

Anorectal Malformations and Posterior Sagittal Anorectoplasty- A Prospective Study for a Period of One Year in North-West Punjab

AMARJIT SINGH KUKA, NISHAN SINGH, SP SINGLA, PUSHPINDER SINGH KUKA

ABSTRACT

Introduction: Anorectal Malformations (ARM) have enormous impact on the patient's quality of life. It is not only a surgical challenge but also a shocking event for parents and whole family. Despite of advances in field of surgery, it challenges the wisdom and expertise of surgeons.

Aim: To determine the incidence of ARM in North West Punjab and to evaluate the success of different types of surgeries such as Posterior Sagittal Anorectoplasty (PSARP), Anterior Sagittal Anorectoplasty (ASARP) and cut back anoplasty. The prevalence of bowel continence after surgery for high or low ARM was also studied along with proportion of cases requiring colostomy.

Materials and Methods: A prospective study was done in thirty (30) patients having ARM in North-West region of Punjab, admitted over a period of one year. In low varieties of ARM, the primary surgery was done at the time of presentation. In high and intermediate varieties, primary defunctioning colostomy was done at the time of presentation followed by definitive repair later. In majority of patients definitive repair was done by PSARP, in few cases by ASARP and very few cases PSARP combined with Abdominal pull through approach. Colostomy closure was done after 4-6 weeks of regular anal dilatation.

Results: The male to female ratio was 1:1, with 60% patients presenting in neonatal period and 40% in post neonatal period. The level of lesion was high in 46.66%, intermediate in 13.33%, and low in 30% patients. Cloaca was seen in 10% of cases. The common presenting symptoms were not passing meconium since birth (50%), absent or abnormal anal opening, vomiting (13.33%) with increasing abdominal distension (26.66%), excessive crying and passing of thin pipe stools with constipation (23.33%), passing stools through vagina (13.33%) or urethra (3.33%) or an abnormal opening elsewhere in the perineum (20%). Associated urogenital anomalies were seen in 13.33% cases. Other anomalies included Meckel's diverticulum (6.66%), pouch colon (3.33%), bicornuate uterus (3.33%) hypospadias with meatal stenosis (3.33%), undescended testes (3.33%), Inguinal hernia (3.33%), bilateral choanal atresia (3.33%), band at ileo caecal region (3.33%) and terminal ileum opening in caecum attached to recto sigmoid region (3.33%). Associated fistulae were seen in 16.66% of patients. Overall morbidity of the definitive procedure was 31.58% and mortality was 10% in present study.

Conclusion: In majority of patient's definitive repair can be done by PSARP, ASARP PSARP combined with abdominal pull-through approach. Primary PSARP can be tried in rectovestibular, rectovaginal fistula and in some low varieties of ARM.

Keywords: Abdominal pull-through, Anterior saggital anorectoplasty, Fistula, Urogenital anomalies

INTRODUCTION

Anorectal Malformation has enormous impact on patient's quality of life. Birth of child with anorectal malformation is not only a surgical challenge but also a shocking event for parents and whole family. The field of surgery has seen enormous advancements and from the ancient times, when infants with ARM were allowed to die to the development of posterior saggital anorectoplasty by Alberto Pena in 1982, we have come a long way [1]. Early diagnosis, advances in anaesthesia, special procedures and specialized post

operative care has made it possible to perform complex procedures successfully. Anorectal malformations have been classified in various ways. However, Krichenbeck classification is most widely accepted [2] [Table/Fig-1].

AIM

The aim of the study was to determine the incidence of ARM in North-West Punjab and to evaluate the success of different types of surgeries such as Posterior Sagittal Anorectoplasty (PSARP), Anterior Sagittal Anorectoplasty (ASARP) and cut back anoplasty. The

prevalence of bowel continence after surgery for high or low ARM was also studied along with proportion of cases requiring colostomy.

