



## THE OUTCOME OF TWO STAGE REPAIR IN ANORECTAL MALFORMATIONS WITH RECTOVESTIBULAR FISTULA.

Shafiq ur Rehman<sup>1</sup>, Yasir Makki<sup>2</sup>, Fareena Ishtiaq<sup>3</sup>, Saad Fazal<sup>4</sup>, Nauman Aziz<sup>5</sup>, Waleeja Shamikha<sup>6</sup>

1. MBBS, FCPS  
Assistant Professor Pediatric Surgery  
Sahiwal Medical College Sahiwal.
2. MBBS, FCPS  
Senior Registrar Pediatric Surgery  
Sahiwal Medical College Sahiwal.
3. MBBS  
WMO Basic Health Unit  
Dhakranwali Kharian.
4. MBBS  
Post Graduate Trainee  
SIMS/Services Hospital Lahore.
5. MBBS, M.Phil  
Demonstrator  
Sahiwal Medical College Sahiwal.
6. MBBS, FCPS  
Senior Registrar General Surgery  
Sahiwal Medical College Sahiwal.

### Correspondence Address:

Dr. Shafiq ur Rehman  
Pediatric Surgery  
Sahiwal Medical College Sahiwal.  
shafiqlangriyal@gmail.com

### Article received on:

17/10/2019

### Accepted for publication:

25/02/2020

**ABSTRACT...** Wound infection and dehiscence after recto vestibular fistula repair may affect the fecal continent mechanisms. A significant number of children with anorectal malformations have long term social, economic and psychological problems due to fecal incontinence. The role of protective colostomy should not be under estimated especially when you are treating the children from poor socioeconomic settings with compromised nutritional status. **Objectives:** The aim of this study was to evaluate the outcome of two stage limited posterior sagittal anorectoplasty with protective colostomy in female patients of congenital recto vestibular fistula. **Study Design:** Prospective study. **Settings:** Department of Pediatric Surgery, DHQ Teaching Hospital Sahiwal. **Period:** January 2016 to December 2018. **Material & Methods:** Thirty four girls with anorectal malformation and recto vestibular fistula underwent two stage anorectoplasty. Divided sigmoid colostomy and limited posterior sagittal anorectoplasty was performed in first stage. Six to eight weeks later stoma was closed in second stage. All the patients were evaluated for fecal continence, constipation, bowel function and complications of stoma formation, definite procedure and stoma closure. **Results:** The age distribution of patients at the time of surgery ranged from 9 months to 5 years (mean 27.32 months). During the first stage of repair, three patients (8.82 %) developed wound infection. Mucosal prolapse was seen in two patients (5.88%). Anal stenosis was noticed in one patient (2.94%). Seven patients (20.58%) developed peri stoma skin excoriation. Stoma prolapse was observed in three patients (8.82%). Four patients (11.76%) developed wound infection after colostomy closure. During the follow up period, constipation was reported in five patients (14.70%) and soiling in one patient (2.94%). **Conclusion:** Two stage correction of congenital recto vestibular fistula under the cover of colostomy is safe strategy. It is strongly recommended in a setup with limited resources and weak infrastructure. It is also useful for patients from poor socioeconomic settings.

**Key words:** Anorectal Malformation, Colostomy, Limited Posterior Sagittal Anorectoplasty, Rectovestibular Fistula, Two Stage Repair.

**Article Citation:** Shafiq ur Rehman, Makki Y, Ishtiaq F, Fazal S, Aziz N, Shamikha W. The outcome of two stage repair in anorectal malformations with rectovestibular fistula. Professional Med J 2020; 27(7):1527-1532.

