


Single Stage Management of Anorectal Malformation in Male Neonates: Experience of a Tertiary Care Centre

Vinit Kumar Thakur^{1*}, Zaheer Hasan¹, Vijayendra Kumar¹, Ramdhani Yadav¹, Sandeep kumar Rahul¹, Digamber Chaubey¹

¹Indira Gandhi Institute of Medical Science, Patna, Bihar

*Address for Corresponder: Dr. Vinit Kumar Thakur, Indira Gandhi Institute of Medical Science, Patna, Bihar (email: kumvinit134@gmail.com)

How to cite this article:

Thakur V, Hasan Z, Kumar V, Yadav R, Rahul S, Chaubey D. Single Stage Management of Anorectal Malformation in Male Neonates: Experience of a Tertiary Care Centre . Iranian Journal of Pediatric Surgery 2020; 6 (1):30 -36

DOI: <https://doi.org/10.22037/irjps.v6i1.31701>

Abstract

Introduction: Anorectal malformation [ARM] can be treated either by staged procedures or by a single stage procedure. In the present study we have performed single stage surgery of ARM in male neonates.

Materials and Methods: Retrospective review of cases of ARM operated over 4 years at Indira Gandhi Institute of Medical Sciences, Patna, and Bihar from January 2015 to December 2018 were undertaken. In this study we have compared the results of single stage repair of Anorectal malformation with that of staged procedures performed earlier. Fifty four patients of Anorectal malformation were operated by single stage procedure (category 1) this was compared with staged procedures (category 2).

Forty three patients underwent primary PSARP (posterior sagittal anorectoplasty) and the remaining 11 primary APP (abdomino-perineal pull through) procedure. The result of these single stage procedures were compared with that of staged procedures (category 2) in which a total of 39 cases were operated from 2012 to 2014.

Result: Mean post-operative hospital stay in category 1 was 11 days and 32 days in category 2. In category 1, 20 patients were analysed. In which PSARP procedure was done in 15 and in 5 patients APP procedure was performed. The Rest of the patients are in follow up. Kelly score was good to fair in 17 patients (85%) and poor in 3 (15%) patients. In category 2 it

Keywords

- Anorectal malformation
- Neonate
- Single stage procedure

was good to fair in 28 (71.7%) cases and poor in 11 cases (28%).

Conclusion: Single stage surgical repair of anorectal malformation in male neonates is reliable and can be safely performed. Early results are encouraging; however, a long term follow-up is required to get any definite conclusion.

Introduction

Anorectal malformations (ARMs) are congenital anomalies that specifically affect the anorectal anatomy in which the anus is either nonexistent or malformed. It has an incidence of 1 in 2500-5000 births; yet might be more common in some developing countries.¹ Traditionally in Anorectal malformation, neonatal diverting colostomy followed by PSARP/APP and subsequent colostomy closure is done however, primary surgery without using colostomy is an emerging trend as colostomy is often associated with morbidity and mortality and generally not favoured by parents in the modern society aesthetically. Keeping this in mind, we have performed primary surgery for male patients presenting with high type and intermediate type anorectal malformation.

Materials and Methods

Retrospective review of cases of ARM managed from January 2015 to December 2018 was undertaken. The study was approved by the Ethics Committee of the Indira Gandhi Institute of Medical sciences, Patna, Bihar. In category 1, a total of 54 male patients of Anorectal malformation underwent single stage operation. The result of this procedure was compared with that of staged procedure, category 2, in which a

total of 39 cases were operated on before 2015. Both groups were compared with respect to age at the time of definitive surgery, duration of surgery, hospital stay, post-operative complications as well as faecal continence in patients of more than 3 years. Routine blood count, biochemistry, renal function test, Ultrasonography of abdomen and echocardiography were performed in all patients along with Prone cross table lateral X-ray with focus on greater trochanter for detecting the level of gas shadow. If the gas shadow was above the Pubococcygeal line (PC), we did Abdomino-perineal pull through procedure (APP) and if found below the PC line we performed posterior sagittal anorectoplasty (PSARP) procedure. Out of 54 patients of ARM, 43 patients were operated by PSARP (posterior sagittal anorectoplasty) and 11 by APP (Abdomino-perineal pull through) for high ARM. In two patients Abdomino-PSARP procedure were done because pouch could not be located by the PSARP approach. Our exclusion criteria were as follows:

- (1) Low ARM,
- (2) Presenting with gut perforation,
- (3) Sepsis
- (4) Associated with complex anomalies