MATERIALS AND METHODS

This prospective study comprised of 30 cases of Anorectal Malformations who were admitted during 1st October 2008 to 30th September 2009 in the Surgery Department of Guru Gobind Singh Medical College, Faridkot, Punjab, India. The study was approved by the College Ethical committee. In all cases, as soon as patient was brought to the hospital, a quick assessment was done. In all patients presenting with neonatal intestinal obstruction an intravenous line was secured, intravenous fluids started, nasogastric tube put in, followed by broad spectrum antibiotics and injection vitamin K.

Inclusion criteria

History of-

1. Not passing stools since birth
2. Absence of normal anal opening
3. Passing stools from abnormal routs i.e. vagina, urethra or ectopic sites,
4. Abdominal distension,
5. Vomiting, excessive crying,
6. Off and on constipation,
7. Thin pipe stools

Exclusion criteria:-

1. Other causes of abdominal distension and vomiting.
2. Those who did not give consent for inclusion in the study.
Local examination of anus, anal dimple, perineum and ectopic opening were done and recorded.

Per abdomen examination for distension, tenderness, lump, visible peristalsis, bowel sounds and detailed examination of previously done colostomy was done. General physical and systemic examinations were done to rule out associated congenital anomalies.

Radiological studies like invertogram and lateral prone cross table view X-rays were done as required. Thus level of lesion i.e. low, intermediate or high was decided. If required, ultrasound abdomen was also done in some patients to rule out other associated abnormalities.

In low varieties of ARM the primary surgery was done at the time of presentation. They had good fecal continence post-operatively provided the perineal muscles and rectum were not injured during surgery and major infection did not occur. In high and intermediate varieties, primary defunctioning colostomy was done at the time of presentation followed by definitive repair later. In majority of patients definitive repair was done by PSARP, in few cases by ASARP and very few cases PSARP combined with abdominal pull through approach. Care was taken to prevent heat loss prior, during and after the surgery.

Colostomy closure was done after 4-6 weeks of regular anal dilatation to prevent stenosis. Two weeks

after the definitive procedure anal dilatation done as per schedule until rectum reaches the desired size, frequency of dilatation reduced once parents state that dilator goes easily without pain patient admit little finger easily. Little finger of parents is best dilator as per our judgement. Colostomy closed if dilatation is satisfactory. A patient who had 1-3 bowel movements per day and remained clean in between bowel movements was considered as good prognosis. The approach to male and female patients with ARM is shown in [Table/Fig-2,3] respectively [3].

RESULTS

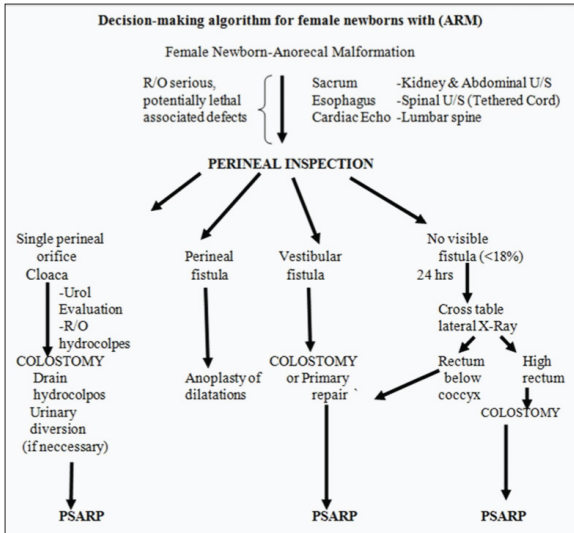
In the present study, there were 15 males (50%) and 15 females(50%) with male to female ratio of 1:1, with 18 (60%) patients presenting in neonatal period and 12 (40%) in post neonatal period. The type of ARM, the sex incidence and type of lesion is shown in [Table/Fig-4,5]. The level of lesion was high in 14(46.66%), intermediate in 4(13.33%), and low in 9(30.00%) patients. Cloaca was seen in 3(10%) of the study cases.

