

# Abdominoperineal posterior sagittal anorectoplasty without protective colostomy in high arm pouch colon surgery



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## ABSTRACT

**Background:** In high anorectal malformations (ARM), surgical management includes initial colostomy at neonatal period followed by abdominoperineal posterior sagittal anorectoplasty (PSARP) without a protective colostomy. **Aims and Objectives:** The aims of this study were as follows: (i) To evaluate whether 2 stage PSARP gives better outcome for high ARM than three stage and (ii) to assess overall stay duration, cost incurred, and complications associated with two stage PSARP. **Materials and Methods:** A total 30 patients were studied. Fifteen patients with classical abdominoperineal PSARP with protective colostomy (three stages) constituting Group A studied retrospectively and 15 patients were studied in Group B who underwent neonatal colostomy followed by abdominoperineal PSARP without protective colostomy (Two stages). Comparative study was done between the two groups for a better choice of procedure in high ARM patients. **Results:** In Group A, blood transfusion required was more than Group B. Mean expense incurred in Group A was more than Group B. Complications in Group A occur in eight patients, whereas in Group B, complications occurred in six patients. **Conclusion:** Stages of high ARM and pouch colon are better alternative to three stages repair in all aspect.

**Key words:** High anorectal malformation; Without colostomy; Abdominoperineal posterior sagittal anorectoplasty

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## INTRODUCTION

Anorectal malformations (ARMs) are relatively frequently encountered anomalies that represent an important component of pediatric surgical practice. The anomaly is identified by the absence of anal opening in its normal anatomical position. In many cases, there is a fistulous opening into the urinary tract in the male or in the genital tract in the female. Surgical correction of ARM at times is difficult as the rectum and anus have lost their relationship to the sphincter muscles and these muscles may be abnormal in their development and nerve supply.

### Aims and objectives

The aims of this study were as follows: (i) To evaluate whether 2 stage PSARP gives better outcome for high

ARM than three stage and (ii) to assess overall stay duration, cost incurred, and complications associated with two stage PSARP.

### Classification

ARM represents a wide spectrum of defects (Figure 1). There are different classifications in use in different centers throughout the world making comparisons difficult.

Pena's classification (Table 1) is based on the concept of high and low lesions and their requirement for colostomy or not, and hence, the primary principle is the correct management algorithm in the neonate.<sup>1</sup>

In 2005, in an International Congress for the development of standards for the classification, treatment and follow-up

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**Table 1: Pena classification for Anorectal malformations (1995)**

| Male                             | Female                            |
|----------------------------------|-----------------------------------|
| Perineal fistula                 | Perineal fistula                  |
| Rectourethral fistula            | Vestibular fistula                |
| Bulbar                           |                                   |
| Prostatic                        |                                   |
| Rectovesical fistula             | Persistent Cloaca                 |
|                                  | <3 cm common channel              |
|                                  | >3 cm common channel              |
| Imperforate anus without fistula | Imperforated anus without fistula |
| Rectal atresia                   | Rectal atresia                    |

of ARM took place in Kricken back Castle in Westphalia, Germany. At this meeting, a new international classification system (Kricken back) was development (Table 2).<sup>2</sup>

Efficacious and cost-effective care of patients with ARM being a complicated one, pena advocated a careful thought out plan in the neonatal period for high ARM and pouch colon for three stages repair comprising: diverting colostomy in neonatal period, definitive surgery when 6–8 months old, then closure of colostomy few month late. This staged method offered good result, but it involves 3 admissions, often the patient has to travel long distances during which they would be separated from the rest of their family with several other children.

Wilkins and Peña<sup>3</sup> though proposed formation of initial colostomy for these children to decompress them in neonatal period (Figures 2 and 3) and to protect the subsequent definitive operation, they also expressed that colostomy represents a significant source of morbidity and colostomy complications in these children's were highlighted by Patwardhan et al.<sup>4</sup>

These multistage operations have a number of disadvantages as follows:-

1. High cost of treatment
2. Inherent problems of colostomy-diarrhea, dehydration, prolapse, infection, skin excoriation, stenosis and chronic blood loss leading to anemia, malnutrition, growth retardation etc.
3. Physical, psychological, and financial strain for the parents
4. Problem of anesthesia
5. A number of patients are lost during follow-up after colostomy.

