the second stage that protects the wound from contamination and omits one additional stage. This method not only has the advantage of fecal diversion and decreases the risk of infection but also has the disadvantage of a 2-step operation and risk of colostomy-related complications, (mentioned in this discussion).

The purpose of this study is to evaluate the feasibility and efficacy of the one-stage repair in the treatment of female neonates with a rectovestibular fistula and its comparison with the three-stage conventional method. Assessment of two-stage repair is not the aim of our study because it has not any technical advantage compared to one-stage repair.

MATERIALS AND METHOD

This study was performed between March 2011 and March 2013 in the Kermanshah University of Medical Sciences Pediatric Surgery Center (Mohammad Kermanshahi and Emamreza Hospitals) that are referral centres for paediatric surgery for the Middle-west of Iran. As a randomized, single-blind clinical trial, 40 female patients with ARM and rectovestibular fistula were included in the study. Allocation of the first case was random, and all cases were then allocated alternately (every other subject was assigned to a treatment

group) until each group received 20 cases equally. One group with 20 patients underwent the three-stage procedure (control group), whereas the other group with 20 patients was treated by the one-stage method (study group). All female neonates with rectovestibular fistula were included in the study and all male patients and females with another type of malformation (Cloacal anomalies, without fistula, perineal fistula, etc.) were excluded from the study. Only one case of rectovestibular fistula was excluded from the study group due to obstructive symptoms (impaction and obstruction of a narrow fistula tract by meconium that did not respond to bougienage and dilatation).

The presence of other possible accompanying anomalies was determined by echocardiography and abdominal sonography. All cases presented in the first few days of life except two cases that came later (one in 20 days and one after 3 months; both had effective defecation via a vestibular fistula). All surgeries were performed by one paediatric surgeon with 11 years' experience in paediatric surgery and 20 years' experience in general surgery. The parents of all children in the study signed an informed consent form that contained the necessary information. The study was approved by the Ethics Committee of the University and informed consent forms were obtained from all parents.

In the control group (three-stage repair), the neonates underwent a double-barrel sigmoid colostomy in the first few days after birth. Then, the patient was discharged, and after about 6 weeks, the PSARP was performed. At this stage, the patient was placed in a prone jackknife position and the pelvis elevated. The exact location of the anus and anal muscles complex was determined using electrical stimulation. The skin and subcutaneous tissue of the perineum was incised in the midline from the tip of the coccyx to near and around the fistula. Next, the muscles, rectum, and vagina were separated. First, the posterior wall and then the lateral sides were freed, and finally, the anterior rectal wall attached to the vagina was separated with caution. The anus and rectum were placed in their true and correct positions and fixed there with sutures. Then, a perineal and incisional line repair completed the operation and the procedure.

Administration of intravenous antibiotics continued for 48 h and then the treatment was changed to oral antibiotics. Oral feeding was immediately permitted due to the presence of fecal diversion by colostomy that protected the surgical incisional site from contamination and dehiscence. Anal bougienage will

be done 2 weeks after anorectoplasty and is continued daily by the patient's parents. When the anorectoplasty heals completely, about 4-6 weeks after PSARP, the colostomy can be closed as the third stage (when the patient is between the ages of 10 and 12 weeks. Thus, the patient will be hospitalized for the 3rd time and will undergo the colostomy closure operation.

In the study group (one-stage repair), all neonates were permitted to receive oral feeding and could defecate via a vestibular fistula. After 4-6 weeks, when the infant's weight reached about 4-5 kg, then it was then better positioned to undergo PSARP. This delay improved technical problems, especially vaginal separation from the rectum that is most challenging in this operation. In fact, we performed the operation in a one-stage repair, which took place 1-1½ months after birth in order to make time for the infant's weight gain and growth, greater ease of operation regarding technical issues and reducing the risk of trauma to the vagina. In the one-stage repair, all cases were not permitted to receive oral feeding for 5 days post-operation and total parenteral nutrition was started during the 1st post-operation day and continued for a few days until oral feeding resumed. This was because there was not diverting colostomy and this can increase the risk of surgical site infection (SSI) by fecal material after oral feeding.