DOI: 10.29309/TPMJ/2020.27.07.4257

## INTRODUCTION

Anorectal malformations, one of the most common congenital defects, present with wide spectrum of severity. The incidence of anorectal malformations is different in different geographical areas of the world.<sup>1</sup> Generally reported worldwide incidence is 2.0-2.5 per 10000 live births.<sup>2</sup> About 10% are recto vestibular fistulae.<sup>3</sup> Rectovestibular fistula is the most common form of anorectal malformation in female children.<sup>4</sup> A large number of procedures have been used for the repair of congenital recto vestibular fistula. However limited posterior

sagittal anorectoplasty innovated by Pena and anterior sagittal anorectoplasty introduced by Okada are most popular, well established and most commonly practiced techniques.<sup>5,6,7</sup> Aziz et al found no statistical significance between two techniques.<sup>8</sup> Conventional treatment of anorectal malformations was a three stage procedure. To reduce the duration from stoma formation to stoma closure and to minimize the colostomy related morbidity, two-stage repair was used. With the development of technology and advancement in medical sciences, new trends and modifications

have been introduced in the management of anorectal malformations. Laparoscopic assisted and image guided has been developed, which are less invasive and less damaging to muscles and nerves.<sup>9</sup> Advancements in anaesthesiology and neonatology has made it possible to perform primary neonatal anorectoplasty. However the management of anorectal malformations is still associated with significant morbidity, usually in the form of fecal incontinence. In spite of all advancements, modifications and new trends, the role of protective colostomy in the management of recto vestibular is still an unsettled issue. Two stage repair of recto vestibular fistula in girls under the cover of colostomy is truly beneficial.<sup>10</sup> Presence of protective colostomy has a significant effect on the outcome of the management of recto vestibular fistula.<sup>11</sup> Two stage repair of recto vestibular fistula is safer and more beneficial than one stage.<sup>12</sup>

Pediatric surgery is newly established department at DHQ Teaching Hospital Sahiwal. Resources are limited and infrastructure is in developing phase. Most of our patients come from under privileged areas with compromised nutritional status. The aim of our study is to evaluate the outcome of two stage limited posterior sagittal anorectoplasty with protective colostomy in patients of anorectal malformation with recto vestibular fistula, in our circumstances.

## OBJECTIVE

The objective of this study was to evaluate the outcome of two stage repair in anorectal malformations with rectovestibular fistula.

## MATERIAL & METHODS

### Settings and Design

This is a prospective study conducted in the Department of Pediatric Surgery, DHQ Teaching Hospital Sahiwal, during the period from January 2016 to December 2018. Study was approved by ethical review committee of Sahiwal Medical College and Allied Hospitals, Sahiwal.

### Inclusion Criteria

Girls with anorectal malformation and recto

vestibular fistula presented between nine months to five years age and managed previously with conservative measures.