The type of The common presenting symptoms were not passing meconium since birth (50%), absent or abnormal anal opening, vomiting (13.33%) with increasing abdominal distension (26.66%), excessive crying and passing of thin pipe stools with constipation (23.33%), passing stools through vagina (13.33%) or urethra (3.33%) or an abnormal opening elsewhere in the perineum (20%). Associated urogenital anomalies were seen in 4 (13.33%) cases. Other anomalies included Meckel's diverticulum (6.66%), Pouch colon (3.33%), bi cornuate uterus (3.33%) hypospadias with meatal stenosis (3.33%), undescended testes (3.33%), Inguinal hernia (3.33%), bilateral choanal atresia (3.33%), band at ileo caecal region (3.33%) and terminal ileum opening in caecum, attached to recto sigmoid region (3.33%). The association of fistula with vestibule, vagina and urethra was seen in 16.66% of patients. In 5(16.66%) patients associated fistulae were seen which included rectovestibular in 3(10%), rectovaginal and rectourethral fistulae in 1(3.33%) each.

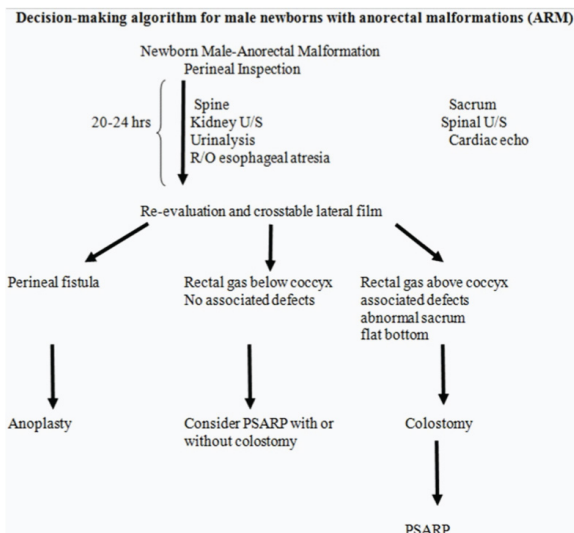
The type of surgery done is shown in [Table/Fig-6]. Single stage repair was done in 12 (40%) and multi stage repair was done in 18(60%). Type of approach for definitive repair is shown in [Table/Fig-7] and the outcome of surgery in [Table/Fig-8]. Complications of

Major clinical groups	Perineal (cutaneous) fistula Rectourethral fistula Bulbar Prostatic Rectovesical fistula Vestibular fistula Cloaca No fistula Anal stenosis
Rare/Regional variants	Pouch colon Rectal atresia/stenosis Rectovaginal fistula H type fistula Others

[Table/Fig-1]: Krickbeck international classification 2005 [2].



[Table/Fig-2]: The approach to male patients with ARM [3].



[Table/Fig-3]: The approach to female patients with ARM [3].

colostomy included excoriation in 2(6.66%), stenosis in 2(6.66%) and wound infection in 1(3.33%). Overall morbidity of the definitive procedure was 31.58% in present study. Overall mortality was 10.00% in present study. Follow-up was done for faecal continence and cosmesis.

DISCUSSION

Congenital anorectal malformations have been known since ancient times. It is commonest cause of neonatal intestinal obstruction. In the present prospective study

Type of Lesion	Male	Female	No. of Cases	%
Anorectal agenesis with rectourethral fistula	1	0	1	3.33
Anorectal agenesis with rectovaginal fistula	0	1	1	3.33
Anorectal agenesis without fistula	11	2	13	43.33
Anal agenesis with recto vestibular fistula	0	3	3	10
Anteposed anus with anal stenosis	2	5	7	23.33
Covered anus	1	1	2	6.66
Cloaca	0	3	3	10

[Table/Fig-4]: Type of anorectal malformation.

of 30 patients, 18 (60%) patients presented in neonatal period and 12 (40%) in post neonatal period as compared to the British Columbia Hospital study comprising 120 patients, in which 105 (87.50%) patients presented in neonatal period and 15 (12.50%) in post neonatal period [4]. Factors responsible for higher rate of delayed presentation in present study were lack of awareness, illiteracy, poverty and negligence of parents.

