Outcome and Complications of Anorectal Malformations

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Abstract: Anorectal Malformation [ARM] is a relatively common, complex anomaly to treat, improvement in management has improved life style of ARM child. We have studied every case of ARM admitted in GMKMCH between January 2012 and December 2016 (total 81). Children admitted are first stabilized and examined clinically after 24 hours subjected to invertogram and treated according to type of anomaly. Male babies with low anomaly are treated with anoplasty. Female babies are subjected to posterior trans position of anus at 3 months of age. High and intermediate anomalies are treated with 3 stage procedures. It was found that ARM is common in low socio economic group with equal gender incidence. Half of the cases are associated with other anomalies which is most common cause of death. Laparoscopic Assisted Anorectoplasty [LAARP] is better for high ARM and Posterior Sagital Anorectoplasty [PSARP] for intermediate ARM. Functional outcome of low ARM is good compared to high and intermediate ARM

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I. Introduction

ARM is a relatively common congenital anomaly that we come across in our pediatric surgery practice. The severity of the lesions varies from fairly minor lesions (e.g. Covered Anus) to most complex abnormalities such as Cloaca Exstrophy and Rectal atresia. ARM forms a significant load in developing countries, not only in the emergency situation but also in terms of long-term corrective procedures. Although there have been major advances in the management of these children during the last 15 years, these patients still represent a continuing challenge as a result of the significant reconstructive problems involved, as well as the fact that a significant number suffer from faecal and urinary incontinence, as well as the possibility of inadequacies - sexual, non correctable defects, not to talk of the associated anomalies in later life. With development in the Surgical Specialities the management has improved and enable the children to lead normal livelihood, and we now see patients who have married and borne children with a normal life span.

II. Materials and Method

Every case of Anorectal malformation admitted in newborn unit of Govt Mohankumaramangalam Medical College Hospital(GMKMCH), salem during the period from January 2012 to December 2016(90 patients) and completed all the stages of surgical procedures with in this period and with the follow up of lyear.All babies with ARM patients admitted are first stabilized and kept under observation for16 to 24 hours. The child is examined clinically If there is clinical evidence of perineal fistula, bucket handle deformity or mid line raphe fistula the child is subjected to Anoplasty. Child is followed with anal dilatation. If in first 24 hours if there is meconuria and flat bottom the child is subjected to colostomy. Then at 6 months child is taken for definitive procedure (PSARP) done. If no clinical evidence of type of anomaly then invertogram is taken at 16 to 24 hours. If on invertogram low ARM is diagnosed child is subjected to Anoplasty. If high or intermediate anomaly, child is subjected to colostomy and followed at 6 months with definitive procedure. In all cases Colostogram is taken before definitive procedure. After 3 months colostomy closure was done. In newborn female child, child is kept under observation for16 to 24 hours. The child is subjected to through clinical examination. If there is clinical evidence of cutaneous fistula, the child is subjected to Anoplasty. Child is followed with anal dilatation. In case of anovestibular fistula or vulvar anus/fistula posterior transposition is done at 6 months and followed with anal dilatation. In case of cloacal anomaly, colostomy is done, followed by definitive procedure Posterior Sagital Anorectovagino uretroplasty (PSARVUP) at 6 months. If no clinical evidence of fistula then invertogram is taken at 16 to 24 hours. If on invertogram low ARM is diagnosed child is subjected to Anoplasty. If high or intermediate anomaly, child is subjected to colostomy and followed at 6 months with definitive procedure. In all cases associated anomalies are diagnosed during first month of life by subjecting to investigations. Outcomes measured in terms of mortality, morbidity and complications.

Table - 1 : Socio-economic status					
	Our study A. E. Archibong, et al. ¹				
Income	Less than Rs.	Mores than Rs.	Less than Rs.	More than Rs.	
Group	12000/annum	12000/annum	12000/annum	12000/annum	
Number of	81	-	125	30	
patients					

III. Results				
'ahle	_	1		Socio-economic status

The incidence of ARM in affluent patients with income group >1200 is very meager and practically nil. we understand from our senior colleagues who practice outside that ARM predominantly affects lower socioeconomic status patients. According to A. E. Archibong low socio economic group patients had higher incidence of ARM compared to affluent patients.