In a two stage operative procedure rather than three stages with different hospital admissions, effectively leads to significant reduction of hospital costs.

The literature of surgical management of ARM has evolved extensively over times. The surgical approach for

repairing these defects changed dramatically in 1980 from sacroperineal to posterior sagittal anorectoplasty (PSARP). Now, it is time to shift toward two stages repair in high ARM and pouch colon whenever possible. Two stage repair often has equivalent or better results yet less morbidity than the three staged repair and physical trauma to the pediatric patient and emotional or mental extortion to their parents are automatically avoided in 2 stage repairs.

## MATERIALS AND METHODS

It was an institution-based observational study conducted in Pediatric Surgery department of Medical College, Kolkata.

As per available statistical data, total 30 such cases were selected to complete the total sample size. They were divided into two groups:

- Group A – neonatal colostomy followed by abdominoperineal pull through and the colostomy closure (Figures 4 and 5)
- Group B – neonatal colostomy followed by abdominoperineal pull though without protective colostomy.

### Inclusion criteria

Patients having high anorectal malformations were included in the study.

### Exclusion criteria

Patients having low ARM were excluded from the study.

### Study design

It is a cross-sectional prospective and retrospective observational study, where 15 consecutive patients undergoing two staged procedure (neonatal colostomy followed by abdominoperineal pull though without protective colostomy) for high anorectal anomalies and pouch colon were recruited for study using inclusion and exclusion criteria. Retrospective analysis of standard three staged procedure (neonatal colostomy followed by abdominoperineal pull through and then colostomy closure) was also studied. Written informed consent was taken in all cases.

### Study tools

1. Pretested and predesigned pro forma
2. Clinical assessment: Detail history, general examination of spine, examination of perineum, examination of gluteal muscles, examination of anomalously placed anus, and external opening (if any)
3. Biochemical investigation: Complete hemogram, coagulation profile, electrolytes, and nutritional status
4. Radiological evaluation: USG whole abdomen, dye-study, and chest X-ray
5. Cardiac evaluation: Echocardiography.

### Study technique

The principal investigator collects data by use of questionnaires and direct observation of the patient in the pre-operative, peri-operative, and post-operative periods.

The results of the two staged procedure (neonatal colostomy followed by abdominoperineal pull through without protective colostomy-Group B) for high anorectal anomalies and pouch colon were compared with 15 cases (History control) that underwent all three stages-(Group A) over a period of 2 years done between January 2017 and December 2018.

### Statistical analysis

Statistical analysis was performed with help of EPI Info (IM) 7.2.2.2. EPININFO is a trademark of the Center for Disease Control and Prevention (CDS).

Besides cross-tabulation and frequency distributions, t-test was used to assess the significant difference between means, odds ratio (OR) with 96% confidence interval (CI) was calculated to measure the validity of the study. pSD was calculated -0.06, considered statistically significant.

## RESULTS

Fifteen cases of two stage procedure (neonatal colostomy followed by abdominoperineal pull through without protective colostomy – Group B) done over a period of 2 years (2017–2018) compared within 15 cases (History control) that underwent all three stages procedure (neonatal colostomy followed by abdominoperineal pull through and then colostomy closure – Group A).

In Group A, there were total 15 patients (Historical control), out of which eight were male and seven were female. In Group B, out of 15, nine were male and six were female.

In Group A, out of eight males, six had rectovesical fistula, one had rectal atresia, and one had pouch colon. In female, out of seven females, three had cloacal anomaly, two had ARM without fistula, one pouch colon, and one rectal atresia (Table 3, Charts 1 and 2).

In Group B, out of nine males, seven had rectovesical fistula and two had pouch colon. In female, out of six females, one had cloacal anomaly, two had ARM without fistula, one pouch colon, and one rectal atresia (Table 4, Charts 3 and 4).