As shown in [Table/Fig-5], the male to female ratio of 1:1 was seen in present study as compared to study done by Chau, where it was 1.6:1. In both studies, the incidence of high variety anorectal malformation was more common than other varieties [5]. Incidence of cloaca was 10% in present study and in Chau series, it was 1.85%. Incidence of low varieties was nearly equal in both studies [5].

The common presenting symptoms in the present study were: not passing meconium since birth (50%), absent or abnormal anal opening, vomiting (13.33%) with increasing abdominal distension (26.66%), excessive crying and passing of thin pipe stools with constipation (23.33%), passing stools through vagina (13.33%) or urethra (3.33%) or an abnormal opening elsewhere in the perineum (20%). Non passage of meconium per anus since birth (76%), abdominal distension (100%), constipation (70%), vomiting (20%) and passing of stools per vagina (3.70%) were presenting symptoms in a similar study done in Calabar, Nigeria [6].

Anorectal agenesis without fistula was the most common type of anorectal malformation (43.33%) in present study and was more common in males as compared to females. Anteposed anus and anal

Level of Lesion	Present Study				Chau Series [4]			
	Male	Female	n	%	Male	Female	n	%
High	12	2	14	46.66	25	14	39	36.11
Intermediate	-	4	4	13.33	19	13	32	29.62
Low	3	6	9	30	22	13	35	32.40
Cloaca	0	3	3	10	0	2	2	1.85
Total	15	15	30		66	42	108	
Percentage (%)	50	50		100	61.11	38.88		100

[Table/Fig-5]: Sex incidence and level of lesion.

stenosis was present in 23.33% patients. Cloaca was seen in 10% patients and recto-vestibular fistula seen in 10% cases [Table/Fig-4]. In the present study, 13.33% cases were associated with urogenital anomalies and 9.99% were associated with gastro intestinal anomalies like Meckel's diverticulum, pouch colon, terminal ileum attached to rectosigmoid junction and band at ileocaecal junction. Also, 9.99% cases were associated with bicornuate uterus, hypospadias, undescendent testis, inguinal hernia and bilateral choanal atresia. No case was detected with vertebral, cardiac anomalies or trachea-esophageal fistula in present study.

In a study by Simmi K. Rattan, ARMs were associated with urogenital anomalies (39%), gastrointestinal anomalies (9%) and vertebral (28%), cardiac (10%) and trachea-esophageal fistula (7%). Only 4% cases were associated with other anomalies [7].

In a study by Kumar V and colleagues, associated Esophageal Atresia and Tracheoesophageal Fistula (TEF) with left amastia and multiple congenital anomalies was seen in association with high variety of anorectal malformation and hypospadias. This case was successfully managed b two staged operations to tackle the various associated congenital anomalies [8].

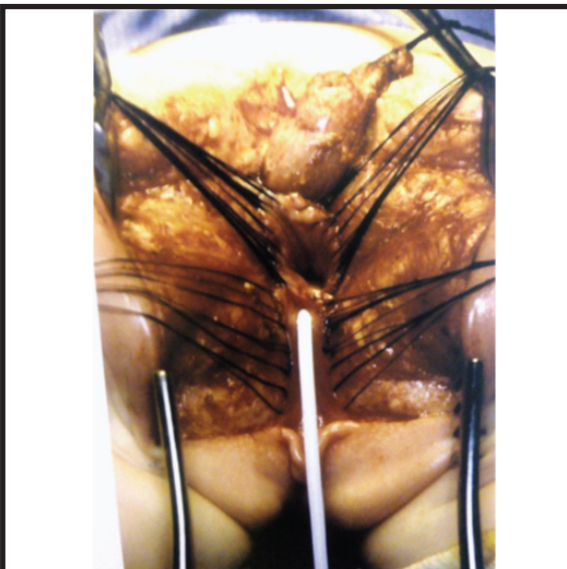
Kava MP and colleagues from Mumbai, India reported that 5% of patients with Down's syndrome suffer from high ARM [9].

In present study, most of associated congenital anomalies were seen in high variety of ARM (20%). Similar findings have been reported by other authors [7].