### Exclusion Criteria

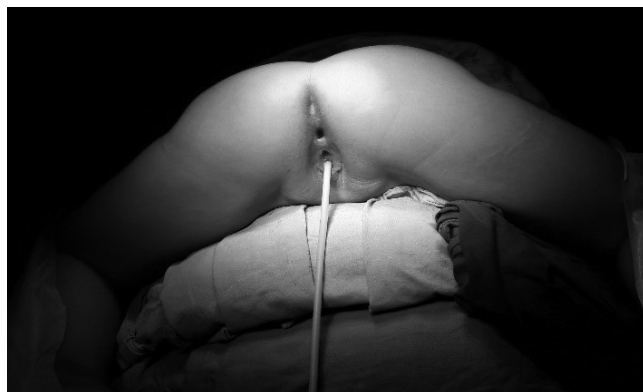
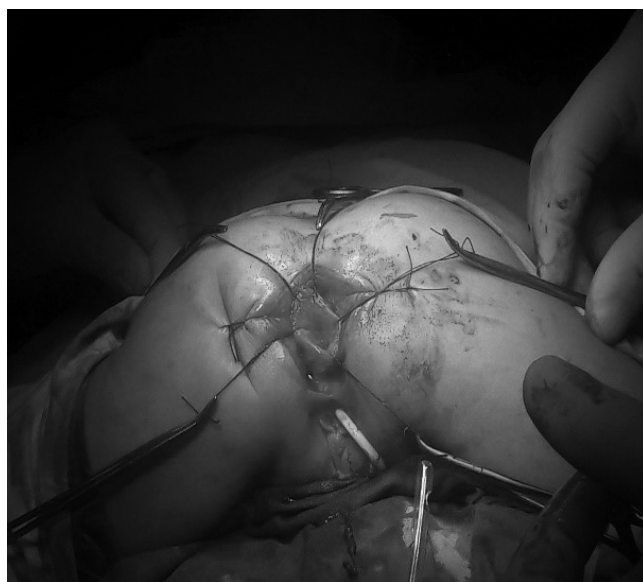
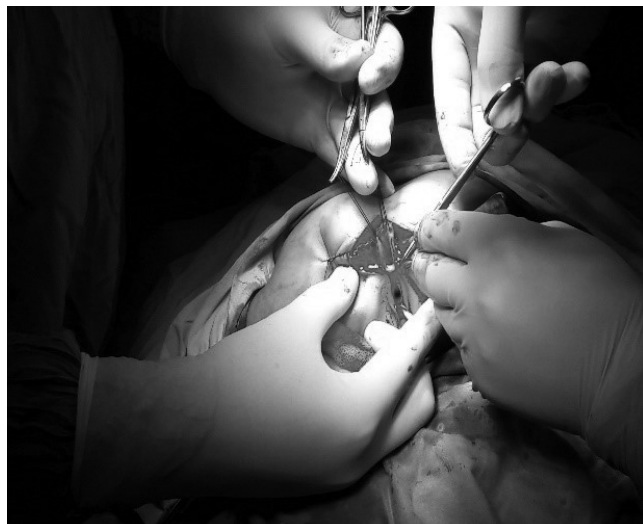
1. All male patients with anorectal malformation.
2. All female patients with another type of anorectal malformation.
3. Patients with major associated anomalies.
4. Patients presented in neonatal period and managed with diverting colostomy.

All patients were admitted through outdoor patient department, two days before operation. Pre-operative evaluation was done with medical history, routine clinical examination and lab investigations. Perineal examination was particularly performed in all cases to evaluate the vestibular orifices. Associated anomalies were assessed by abdominal sonography. Plain x-ray spine and echocardiography was advised only in selected cases. Bowel preparation was carried out in all patients. Rectal wash with normal saline, 20ml/kg/body weight, eight hourly, was started 48 hours before operation. Nothing per oral started one day preoperatively. Third generation cephalosporin and metronidazole infusion was started 24 hours before operation and continued postoperatively for two to three days. All patients underwent two stage limited posterior sagittal anorectoplasty. Divided sigmoid colostomy and definite procedure was performed in first stage. Six to eight weeks later stoma closure was done in second stage. The basic principles of posterior sagittal approach, as described by de Vries and Pena, were followed. Data collected included age, type of anorectal malformation, fistula location, hospital stay in first and second stage of procedure, duration from stoma formation to stoma closure and operation time. Immediate post-operative complications of stoma formation, limited posterior sagittal anorectoplasty, and stoma closure were noted. Stoma related morbidity like peri stoma excoriation, prolapse, stenosis etc. were also recorded. During the follow up period, all patients were evaluated for constipation and fecal incontinence according to Krickenbeck classification. Sphincter was assessed according to Kelly's clinical score.

Bowel habits (feeling of urge, capacity to report urge, ability to hold back) were assessed in children above three years of age. The patients were followed in outdoor patient department, weekly for one month, monthly for three months and then every three months till the child became toilet trained.

### Statistical Analysis

Statistical analysis was performed using SPSS. Quantitative data was shown as mean  $\pm$  standard deviation. Student t test was performed and bias was set at  $p < 0.05$  with confidence rate of 95%.