Procedure

All the surgeries were performed under general anaesthesia with endotracheal intubation. Primary PSARP procedure was done using midline perineal incision after taking stay suture over the proposed anus. Muscle and sphincter was cut in the midline using needle tip cautery. We first deflate the air from terminal pouch using syringe simultaneously we avoid using Babcock forceps as it is crushing to native bowel; rather we use stay suture and vascular forceps to hold the bowel. We also use wet gauze piece for mobilising the rectal pouch. Terminal pouch is opened and meconium sucked out subsequently, sub mucosal dissection proximal to fistula site is carried out and the pouch is mobilised. Then the fistula is closed; perineal body reconstituted and anorectoplasty done. In the APP procedure the abdomen is opened by a left lower paramedian incision. Bowel is deflated and traction to terminal pouch is given using an infant feeding tube. Dissection of rectum is started at the site of peritoneal reflection, ligation of fistula done, and subsequent pull-through of colon followed by anoplasty is carried out. In the post-operative period patients were nursed in surgical neonatal intensive care unit in prone position to prevent faecal contamination. Post-operative dressing was done using diluted povidone iodine and mupirocin ointment.

Result

In the category 1 median age for definitive surgery was 3 days and 360 days in category 2. Among the 54 cases we had 43(79.6%) cases of Intermediate

ARM and 11(20.3%) cases of high ARM. Mean duration of surgery in category 1 was 1.15 hours and 3.5 hours in category 2 which includes initial colostomy, PSASRP or APP and colostomy closure. In the category 1 post-operative feeding was started from 3rd (mean) post-operative day, after abdominal distension had resolved. Urinary catheter was removed by day 7 or 8 post-operatively in primary surgery. Mean post-operative hospital stay in category 1 was 11 days and 32 days in category 2. The difference was found to be statistically significant. Patients were advised to undergo anal dilatation programme after two weeks of surgery. During the post-operative period in both categories patients were kept on enema to keep the rectum empty which was required less frequently in category 1 in the subsequent follow up visits. Mean weight gain for similar age groups was better in category 1 than in category 2. In patients with high ARM defecation was more frequent, resulting in associated perineal excoriation which were lessened in the subsequent visits by applying conservative management. In eight patients (14.8%) in category 1 there was perineal excoriation and five patients suffered from minor wound infection. They were managed using dressing and zinc oxide based ointments. In category 1, 41 (75.9%) cases had stool frequency 3-5 times per day while in category 2, 20 (51%) patients had the same frequency at the age of three years. This difference was found to be statistically significant (p -value < 0.001). Various complications associated with both procedures are shown in [Table 1](#).

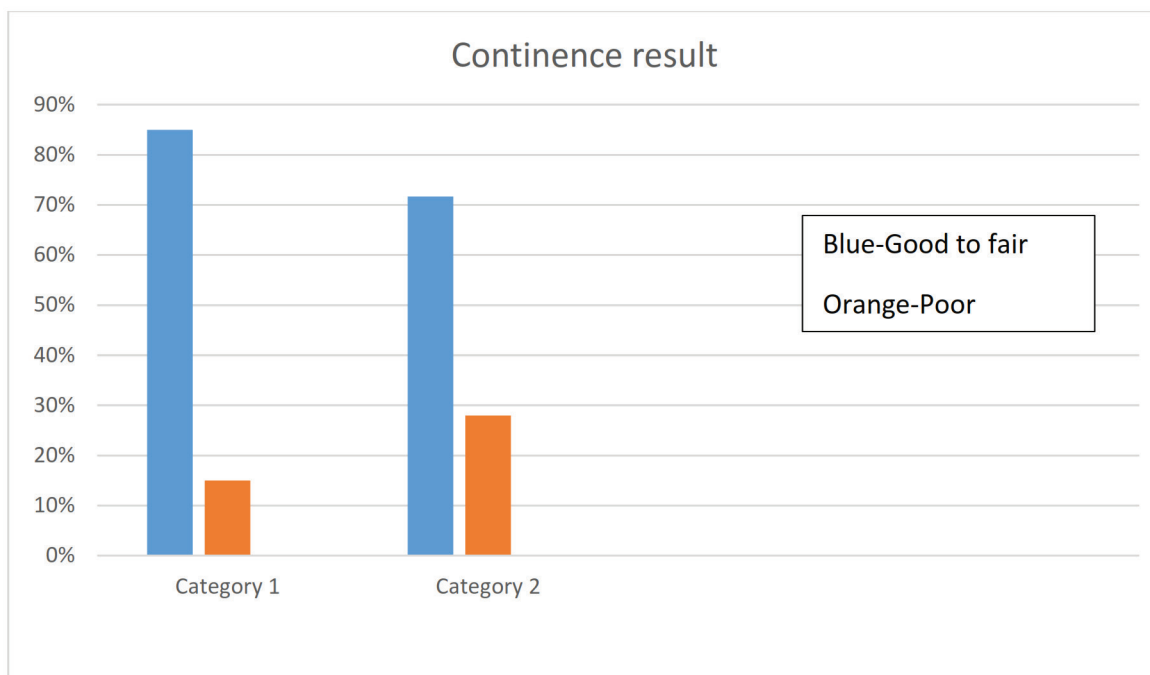
Table 1: Complications of Category 1 and Category 2