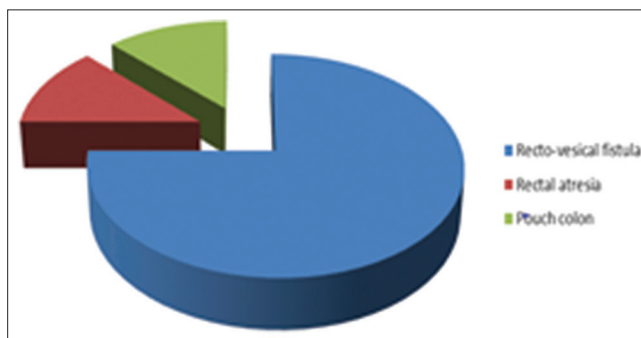
In Group A, each patient underwent three operations (neonatal colostomy followed by abdominoperineal pull

**Table 2: International Classification (Krickenn back)**

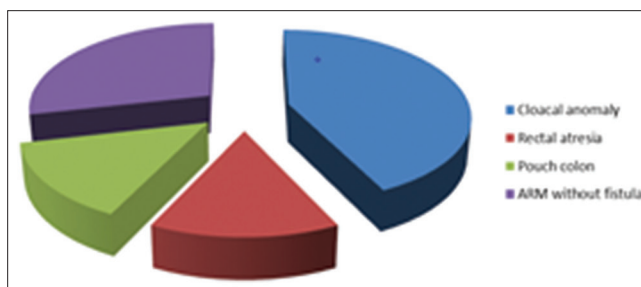
|                           |   |
|---------------------------|---|
| M - Major clinical groups | Per perineal (cutaneous) fistula, rectourethral fistula, bulbar prostatic, rectoversical fistula, vestibular fistula, per persistent cloaca, no fistula A – anal stenosis |
| Rat - Rare/               | P – Pouch Colon   |
| Re - Regional             | R – Rectal atresia/stenosis, rectovaginal fistula   |
| Va - Variants             | H – H-type fistula  |
|                           | Ot – Other  |

**Table 3: For Group A**

| Diagnosis            | Male | Female |
|----------------------|------|--------|
| Rectovesical fistula | 6    | 0      |
| Pouch colon          | 1    | 1      |
| Cloacal anomaly      | 0    | 3      |
| ARM without fistula  | 0    | 2      |
| Rectal atresia       | 1    | 1      |



**Chart 1:** Distribution of various diagnoses in male patients in group A



**Chart 2:** Distribution of various diagnoses in female patients in Group A

through and then colostomy closure). Hence, 15 patients underwent total 45 operations. Total 28 units blood transfusion required in Group A patients. In Group B, each patient underwent two operative procedures. Hence, total number of operations in this group were 30 and blood transfusion requirement in this group was 15 units (Table 5 and Chart 5).

Hospital stay in Group A patients ranges from 24 days to 36 days with a mean of 28.66 days. In Group B, hospital stay ranges from 15 days to 26 days with the mean of 19.93 days (Table 6 and Chart 6).