### RESULTS

The age distribution of patients at the time of surgery ranged from 9 months to 5 years (mean; 27.32 months). Duration of hospital stay for first stage procedure (colostomy+limited posterior

sagittal anorectoplasty) ranged from 4-6 days (mean; 5.20 days). Length of hospital stay for second stage procedure (colostomy closure) was 9-11 days (mean; 9.17 days). Duration with colostomy was 49-83 days (mean; 64.85 days). Operation time for first stage procedure was 95-

110 minutes (mean; 99.14 minutes) and for second stage was 45-60 minutes (mean; 52.97 minutes). During the first stage of repair, three patients (8.82 %) developed wound infection. Mucosal prolapse was seen in two patients (5.88%). Anal stenosis was noticed in one patient (2.94%). Seven patients (20.58%) developed peri stoma skin excoriation. Stoma prolapse was observed in three patients (8.82%). Four patients (11.76%) developed wound infection after colostomy closure. During the follow up period, constipation was reported in five patients (14.70%). Four patients were grade 11 and managed with diet modification and laxatives. One patient developed grade 111 constipation and was resistant to toilet training, diet modification and laxatives. However this patient improved with daily rectal wash. Soiling in one patient (2.94%). Strong contraction and effective squeeze of sphincter was seen in all 34 patients (100%). Bowel function assessment was done only in those patients who were above three years. In our study 27 patients were above three years of age. Feeling of urge for defecation was noted in all 27 patients (100%). Ability to verbalize and hold back the defecation was in 26 patients (96.29%).

	Range	Mean
Age	9-60 months	27.32
Hospital stay for 1 <sup>st</sup> stage (Colostomy + PSARP)	4 – 6 days	5.20
Hospital stay for 2 <sup>nd</sup> stage (Colostomy closure)	9-11 days	9.17
Duration with colostomy	49-83 days	64.85
Operation time (stage 1)	95-110 minutes	99.14
Operation time (stage 2)	45-60 minutes	52.97
Follow up period	6 – 26 months	13.94

**Table-I. Demographic date**

Post Op Complications	Number	Percent
Wound Infection	03	8.82%
Anal stenosis	01	2.94%
Mucosal Prolapse	02	5.88%
Constipation	05	14.70%
Fecal Incontinence	01	2.94%

**Table-II. Definite procedure related complications**

Colostomy formation related complications		
Complications	Number	Percent
Skin excoriation	7	20.58%
Prolapse	3	8.82%
Colostomy closure related complications		
Wound Infection	4	11.76%

**Table-III.**

Bowel Function	Number	Percent
Feeling of urge	27/27	100%
Capacity to verbalize or report the feeling of urge	26/27	96.26%
Ability to hold back defecation	26/27	96.29%

**Table-IV. Assessment of bowel function in patients > 3 years**

## DISCUSSION

The patients of rectovestibular fistula born with excellent potential in terms of fecal continence and bowel function.<sup>13,14</sup> However they may end up with fecal incontinence due to deficient surgical management. Therefore, depending upon the circumstances in which you are working, available facilities, and expertise of surgeon, the optimal strategy should be adopted to achieve the good bowel function. Selection of technique either PSARP or ASARP, does not matter very much. It depends on expertise, preference and convenience of the surgeon. However it still remains controversy whether the one stage or the two stage repair should be adopted to treat the anorectal anomaly with rectovestibular fistula. A host of pediatric surgeons harbor the idea that formation of colostomy protects the definite procedure. It has been widely accepted that two stage procedure is associated with less risk of surgical site infection and wound dehiscence because of fecal diversion by the colostomy. On the contrary, there are several reasons behind the preference of one stage procedure, for it can prevent repeated anesthesia and surgeries and high complications associated with colostomy and colostomy closure. But apart from that we should be more concerned about the bowel function after definite procedure.