	Category 1 n (%)	Category 2 n (%)
Constipation	3 (5.5)	7 (17.9)
Stenosis	0 (0)	3 (7.6)
Mucosal prolapse	1 (1.8)	3 (7.6)
Neurogenic bladder	1 (1.8)	2 (5.1)
Urethral injury	0 (0)	2 (5.1)
Perineal excoriation	8 (14.8)	10 (25.6)
Wound infection	5 (9.2)	2 (5.1)
Colostomy prolapse	0 (0)	4 (10.2)

Perineal excoriation and wound infection were encountered more commonly in category 1 than in category 2. Mucosal prolapse was detected in one case in category 1 and two cases in the category 2.

Continence was assessed by Kelly's score (continence, staining and sphincter squeeze) in the 3 year old age group. In category 1, 20 patients

could be analysed. Of which PSARP procedure was done in 15 cases and in 5 patients APP was performed. The remaining patients of this category are under follow up. Kelly's score was good to fair in 17 patients (85%) and poor in 3(15%) patients. In category 2 it was good to fair in 28 (71.7%)cases and poor in 11 cases (28%) **Figure 1**.

**Figure 1:** showing continence result of category1 and 2

There was one death in category 1 in the post-operative period due to sepsis whereas in category 2 there were two deaths due to colostomy related complication.

Discussion

ARM has been a source of concern for many centuries. The etiology of ARM is unclear and likely multifactorial. Its treatment mainly consists of anoplasty, PSARP and abdominoperineal pull-through for the selected group of high ARM. Recently, Georgeson et al,² performed a one-stage AP pull-through using a laparoscope that provided excellent exposure deep in the pelvis. They used a harmonic scalpel to divide the fistula, and passed the colon through an intact sphincter to the anal dimple where an anoplasty was performed. The choice between selecting the one-step or three-step procedure to treat ARMs has always been a subject of debate. Reasons for choosing the 3-stage repair are lesser risks of surgery due to faecal 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 single stage repair are multiple, for example, avoidance of multistage operations will lead to saving time and costs, less morbidity for children and avoidance of colostomy related complications. In addition dissection is easier and takes less time in the neonatal period due to virgin tissue planes.³ Moreover, the children with colostomy will have problems in peer relationships, school absentees and may develop behavioural problems. This led us to evolve single-stage management of all ARM cases. Definitive procedure in the neonatal period

itself is recommended since cerebral cortical nerve fibers evolve in the first years of life and having a sensation of rectal fullness is necessary for these fibers to develop completely so that continence can be achieved to its maximal potential.^{4,5,6} Several authors in the literature have reported that one-stage repair of ARM cases by PSARP in neonates was safe and feasible in selected group of patients.^{7, 8, 9} Elhalaby in his study on 38 patients demonstrated that primary repair of high and intermediate ARMs in infants were feasible.¹⁰ Another study in 2005, conducted by Gangopadhyay et al, in 105 patients, showed that an initial one-stage surgery method had better apparent results and fecal control, and was associated with a reduced mortality rate and reduced cost.¹¹ The safety of the approach depends on the exclusion of associated anomalies with anorectal malformation.¹² 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.¹³ To assess post-operative functional result we have used stool frequency. Many scientists have worked on developing this clinical scoring method such as: Kelly, Templeton et al, Kiesewetter and Chang, and Stephens and Smith. The score is based on the degree of continence and the quality of life after management.¹⁴ Up to now many studies have tried to prove the advantage of one method of repairing ARM such as PSARP over another; by comparing the level of continence of patients. In this regard Rintala and Lindahl showed that in terms of long-term bowel function and faecal continence, PSARP is better than sacroperineal and sacro-abdominoperineal pull-through.¹⁵ In contrast, Gil-Vernet et al. reported abdomino-

perineal pull-through plus PSARP to have better results in patients with high ARM compared to PSARP alone.¹⁶ In our study category 1 has better continent result compared to category 2. Since the definitive procedure was done very early, anorectal continuity was achieved early which we believed helped in this regard. Our early results are comparable to other studies.^{4,3} The early post-operative complications in the present study are acceptable and its functional results are better than that of the conventional multi-stage procedure. Only few studies have been conducted involving exclusively male neonates with anorectal malformation with PSARP procedure.^{17, 18, 19}

Conclusion

Single stage surgical repair of anorectal malformation of high and intermediate type in male

neonate is reliable and can be safely performed and early results are encouraging; however, a long term follow-up is required to reach any definite conclusion.