In 5 (16.66%) patients associated fistulae were seen which included rectovestibular in 3(10%), rectovaginal and rectourethral fistulae in 1(3.33%) each. In a study

Type of Colostomy/ Heostomy	Present Study (n=30)					Heinen FL (n=227) [10]				
	High	Intermediate	Low	Cloaca	%	High	Intermediate	Low	Cloaca	%
Sigmoid loop	13	1	0	1	50	40	25	4	3	31.7
Transverse loop	1	0	0	0	3.33	87	5	0	0	40.52
Pouch colostomy	0	0	0	1	3.33	0	0	0	0	0
Ileostomy	0	0	0	1	3.33	0	0	0	0	0
Percentage	46.66	3.33	0	10	59.99	55.94	13.21	1.76	1.32	72.33

[Table/Fig-6]: Type of Colostomy / Ileostomy done



[Table/Fig-7]: Posterior sagittal anorectoplasty- intraoperative photograph.

by Sanchez Martin et al., 39.99% of patients had associated fistulas. Rectovaginal fistula was present in 33.33% cases, anovestibular fistula in 2.50% cases and rectovestibular fistula in 1.66% patients of anorectal malformations [10]. In present study no case of fistula was present in low variety but in Sanchez Martin series, 8.33% were seen in low variety of ARM [10].

In a study by B. Mirza of 100 patients with ARM, 66% underwent sigmoid loop colostomy, 18% patients underwent anooplasty and exploratory laparotomy was performed in 8% patients. About 4% patients of stenosed vestibular fistula were managed by dilatation while patients with complex malformations (3%) were counseled for surgical intervention at a later date [11]. Heinen FL has done a study of 227 patients of anorectal malformations. He treated patients with PSARP; 72.23% patients underwent colostomy out of which 40.52% underwent transverse loop colostomy and 31.71 underwent sigmoid loop colostomy followed

Level of Leison	Present Study (n=30)					Chau Series (n= 90) [4]				
	PS ARP	PSARP with Abdomino Perincol pull through	AS ARP	Cut back Anooplasty	Anal Dilatation	PS ARP	PSARP with Abdomino Perincol pull-through	AS ARP	Cut Back Anooplasty	Anal Dilatation
High	4	1	0	0	0	37	2	0	0	0
Intermediate	3	0	1	0	0	32	0	0	0	0
Low	1	0	1	5	2	0	0	0	30	5
Cloaca	1	0	0	0	0	2	0	0	0	0
Precentage	30.00	3.33	6.66	16.66	6.66	65.74	1.85	0	27.77	4.62

[Table/Fig-8]: Type of approach for definitive repair.

by PSARP after two months [12]. In present study 50.00% cases underwent sigmoid loop colostomy, transverse loop colostomy was done in 3.33%. Pouch colostomy and ileostomy was done in two cases of cloaca because of associated congenital anomalies [Table/Fig-6].

Chowdhary SK et al., had carried out a retrospective study of 50 newborns with congenital anorectal malformations at Chandigarh, Punjab, India. They concluded that sick, small and septic babies arriving late to the unit do not appear to tolerate general anesthesia and divided sigmoid colostomy well. However, the procedure is advantageous in the long run. Divided sigmoid colostomy has excellent results in babies more than 2.5kg weight but in context of the developing world and limited critical care availability transverse loop colostomy under local anaesthesia may save lives [13].

In a study by Mirza in 100 patients with ARM, 15% developed post operative complications after colostomy which included wound infection in 10, pericostomy intestinal evisceration in two and gangrene and colostomy retraction in one patient each. Four patients had to be re-operated for these complications [11]. In another study by Figueroa M in 185 patients, colostomy complications included retraction (7 patients), Prolapse (7 patients), closure of distal opening (5 patients) proximal stenosis (3 patients), ostomy necrosis (1 patient) parastomal hernia (2 patients) [14]. In present study 43.33% developed no complication, skin excoriation in 6.66% stenosis in 6.66% and wound infection observed in one patient. In our study prolapse and retraction of colostomy were not seen. All the complications were treated conservatively in present study.