Complication rates after surgery for anorectal

malformation range from 10 to 30%.<sup>1,15,16, and.17</sup> In our study, we achieved very good results by adopting two stage procedure for repair of rectovestibular fistula. Although most of our patients belonged to older age group with high risk of contamination. Operating in the neonatal period has advantage of sterility of meconium.<sup>18,19</sup> Majority of patients came from rural area with compromised hygienic condition and nutritional status. In our study, three patients (8.82%) developed wound infection after definite procedure. Elsaied et al, reported high incidence of wound infection (40%) after PSARP without covering colostomy.<sup>10</sup> Khalifa et al, reported wound infection (41.7%) and wound disruption (29.2%) after ASARP without protective colostomy.<sup>12</sup> In another study, wound infection and dehiscence was reported in 30% of patients after single stage PSARP.<sup>20</sup> Redo surgery for anorectal malformation patients, is usually associated with poor functional outcome. The use of magnetic resonance imaging during revision surgery for recto vestibular fistula is recommended to improve the functional outcome.<sup>9</sup> In our study redo surgery was not required in any patient. High incidence of redo surgery after single stage repair of recto vestibular fistula has been reported in the literature.<sup>10,12,20</sup> Fecal incontinence was noted in one patient (2.94%). It was overflow incontinence, developed after constipation and fecal impaction and managed conservatively. Most of our patients belonged to older age group and more prone to develop constipation. It was observed in five patients (14.70%). High occurrence of constipation, both in single stage and two stage repair, has been reported in the published literature.<sup>10,12,21</sup> Fecal incontinence was noted in one patient (2.94%). It was overflow incontinence, developed after constipation and fecal impaction and managed conservatively. Very good bowel function is expected and should be achieved in each and every patient of rectovestibular fistula.

Approximately 90% of patients with rectovestibular fistula achieve continence and good bowel function by the age of three years.<sup>22,23</sup> Out of 34 participants of study, functional assessment was carried out for 27 patients who were above three years of age. Feeling of urge was present in all

27 patients. Whereas 26 patients (96.26%) were able to verbalize the feeling of urge and hold back defecation. Most common stoma formation related complication was peristoma skin excoriation. It was observed in seven patients (20.58%). Three patients (8.82%) developed colostomy prolapse. Four patients (11.76%) developed wound infection after colostomy closure. All complications associated with colostomy were minor and temporary.

## CONCLUSION

Despite the complications associated with colostomy, two stage repair of recto vestibular fistula has a significant beneficial effect on the functional outcome. Two stage limited PSARP under the cover of colostomy is a safe strategy. It is strongly recommended in a setup with limited resources and weak infrastructure. It is also useful for patients from poor socioeconomic settings.

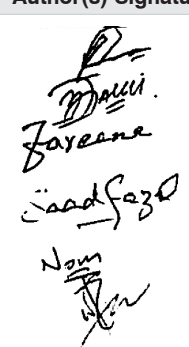
**Copyright© 25 Feb, 2020.**

## REFERENCES

1. Yifeyeh AC, Duduyemi BM, Enimil A, Amoah M, Nimako B. **Malformations: A 5-Years review of the presentation and management in a Teaching Hospital in Ghana.** Afr J Paediatr Surg. 2018; 15(2):118-120.
2. Murphy E, Puri P, Hutson JM, Holschneider AM. **Incidence and frequency of different types and classification of anorectal malformation.** Berlin Heidelberg Springer Veriag; 2006: 163-184.
3. Minneci PC, Kabre RS, Mak GZ, et al; Medwest Pediatric Surgery Consortium. **Screening practices and associated anomalies in infants with anorectal malformations: Results from the Midwest pediatric surgery consortium.** J Pediatr Surg 2018; 53(06):1163-1167.
4. Spouge D, Baird PA. **Imperforate anus in 700,000 consecutive liveborn infants.** J Med Genet Suppl 1986; 2:151-61.
5. Holschneider AM. **Secondary sagittal posterior anorectoplasty.** Prog Pediatr Surg.1990; 25:103-17.
6. Pena A, Devries PA. **Posterior sagittal anorectoplasty: Important technical considerations and new applications.** J Pediatr Surg.1982; 17:796-811.
7. Okada A, Kamata S, Imura K, Fukuzawa M, Kubota A, Yagi M, et al. **Anterior sagittal anorectoplasty for rectovestibular and anovestibular fistula.** J Pediatr Surg.1992; 27:85-8.