Table - 2: Incidence				
	1 ST YEAR	2 ND YEAR	3 RD YEAR	
Number of Cases of ARM	33	27	21	

The incidence of ARM is decreasing. Previously in the yester years we were having a heavy workload because of these congenital diseases. Now, the number of cases has come down. Reasons probably may be due to the better nutritional status and better ante natal care which our people enjoy. Further the Ultra Sound may help in discerning these afflicted babies leading to spurning of such fetuses which suffer from malformation disorders. This is reflected in this study as evinced by the decreasing incidence noted through the years.

Table –3: Sex Ratio					
Gender	Stephens ⁴	Endo et al ⁵	Our study		
Male	57%	57%	54%		
Female	43%	43%	46%		

In our study, the male : female ratio associated with ARM is almost equal, with a 54 : 46 male : female ratio. Our results are almost similar to the study of Stephens and Endo et al. In these study also male female ratio is almost equal. There is no difference in incidence of ARM in both the sex group even though certain type of ARM is exclusive for either male or female gender.

Table – 4. Trequency of type of ARM					
Authors	High %	Intermediate%	Low %		
Our study	27	24	49		
$Cook^6$	28	23	51		
Stephens ²	46		54		
Chen ⁷	20	47	33		
Endo et al⁵	26	11	57		

Table – 4: Frequency of type of ARM

In our study Low anomaly is most common occurring anomaly followed by High and Intermediate anomaly, which is also shown by other studies – Cook et al and Endo et al. Difference in incidence shown by Chen et al and Stephen et al is because of difference in classification of ARM used by them. We used International Classification of ARM.

 Table- 5 : Anomalies Associated with Anorectal malformation

	Our study	Ratam ⁸	Smith ³	Kiesewetter ⁹
Anomalies %	52	58	61	54
Vertebral / Skeletal	42	41	26	6
Cardio vascular	22	10	9	7
EA / TEF	6	6	4	9
GIT	6	9	8	4
GU	34	39	25	40
Genitalia	8			
Downs	2			

Almost half of the cases with ARM has associated anomalies (52%) and it is the most common cause of death in ARM patients (5 out of 8 mortalities in our study). It is recommended that all patients with anorectal malformations should have all necessary investigations to search the associated anomalies different systems. Urinary anomalies were high in both sexes in high ARM. Patients with urogenital anomalies require careful assessment and timely intervention for better out come. However large number of patients and poor

primary health care services make us confine to do basic investigations rather than follow a protocol. So we routinely do Ultrasonography of abdomen, X-ray spine of all patients, echocardiogram and neurosonogram. Special investigations for example: intravenous Urography, MCUG and fistulogram are done in appropriated cases. Actual incidence of urogenital anomalies may be higher if thoroughly investigated.

	Invertogram	Operative findings
High	22	17
Intermediate	11	12
Low	4	8
Total	37	37

Table- 6: Correlation between invertogram and operative finding

The overall sensitivity of invertogram in detecting type of anomaly is low. In our study out of 22 cases diagnosed as high by invertogram 17 were confirmed to be high, four were low and one intermediate, totaling five. In these five cases, four of them would have been subjected to colostomy because of the false positive the Invertogram. So, we would like to stress, that though Invertogram is being done as a routine, clinical assessment is the ultimate parameter for judgment. Cases with epithelisal pearls, bucket handle deformity are pathognomic of low anomaly and are treated with perineal exploration irrespective of Invertogram findings. Surasak Sangkhathat suggested high sensitivity and specificity with MRI. So through clinical examination is needed in case of doubte MRI for diagnosis of type of ARM (e.g.; avoids 3 staged procedure for misdiagnosed Low type ARM).

	Prove		
No.		Pelvic colostomy	Transverse colostomy
			-
		%	%
1.	Skin excoriation	19.3	30
2.	Prolapse	12	17.5
3.	Bleeding	7.6	17.7
4.	Obstruction	-	11.5
5.	Wound infection	5.3	-
6.	Retraction	3.3	5.2
7.	Redo	4.3	5.2
8.	Mortality	-	10

 Table – 7: Comparison between Pelvic and Transverse colostomy

The common complication of colostomy is skin excoriation 19.3% in pelvic colostomy and 30 % in transverse colostomy. Similarly all the other complications such as Prolapse, Bleeding, Obstruction, Wound Infection, Retraction, and Mortality are more in with Transverse colostomy than with Pelvic colostomy. In addition to the above complications Transverse colostomy have additional complications such as electrolyte imbalance and malnourishment problems. This show that Pelvic colostomy is better option when compared to Transverse colostomy which was also in accordance with study by Chandramouli .Sigmoid colostomy should be performed whenever possible except in situations of very high anomaly where surgeon suspects that distal bowel won't be sufficient for further pull through procedures. Close attention to technical details, principles of stomal care, and proper parental instruction should minimize morbidity. Concluding that while transverse colostomy is surgeon friendly for subsequent procedures, pelvic colostomy is patient friendly for maintenance and lesser complication rate.