In present study all the high variety anorectal malformations and cloaca patients were treated with multi stage strategy. All the low variety of anorectal malformation cases treated in single stage. In intermediate variety three patients treated with single stage strategy and one patient treated with multi stage strategy. So 40% patients were treated with single stage strategy and 60% with multistage strategy [Table/Fig-7]. A study on one stage correction of intermediate imperforate anus in males in Nigeria was done in 15 patients. Two patients were subjected to diverting colostomy due to septicaemia. Rest underwent PSARP. The results of this study showed that males with intermediate anus can have a safe PSARP without needing colostomy. In developing countries, this is of importance since having a single surgery has many advantages over having three surgeries. Results in high variety are poor with single stage repair even with best care, so they should be treated by multistage repair for favorable results [15]. In a study done by Nagdeve in 12 males with high ARM with a well descended rectum, primary repair was done. The patients were found to have good continence without significant morbidity. However, its preference over multistage repair depends

on long term anorectal function, which could not be assessed immediately following surgery [16]

In present study 68.42% patients had good results, 10.52% patients had wound infection; one patient developed both anal stenosis and urinary incontinence, which were corrected by doing PSARP. One cloaca patient having rectourethral fistula awaits surgery. Results of surgery in intermediate and low variety were good.

In terms of definitive procedure, in present study, in 30.00% patients, PSARP was done, in 3.33% cases PSARP was combined with abdominoperineal pull through. ASARP was done in 6.66% patients, cut back anoplasty in 16.66% and anal dilatation done in 6.66% cases. In Chau series of 108 patients of imperforate anus, PSARP was done in 65.74% patients, PSARP with pull through in 1.85%, cut back anoplasty in 27.77% and anal dilatation in 4.62% patients [5]. [Table/Fig-8].

In a study done by Nam SH on 311 patients with ARM, 84.8% patients had favourable outcome i.e. showed voluntary bowel movements; 30.7% had constipation and 6.5% showed soiling. 82.2% of children had good continence, 2.7% showed fair continence, and 15.2% showed poor continence. [17].

In a study done in Japan, it was concluded that a secondary operation through a posterior sagittal approach can be performed without a diverting colostomy and restore fecal continence in adolescents and adults [18].

In the present study mortality incidence was 10% , seen in high variety of ARM and cloaca. In a study by Poley et al., in 286 patients, mortality incidence was 11.27%, also observed in high variety of ARM. It was concluded that morbidity and mortality was highest in the youngest group. However, the prognosis was deemed to be favourable for majority of the survivors of surgery. This information can be used to reassure parents of patients [19].

In the present study we observed that low varieties of anorectal malformation do not require defunctioning colostomy and the primary surgery should be done at the time of presentation. They have good faecal continence post-operatively provided the perineal muscles and rectum not injured during surgery and major infection does not occur. High and intermediate varieties need primary defunctioning colostomy at the time of presentation followed by definitive repair at the later date when weight of child appropriate. In majority of patient's definitive repair can be done by PSARP, ASARP PSARP combined with abdominal pull through approach. Primary PSARP can be tried in rectovestibular, rectovaginal fistula and in some low varieties of ARM. Excoriation of skin after defunctioning colostomy can occur should be prevented with good hygiene, zinc oxide paste, wooden paints applied on the abdominal skin surrounding the stoma and using colostomy bags.

After the definitive repair colostomy closure is done after 4-6 weeks of regular anal dilatation to prevent stenosis. Regular follow-up is required. However, better anaesthetic management, preoperative preparation, intra operative care, post operative care, fluid and electrolytic balance as well as care in paediatric ICU lower down the mortality and morbidity of colostomy as well as of definitive procedures and colostomy closure.

LIMITATION OF STUDY

The present study suffered from the following limitations:

- Small sample size
- Short follow-up period.

Similar studies with longer follow-up periods and bigger sample sizes are needed to shed more light on the plight of neonates who suffer from ARM and the effect of corrective surgery on their quality of life.

CONCLUSION

Low varieties of anorectal malformation do not require defunctioning colostomy and the primary surgery should be done at the time of presentation while in high and intermediate varieties of ARM, definitive repair can be done by PSARP, ASARP PSARP combined with abdominal pull-through approach.

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