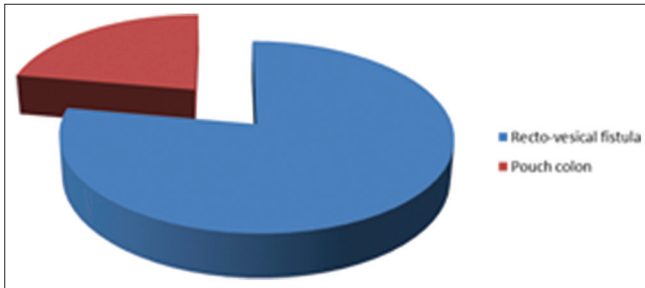


Chart 3: Distribution of various diagnoses in male patients in Group B

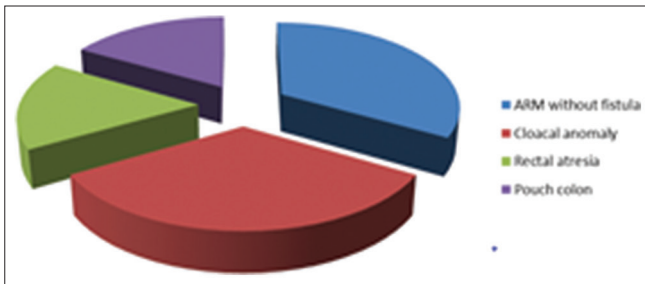


Chart 4: Distribution of various diagnoses in female patients in group B

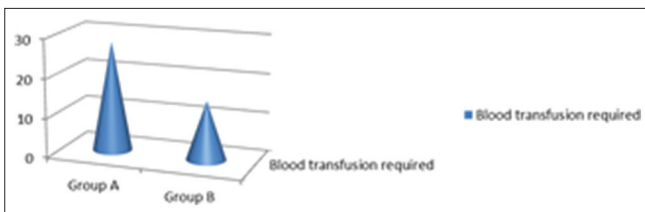


Chart 5: Blood transfusion required in Group A and Group B

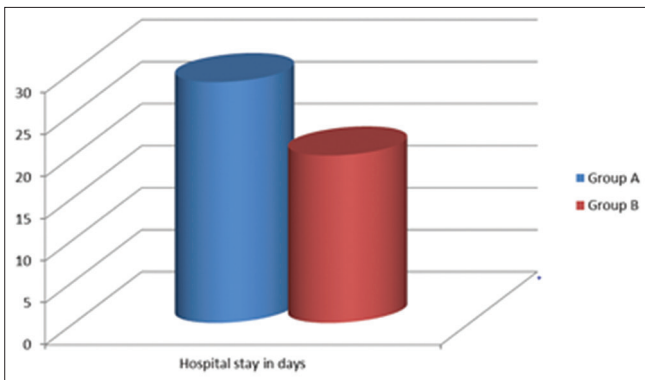


Chart 6: Hospital stay in days in Group A and Group B

Expense incurred by hospital and guardians, in Group A, it ranges from Rs. 17,500 to 28899 with the mean of Rs. 22,160. In Group B, it ranges from Rs. 11,000 to 17899 with the mean of Rs. 14,233 (Table 7).

Complications in Group A occurred in eight patients out of which six had excoriation of peri colostomy skin, one had skin dehiscence of perineal wound and one patient had wound infection following colostomy closure. In Group B, out of 15 patients, complications occur in six patients, out

Table 4: For Group B

| Diagnosis            | Male | Female |
|----------------------|------|--------|
| Rectovesical fistula | 7    | 0      |
| Pouch colon          | 2    | 1      |
| Cloacal anomaly      | 0    | 1      |
| ARM without fistula  | 0    | 2      |
| Rectal atresia       | 0    | 1      |

Table 5: Requirement of blood transfusions

| Blood transfusion requirement | G - Group A | Group B  |
|-------------------------------|-------------|----------|
|                               | 28-28 units | 15 units |

Table 6: Mean hospital stay in patients

| Hospital stay                    | Group A         | Group B         |
|----------------------------------|-----------------|-----------------|
| H - Hospital stay (Mean=in days) | 28.66±3.73 days | 19.93±2.98 days |

t-test (P<0.005)

Table 7: Expenses incurred to Guardians

| Expenses(Rs)                | Gr - Group A   | Group B        |
|-----------------------------|----------------|----------------|
| Ex - Expense incurred (Rs.) | 22.160±3672.83 | 14.233±1821.17 |

t-test (P-value) <0.0001

Table 8: Post Surgical Complications

| Co - Complications                                    | Gr - Group A | Group B |
|---|--------------|---------|
| Per - Pericostomy skin excoriation                    | 6            | 4       |
| Partial dehiscence of perineal wound                  | 1            | 1       |
| Abdominal wound infection following colostomy closure | 1            | 1       |

of which four had excoriation of pericostomy skin and one had skin dehiscence of perineal wound and one patient had wound infection following colostomy closure. All the complications were managed conservatively successfully (Table 8 and Chart 7).

## DISCUSSION

The PSARP devised by Peña and Devaries (1982) has revolutionized the management of ARM by providing complete exposure of the anatomy of the anorectal region during surgery. The conventional approach for the surgical correction of high ARM in a newborn entails a high sigmoid colostomy, PSARP as described by Pena, and colostomy closure. Problems encountered include comorbidity associated with a colostomy, increased costs of 3-stage operations, and number of drop-outs after colostomy especially in developing world.<sup>5</sup> Another