 Table – 8:
 Complication of colostomy closure

	Our study	Chandraemouli ¹⁰		
Wound infection	12.2	12.6		
Anastomotic leak	4.2	7.1		
Adhesive obstruction	4.2	5.2		
Stitch granuloma	12	10.5		
Incisional hernia	2.2	2.6		
Mortality	-	1.8		

In our study wound infection occurred in 6 cases(12.2%). Incisional hernia in 1 case in which wound infection was very severe. Anastomotic leak occurred in 2 cases and were managed conservatively. Adhesive obstructions in 2 cases were also managed conservatively. All this showed that early colostomy closure reduces morbidity and mortality of colostomy. Hence, it is recommended that Post PSARP patients should have their colostomy closure within three months to obviate complications of the pulled through bowel. Frequently we see

patients coming years after the primary procedure. On our part we should motivate and give dates with in three months for the patients at the time of discharge after PSARP.

	Our s	study	C. Devos, M. Arnold et al ¹¹	
Complications	Post sagittal	LAARP	Post sagittal	LAARP
	approach		approach	
Wound sepsis	0	1	0	2
Wound dehiscence	2	0	2	0
Retractions	2	1	1	1
Faecal fistula	1	0	1	0
Rectal stricture	0	1	0	1
Anal stenosis	1	2	1	3
Mucosal prolapse	3	1	3	2
Redo Anorectoplasty	3	1	3	0
Redo Anoplasty	1	2	1	0
Laparotomy needed	11			
Continence				
Good	46	47	48	43
Fair	33	32	30	30
Poor	21	21	30	25

p. Assisted approaches
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A comparison of laparoscopic assisted (LAARP) and posterior sagittal (PSARP) anorectoplasty in the outcome of intermediate and high anorectal malformation showed no significant difference in complications of assisted (LAARP) and posterior sagittal (PSARP) anorectoplasty but have specific associated problems. The increased association of anal stenosis in the LAARP procedure might be due to the fact that the perineum is not as extensively opened as in PSARP, leading to a smaller fashioned anoplasty. However, PSARP group showed a high number of patients needing management for both prolapse. Although a long 'learning curve', with laparoscopic surgical techniques, extending to all participating staff and even equipment maintenance. Both the LAARP and PSARP procedures can successfully treat ARM with comparable outcomes. It appears that LAARP is optimal for high ARMs that would otherwise require a laparotomy to facilitate adequate mobilization.We suggest that were sacro abdominal pull through is contemplated a lap assisted PSARP would be of value as it obviates the need for laparotomy and it is not a very technically demanding procedure through the laparoscope.

		0 1	1	11
	Post sagittal	LAARP	Chi square value	
	approach			
Wound sepsis	0	1	0.663	Not Significant
Wound dehiscence	2	0	0.961	Not Significant
Retractions	2	1	0.663	Not Significant
Faecal fistula	1	0	0.604	Not Significant
Rectal stricture	0	1	0.663	Not Significant
Anal stenosis	1	2	0.469	Not Significant
Mucosal prolapsed	3	1	0.645	Not Significant
Redo Anorectoplasty	3	1	0.645	Not Significant
Redo Anoplasty	1	2	0.469	Not Significant
Laparotomy needed	11	0	0.122	Not Significant
Continence				
Good	46	47		
Fair	33	32		
Poor	21	21		

 Table – 10 : Complications of posterior sagittal approach & Lap. Assisted approach

Table – 11: Functional Outcome of High Ano rectal malformation	S
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Kelly Score	Good	Fair	Poor
Our Study	46	33	21
Stephen and Smith ²	56	32	12
Trustler & Willkinson ¹²	26	20	54
Partridge and Gough ¹³	33	43	24
Taylor ¹⁴	24	20	56

The continence scoring of our procedure (Posterior sagittal approach) for high and intermediate ARM is better compared to other traditional procedures studied in by different authors Trustler & Willkinson, Partridge and Gough and Taylor.