Ethical Consideration

Ethical committee approval No- 1259/Acad dated 30-11-2016

Acknowledgements

Not applicable

Funding/Support

Not applicable

Conflict of interests

There is no conflict of interests.

References

1. Holschneider AM, Hutson JM: Anorectal Malformations in Children: Embryology, Diagnosis, Surgical Treatment, in Moore SW(ed): Genetics, Pathogenesis and epidemiology of anorectal malformation and caudal regression syndrome. Springer Science & Business Media, 2006, pp3.
2. Georgeson KE, Inge TH, Albanese CT: Laparoscopically assisted anorectal pull-through for high imperforate anus; new technique. *J Pediatr Surg* 2000; 35:927-30.
3. Gangopadhyay AN, Gopal SC, Sharma S, et al: Management of anorectal malformations in Varanasi, India: a long-term review of single and three stage procedures. *Pediatr Surg Inter* 2005; 22:169-172.
4. Albanese C, Jennings RW, Lopoo JB, et al: One stage correction of high imperforate anus in the male neonate. *J Pediatr Surg* 1999; 34:834-836.
5. Dobbing J, Sands J: Quantitative development of growth and development of the human brain. *Arch Dis Child* 1973; 48:757.

6. Moore TC: Advantages of performing the sagittal anoplasty operation imperforate anus at birth. *J Pediatr Surg* 1990;25:276.
7. Elsaied A, Aly K, Thabet W, et al: Two-stage repair of low anorectal malformations in girls: Is it truly a setback? *Ann Pediatr Surg* 2013; 9:69–73.
8. Osifo OD, Osagie TO, Udefiagbon EO: Outcome of primary posterior sagittal anorectoplasty of high anorectal malformation in well selected neonates. *Niger J Clin Pract* 2014; 17:1–5.
9. Liu G, Yuan J, Geng J, et al: The treatment of high and intermediate anorectal malformations: one stage or three. *Journal of Pediatric Surgery* 2004;39:1466-71.
10. Elhalaby EA: Primary repair of high and intermediate anorectal malformations in the neonates. *Ann Pediatr Surg* 2006; 2:117–22.
11. Gangopadhyay AN, Shilpa S, Mohan TV, et al: Single-stage management of all pouch colon (anorectal malformation) in newborns. *J Pediatr Surg* 2005;40:1151–5.
12. Gangopadhyay AN, Pandey V: Simultaneous single-staged repair of anorectal malformation with tracheoesophageal fistula: Lessons learned. *J Indian Assoc Pediatr Surg* 2017; 22:96-100.
13. Adeniran JO: One-stage correction of imperforate anus and rectovestibular fistula in girls: Preliminary results. *J Pediatr Surg* 2002; 37:16.
14. Ong NT, Beasley SW: Comparison of clinical methods for the assessment of continence after repair of high anorectal anomalies. *Pediatr Surg Int* 1990; 5:233-7.
15. Rintala RJ, Lindahl HG: Posterior sagittal anorectoplasty is superior to sacroperineal-sacroabdominoperineal pullthrough: a long-term follow-up study in boys with high anorectal anomalies. *J Pediatr Surg* 1999; 34:334–337.
16. Gil-Vernet JM, Asensio M, Marhuenda C, Broto J, et al: Nineteen years' experience with posterior sagittal anorectoplasty as a treatment of anorectal malformation. *Cir Pediatr* 2001; 14:108–111.
17. Mishra BN, Narasimhan KL, Chowdhary SK, et al: Neonatal PSARP versus staged PSARP: A comparative analysis. *J Indian Assoc Pediatr Surg* 2000; 5:10–3.
18. Mirshemirani A, Kouranloo J, Rouzrokh M, et al: Primary posterior sagittal anorectoplasty without colostomy in neonates with high imperforate anus. *Acta Med Iran* 2007; 45:121–5.
19. Ernesto Leva, Francesco Macchini, Rosella Arnoldi, et al: Single stage surgical correction of Anorectal malformation associated with rectourinary fistula in male neonate. *J of neonatsurg* 2013;2:1-3.