It should be noted that in the event of a very narrow fistula and the inability to achieve normal defecation via a vestibular fistula, should obstructive symptoms occur, first, the fistula dilatation by bougienage is performed. If the obstructive symptoms persist, and if the obstruction does not improve, then the neonate immediately undergoes a colostomy and is excluded from the one-stage group (Only one case was excluded from the study for this reason).

All cases in both groups were kept under close observation during the surgery and in the post-operative period in the hospital, as well as during intermittent outpatient visits in the clinic. The participants in the study were followed up for several months, (the follow-up period was different between 11 and 35 months after completion of the treatments). Then the final results and complications were compared between the two groups. The complications, including SSI and anorectoplasty site dehiscence, anal stenosis, rectal prolapse, rectovaginal fistula and in the control group, complications of colostomy such as prolapse, stenosis, or dehiscence of the colostomy site were also collated. All data that was obtained was collected using a checklist designed to conduct the study. Finally, the data

analysis was done using SPSS 16.0 statistical .based on classification of the SSI. One-stage PSARP, because of direct contact with fecal material, is a contaminated wound with an expected infection rate above 27% and 3-stage PSARP, because of fecal diversion is a clean wound with an expected infection rate of about 1-5%.^[6] Such an expected big difference in the complication rate between two groups can make this sample size statistically reliable. Although more cases make the study more reliable, due to the rarity of this anomaly and limitations of our time and facilities, our sample size consisted of 40 total cases.

Data processing and analysis

The SPSS 16 software was used in this study to analyse the data. To summarize the data and provide descriptive reports, the one- and two-dimensional tables, mean, frequency percentage, and bar graphs were used. To compare the surgery results and post-operative complications in the two studied groups, the analyses of agreement tables and the Mantel-Haenszel chi-square test were used. The significance level was considered as 0.05 in this study.

RESULTS

The mean age and weight of the two groups at the beginning of this study were the same because all patients were in the first few days of life when entered into the study and control group. However, by the termination of the treatment period, there were considerable differences between the mean age and weight in both groups, because 3-stage repair needs more time and children become bigger with more age and weight. The mean age of patients was 4.25 ± 2.06 months. The mean age in the study group (one-stage repair) was 3.15 ± 2.11 months, and the mean age of the control group (three-stage repair) was 5.35 ± 1.3 months at the end point of the treatment period. The mean age values of the two studied groups were statistically significant (P value < 0.001) [Figure 1].

The average weight of the patients was 5087 ± 1229 g. The average weight of the one-stage group was 4600 ± 1130 g, whereas the average weight of the three-stage group was 5575 ± 1150 g. The two groups showed significant differences regarding weighting average (P value < 0.01) [Figure 2].

We had no mortalities in either group. In the control group, only one case of wound infection (superficial incisional infection) was seen (5%), where the individual improved rapidly with conservative management,

whereas, in the one-stage group, 6 cases (30%) of wound infection and dehiscence were seen. From those cases, four individuals had superficial SSI and improved by having only conservative management without any considerable adverse sequel and two cases had deep SSI (First case [5%] had severe dehiscence with complete opening of the wound that resulted in deformity and scar formation that finally resulted in anal displacement. That individual underwent reoperation and the situation was corrected. In the second case, a secondary colostomy became necessary to reduce infection. In the third case, due to low birth weight, vaginal and rectal stricture was caused, which was improved by regular dilatation alone. Mild rectal prolapse was detected in 5 cases in the control group and 3 cases in the study group. Only one case of rectal prolapse in the control group underwent surgical resection and was corrected. The remaining cases were mild and did not need any treatment. Of 20 patients who were in the control group, five cases had associated anomalies and 6 cases in the study group had an associated anomaly also. We had one colostomy site stenosis and one case of colostomy prolapse in the control group that needed a revision of the colostomy, and one case of a colostomy site infection and dehiscence that improved with conservative treatment and a few cases of peristomal skin excoriation that was treated with topical zinc oxide ointment and all of them improved after colostomy closure. Although assessment for fecal continence in small children is not completely feasible but some criteria can be used based on clinical exam. There are various methods of quantitative and qualitative assessment for the degree of fecal continence after anorectoplasty. Wingspread, McGill, Kiesswetter and Kelly's criteria that is the simplest of all scoring systems and is usable even in a 3-month infant. The results were judged on the basis of staining, accidental defecation, and strength of sphincter squeeze on per-rectal examination.^[7] Based on clinical assessment, we had no pattern of incontinency in two group. The rate of constipation was the same in both groups (3 cases in the control group and 4 cases in the study group) [Figure 3].