1 uble 12. 1 unchonal o'alcome of Low The rectal marjor mations				
Kelly Score	Good	Fair	Poor	
Our Study	84	14	2	
Stephen and Smith ²	83	15	2	
Trustler & Willkinson ¹²	80	20	-	
Partridge and Gough ¹³	86	11	3	

 Table – 12 : Functional Outcome of Low Ano rectal malformations

The functional outcome patients treated with low ARM is good. This is also substantiated by other studies by Stephen and smith, Partridge and Gough, Trusteler and Willkinson. This is due to less complexity of the defect and good sphincter muscle complex development. 5 patients had fair out come out of which 4 did not turned up for regular dilatation. 1` patient had anal stenosis due to ischemia for which redo surgery was done with poor out come. A typical problem in treatment of low anomaly is anal stenosis which can be prevented by regular dilatations.

Table - 13 :Mortality		
Causes	Number of patients	
Colostomy	2	
Defining procedure	1	
Associated Anomalies	5	
Total	8	

The most common cause of death in ARM patients is Associated Anomalies. Severe forms of anomalies are associated more often with high ARM). It is recommended that all patients with anorectal malformations should have all necessary investigations to search the associated anomalies different systems. Next common cause of death in our study is due to colostomy. These cases presented very late and had a morbid pre-operative picture itself. Early colostomy closure reduces morbidity and mortality of colostomy

IV. Discussion

We have studied the present series of 90 patients who were admitted with ARM and underwent various surgical procedures. Analyzing them, the following summary were drawn

1) All the patients where in our study were low socio-economic status. The incidence of ARM in affluent patients with income group >1200 is very meager and practically nil 2) The male: female ratio associated with ARM is almost equal, with a 54:46 male: female ratio.3) Low anomaly is common in females and high and intermediate anomaly in males. 4) Almost half of the cases with ARM has associated anomalies (52%) and it is the most common cause of death in ARM patients (5 out of 8 mortalities in our study).Off which most common association is Vertebral/ skeletal anomalies(42%) followed by Urological anomalies(34%). It is these associations which decide the prognosis rather than the lines of management.5) The overall sensitivity of invertogram in detecting type of anomaly is low. Invertogram is done as a matter of routine but should not be taken as a fool proof investigation. Clinical determinants are the deciding norms. 6) Pelvic colostomy is better option when compared to Transverse colostomy. Sigmoid colostomy should be performed whenever possible except in situations of very high anomaly where surgeon suspect that distal bowel would not be sufficient for further pull through procedures.7) Early colostomy closure reduces complications. Frequently we see the proximal bowel is dilated and the distal loop has a very small lumen which results to a anastomosis like end on back, as done in Atresias. This is due to disuse atrophy of the distal bowel. Hence a plea is made for early closure of the colostomy after PSARP.8) Both the LAARP and PSARP procedures can successfully treat ARM with comparable outcomes. It appears that LAARP is optimal for high ARMs that would otherwise require a laparotomy to facilitate adequate mobilization.9) The functional results of posterior sagittal approach is better than traditional procedures for High and Intermediate anomalies.10) The functional outcome patients treated with low ARM is excellent.11) The most common cause of death in ARM patients is Associated Anomalies.

V. Conclusion

The outcome of treatment of ARM has improved, tremendously in the present scenario. What was previously a write off are now becoming correctable and these patients lead a normal life, enter adult hood, and bear children. This is because of improvement in the infra structure, availability of adequate health resources, improvement in techniques and investigation modalities. Further, understanding the complex anatomy of ARM and associated anomalies and early and appropriate surgical treatment, emphatically reduces the mortality and morbidity. The Posterior Sagittal Approach has improved functional outcome of treatment of high and intermediate ARM.Surgical procedures have become standardized and PSARP has become the state of the art procedure. Laparoscopy has become a tool which also helps in the high Supra Levator anomalies, giving equally good if not better results. Complex Cloacal anomalies are now correctable technically, which was previously not possible.Gross defects with associated anomalies, are bearing the crux of the mortality. Though

ARM can be corrected the other anomalies do the patient in. Corrections are now possible at an earlier date, and the patients are fully corrected before the school going age.

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