8. Aziz DAA, Velyayutham R, Osman M, Latiff ZA, Lim FS, Nor MM. **An rectal anomaly with rectovestibular fistula: A historical comparison of neonatal anterior sagittal anorectoplasty without covering colostomy and postoperative anal dilatation to the classical three-stage posterior sagittal anorectoplasty.** Open Access Surgery. 2017; 10:33-44.
9. Ralls MW, Fallon BP, Torres ML, Ehrlich PF, Jarboe MD. **Salvage laparoscopic-assisted anorectoplasty after failed vestibular fistula repair using magnetic resonance imaging guidance.** European journal of pediatric surgery reports. 2019; 7(01):e12-e15.
10. Elsaied A, Aly K, Thabet W, Magdy A. **Two-stage repair of low anorectal malformations in girls: Is it a true setback?** Ann Pediatr Surg. 2013; 9:69-73.
11. Allam AM, Zeid AAA, El Shafei I, Ghanem W, Albaghdady A. **Repair of low anorectal anomalies in female patients: risk factors for wound dehiscence.** Annals of pediatric surgery. 2017; 13(3):140-4.
12. Khalifa M, Shareef K, Ekrashy MA, Gobran TA. **One or two stages procedure for repair of recto vestibular fistula: Which is Safer? (A Single Institution Experience).** African journal of pediatric surgery: AJPS. 2017; 14(2):27-31.
13. Levitt MA, Pena A. **An rectal malformations.** In: Coran AG, Caldamone AA, Adzick NS, et al, eds. Pediatric Surgery. Philadelphia, PA: Elsevier Health Sciences; 2012.
14. Pratap A, Yadav RP, Shakya VC, Agrawal CS, Singh SN, Sen R. **One-stage correction of recto-vestibular fistula anorectoplasty (TFARP).** World J Surg. 2007; 31:1894-7.
15. Lunkong CS, Ameh EA, Mshelbwala PM, Jabo BA, Gomna A, Akinyi OT, et al. **Management of an rectal malformation: Changing trend over two decades in Zaria, Nigeria.** Afr J Paediatr Surg. 2011; 8:19-22.
16. Sowande OA, Adejuyigbe O, Alatise OI, Usang UE. **Sagittal anorectoplasty of anorectal malformation in Nigerian Children.** J Indian Assoc Pediatr Surg. 2006; 11:85-88.
17. Nakayama DK, Templeton JM, Jr, Ziegler MM, O`Neil JA, Walker AB. **Complications of posterior sagittal anorectoplasty.** J Pediatr Surg. 1986; 21:488-492.
18. Sharma S, Gupta DK. **Delayed presentation of anorectal malformation for definite surgery.** Pediatr Surg Int 2012; 28:831-834.
19. Menon P, Rao KL. **Primary anorectoplasty in females with common anorectal malformations without colostomy.** J Pediatr Surg 2007; 42:1103-1106.
20. Amanollahi O, Ketabchian S. **One-stage vs three-stage repair in a rectal malformation with recto vestibular fistula.** Afri J Paediatr Surg 2016; 13(1):20-25.
21. Heinen FL. **The surgical treatment of low anal defects and vestibular fistulas.** Semin Pediatr Surg. 1997; 6:204-16.
22. Pena A, Hong A. **Advances in the management of a rectal malformations.** Am J Surg. 2000; 180:370-76.
23. Wakhlu A, Pandey A, Parsad A, Kureel NS, Tandon RK, Wakhlu AK. **Anterior sagittal anorectoplasty for a rectal malformations and perineal trauma in female child.** J Pediatr Surg. 1996; 31: 1236-40.

### AUTHORSHIP AND CONTRIBUTION DECLARATION

Sr. #	Author(s) Full Name	Contribution to the paper	Author(s) Signature
1	Shafiq ur Rehman	Introduction / Data collection / Results/Materials & methods.	
2	Yasir Makki	Discussion, Conclusion, References, Recommendation.	
3	Fareena Ishtiaq	Discussion, References, Introduction.	
4	Saad Fazal	Introduction, Discussion, References, Recommendation.	
5	Nauman Aziz	Biostatistics	
6	Waleeja Shamikha	Discussion, References, Conclusion.	