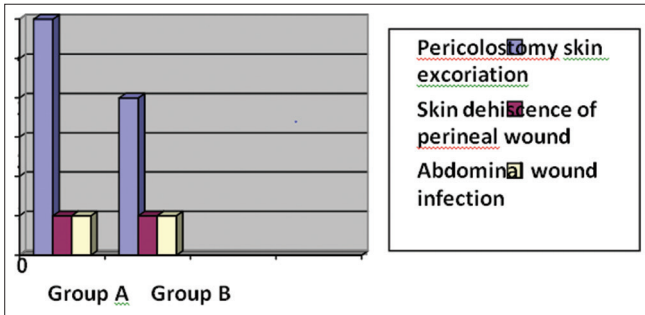


Chart 7: Distribution of various complications in Group A and Group B



Figure 1: Newborn present with anorectal malformation



Figure 2: Prone jack-knife position for operation

important issue especially in developing world is that a number of patients may not return at 6–8 weeks for definitive surgery.

Primary correction for high ARM at birth in neonates is not much popularized and there are only few published studies. Moore first described sagittal anorectoplasty performed through an anterior approach without colostomy in newborns.<sup>5</sup> Adeniran<sup>6</sup> showed that it is feasible for girls with imperforate anus and rectovestibular fistula to



Figure 3: Posterior sagittal midline incision

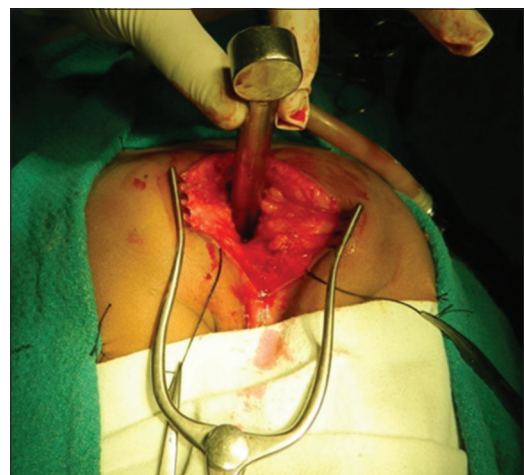


Figure 4: Creation of channel for the new rectum



Figure 5: Wound closure behind rectal tube at proposed rectal site

have safe PSARP without colostomy. The advantages of one, instead of three major operations, are many, especially in developing countries. Liu and Hill series of seven male newborns with rectouniary tract fistula who underwent primary PSARP showed good results.<sup>7</sup> Mishra et al.,<sup>8</sup> Gangopadhyay et al.,<sup>9</sup> Mirshemirani et al.,<sup>10</sup>

in their comparative study of primary PSARP and staged procedures also described good results in neonates who underwent primary corrective procedure at birth. One of the concerns for primary correction in high ARM cases is lack of understanding about local anatomy with subsequent risk of damaging the local structures. Experienced surgeons agree to the fact that most accidents occur in patients with high ARMs. Such mishaps may result in permanent urethral damage, division of the vas, pull through of dilated ectopic ureters, or a neurogenic bladder. To prevent such problems, but also to repair these malformations at an earlier age, Pena suggested to start one stage repair of such malformations in neonates with low lying rectum. Although selection of such patients is very difficult, Albanese has used pre-operative proctoscopy, but identified the fistula in three patients only.<sup>11</sup> Most other surgeons have directly proceeded with the primary PSARP without paying much attention to the urinary communications, which were dealt with only intraoperatively but such blind procedure may be dangerous. In our study, we have done neonatal colostomy followed by definitive procedure without protective colostomy. As we have done the definitive surgery only after proper knowledge of local anatomy, radiological investigations, and after ruling out any other associated anomalies, we have avoided the complications of primary correction and none of our study cases has any evidence of damage to local structures. Furthermore, by avoiding the protective colostomy in second stage, we have avoided complications related to prolonged colostomy to great extent.

Another important concern during definitive surgery without protective colostomy is the wound contamination with stool resulting in wound infection and wound dehiscence. However, we observed that when the colon was empty of stool with proper bowel preparation and when the wound area was kept clean in post-operative period, soiling of wound with stool was never a problem.<sup>12,13</sup>

In this study, we have performed abdominoperineal pull through in all our cases, so we have avoided all the complications associated with distal post colostomy segment like fibrosed thickened pouch due to repeated pouchitis, tapered rectal segment when pouch is small and one of the most dreaded complication, that is, stenosis due to tension at neoanal junction and compromised vascularity. As we have used healthy pre-colostomy bowel segment which was thin, functioning, and well-vascularized, there was no problem related to continence and anal stenosis as far as bowel caused is concerned.