The two surgical methods were significantly different regarding SSIs and dehiscence (*P* value < 0.046). In terms of anal displacement and deformity and iatrogenic vaginal trauma and fistula, the two groups showed no significant differences (*P* value > 0.05) [Table 1].

DISCUSSION AND CONCLUSION

ARMs disease is one of the congenital anomalies with an incidence of about 1 in 5000 in neonates. The most

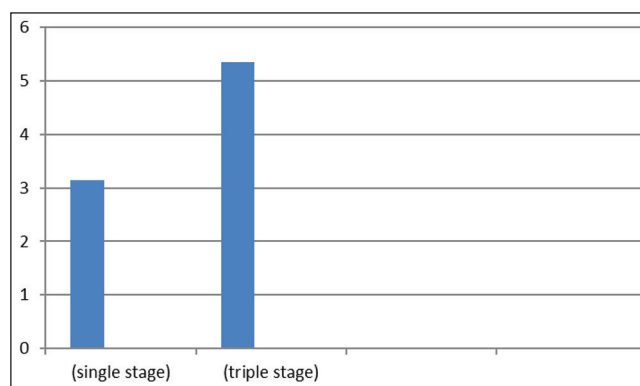


Figure 1: Mean age in months in the two groups. (After completion of treatment)

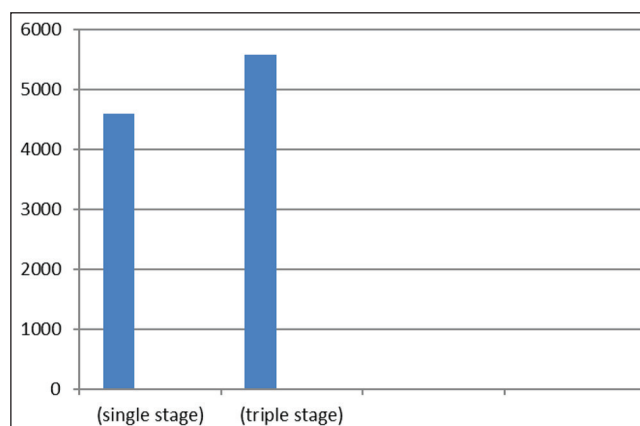


Figure 2: Mean weight in the two groups. (After completion of the treatment) (gr)

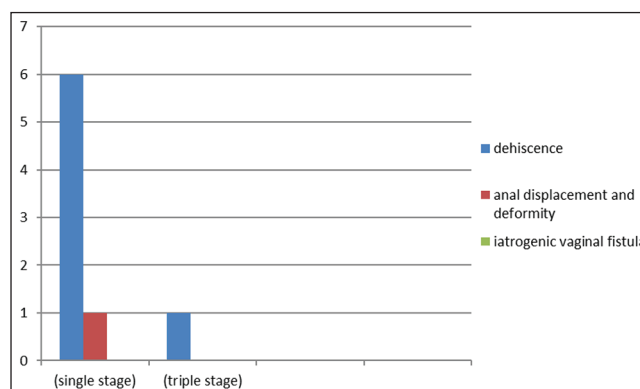


Figure 3: Comparison of surgical complications in two groups

common form of this anomaly in female patients is rectovestibular fistula. Selecting the one-step or three-step surgical procedure to treat this anomaly has been a subject of debate for many years. Reasons for choosing the 3-stage repair are the lesser risk of a SSI because of fecal diversion by colostomy. It might be an easier surgical technique because of the delay in definite repair and weight gain by the infant. On the other hand, the reasons for choosing one-stage repair is multiple, for example, avoidance of multistage operations and saving

Table 1: Major complications seen in the two groups

Type of complication	Type of surgery	Number of observed complications (%)	P
Anorectoplasty site infection and dehiscence	One stage	6 (30)	0.046
	Three stage	1 (5)	
Anal displacement and deformity	One stage	1 (5)	1.000
	Three stage	0	
Iatrogenic rectovaginal fistula	One stage	0	0
	Three stage	0	

time and costs, less stress and insult for children and their parents, less psychosomatic trauma for children, avoidance of a colostomy related complications and risk of an adhesion band in the future because of an abdominal opening. Nagdeve *et al.* showed one-stage repair on male neonates with high anomaly with good results.^[8] Osifo *et al.* showed that one-stage repair is feasible in all ARMs including low or high variation and with meticulous selection, primary PSARP in neonates is safe.^[5] Elsaied *et al.* in Egypt showed that two-stage repair of low ARM in girls is truly beneficial. They could perform a successful operation and achieve continence in their child.^[9] In our study, the rate of SSIs and dehiscence was high in the study group (30%), and this was a negative aspect of one-stage repair. This high rate of infection is due to the absence of a colostomy as a fecal diversion route and direct contact between the surgical site and fecal material immediately after surgery that makes the suture line contaminated. By definition, SSI is further defined as superficial or deep. Superficial incisional SSI – infection involves only skin and subcutaneous tissue of incision. Deep incisional SSI – Infection involves deep tissues, such as facial and muscle layers. Organ/space SSI – infection involves any part of the anatomy in organs and spaces other than the incision, which was opened or manipulated during the operation. According to this classification, most of wound infections in our study group (4 patients) were superficial incisional site infection and self-limited. Considering all of these, makes one-stage repair more logical and ethical and less critical. The only clinical scenario that we would advise diversion is a neonate who cannot defecate via fistula because of a very thin and narrow fistula tract that does not respond to dilatation and bougienage and has obstructive symptoms and we advise one-stage repair for all cases of rectovestibular fistula except primary disability of sufficient defecation via vestibular fistula and obstructive symptoms. The best time for one-stage repair is 4-6 weeks after birth because weight gaining in this delayed interval makes surgery easier and safer technically. Our follow-up period was between 1 and 3 years, and in this age group, assessment of fecal continence by classic methods, such

as wingspread, Kelly, or via the Kiewewetter scoring system, was not completely feasible. However, some of those criteria are performable and measurable in this age group. The pattern of incontinent defecation includes frequent defecation (10-20 times/24 h with permanent soiling), severe perianal skin excoriation and flaccid anal sphincter in a rectal exam are another sign of incontinent defecation. The normal pattern of defecation in children in the 1st month of life is about 6 times/day, and after 2 months, decreases to 1-2 times/day until the child is 2 years old.^[10] Anal sphincter has a squeezing effect during the rectal exam and there is not any considerable perianal skin excoriation. Soiling is not present, and the infant is clean between episodes of defecations. This pattern of defecation, though, is not definitely diagnostic but can be considerably prognostic for fecal continence in the future.

In a prospective study in 2002, Adeniran reported his results of a single-stage operation on imperforate anus patients with rectovestibular fistula. According to his results, initial studies showed that the repair of an imperforate anus with rectovestibular fistula can be safely performed as a one-stage anterior sagittal anorectoplasty, and the advantages of one-stage surgery were considered enormous compared to the three-stage surgery, particularly in developing countries.^[11] Another study in 2005 conducted by Gangopadhyay *et al.* in India on 105 patients, it was found that an initial one-stage surgery method had better apparent results and fecal control was associated with a reduced mortality rate and reduced cost and thus, is the recommended method.^[12] In a prospective study in 2006 conducted by Elhalaby in Egypt on 38 patients, the primary repair of high and intermediate ARMs in infants was studied. The results indicated that the one-stage repair of this type of abnormality in male and female patients was technically feasible. The safety of the approach depends on the omission of special cases of the study. The early post-operative complications in this method are acceptable and its functional results are comparable with the conventional multi-stage procedure.^[13] In another prospective study (2010) conducted by Kuijper and Aronson in the Netherlands on 35 patients, the conclusions indicated that the low-type ARM repair without colostomy with antibiotic therapy and long-term anal dilatation will have low morbidity and good results.

CONCLUSION

Given the considerable benefits mentioned for the one-stage surgical method and considering that most

of the one-stage repair complications were minor and resolved with conservative management, it seems that one-stage repair is a preferred and reliable method for treating the imperforate anus patients with rectovestibular fistula. However, further studies should be performed with a larger sample size and longer follow-up period for completely clarifying this issue.

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Conflicts of interest

There are no conflicts of interest.

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