In our study, there were male preponderance in both groups, in Group A, there were total 15 patients (historical control) out of which 8 (54%) were male and seven were female. In Group B, out of 15 patients, nine were male

(66%) and 6 were female. These results are comparable to other studies which stated that ARM has higher incidence in boys than girls. The sex ratio varies from 66% to 70% in favor of boys.<sup>12</sup> Furthermore, studies have shown a more frequency of high ARM (supralevator lesions) in boys as compared to girls, ranging from 44% to 65%.<sup>13</sup>

In our study in Group A and Group B, most common diagnosis was rectovesical fistula which accounted for 40% of cases in Group A and 56% in Group B. These results supported the other series result which stated that rectovesical fistulas occur in half of all high ARM cases.<sup>12,13</sup>

On combining pouch colon cases of both groups, these account for 16.6% cases out of total 30 cases. In which there was male predominance with 10% contributed by male whereas 8% by females. The results are comparable to other studies which found the incidence of congenital pouch colon from 2% to 15.2% and almost all the series reported male preponderance.<sup>12,13</sup>

In Group A, each patient underwent three operations, so 15 patients underwent total 45 operations. During these 45 operations 28 units, blood transfusion was required. In Group B, 15 patients underwent total 30 operations and blood transfusion requirement in this group was 15 units. On statistical analysis, this difference was found to be statistically significant ( $P < 0.05$ ).

Hospital stay in group A patients ranges from 24 to 36 days (mean of  $28.66 \pm 3.73$  days) with 5–10 days for colostomy, 10–16 days for definitive surgery and 7–10 days for colostomy closure. In Group B, the duration of hospital stay ranges from 15 to 26 days (mean of  $19.13 \pm 2.98$  days) with 5–10 days for colostomy and 10–16 days for definitive surgery. Malnutrition and complications leading to poor general health are some of the factors which lead to prolonged hospital stay in both groups, though the difference found to be statistically significant. Hence, by avoiding one extra stage in the form of colostomy closure, we can cut short overall hospital stay with decreased hospital burden, less demand for workforce and also psychological burden to patient and parents.

Expense incurred by guardians in Group A ranges from Rs. 17,500 to Rs. 28,800 (mean of Rs.  $22,160 \pm 3672,83$ ). In Group B, range was Rs. 11,000–17,899 (mean of Rs.  $14,233 \pm 1821,17$ ). The results of Group A was compared to Group B in our study, found to be strongly significant ( $P < 0.0001$ ).

Complications in Group A occurred in seven patients, out of which six had excoriations of pericostomy skin, one had parital perineal wound dehiscence, and one had infection

of wound following colostomy closure. In Group B, out of 15 patients, complications occurred in six patients, out of which four had excoriation of pericostomy skin, one had parital perineal wound dehiscence, and one had infection of wound following colostomy closure. All cases were managed conservatively successfully.

Hence, in our study, we found that two stage repair of high ARM and pouch colon not only has obvious advantages over three stage correction without increasing the risk to patients but also has financial and psychological benefits to parents. Furthermore, there is an institutional advantage of decreasing waiting time and hospital stay, blood requirement, and reduced cost.

However, it should be noted that the number of patients in recent study are few, whether this approach is preferable over classical 3 – stage repair depends on multi-institutional experience in large number of patients and on the long-term anorectal function. Therefore, the long-term outcome of these patients should be the subject of future studies.

#### Limitations of the study

This was a single centre study with a very limited sample size of only 30 patients. A larger multicentre study with larger sample size is required for drawing further inferences.

## CONCLUSION

Stages of high ARM and pouch colon are better alternative to three stages repair in all aspect.

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#### Authors' Contributions:

**SN and SK**- Concept and design of the study, prepared first draft of manuscript; **SN and SM**- Reviewed the literature, and manuscript preparation; **SK and SM**- Concept, coordination, statistical analysis and interpretation, interpreted the results, revision of the manuscript.

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