Original Article

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Comparison of MRI with Distal Colostogram in Patients with Anorectal Malformations- A Cohort Study

Radiology Section

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ABSTRACT

Introduction: Anorectal Malformations (ARMs) are the second most common paediatric surgical emergencies with management strategy is restoration of near normal anatomy and physiology which require accurate and proper imaging diagnosis.

Aim: To evaluate Magnetic Resonance Imaging (MRI) findings and its comparison with Distal Colostogram (DC) against surgical findings in anorectal malformation patients.

Materials and Methods: A cohort study was done at two centers Jawaharlal Nehru Medical College, Aligarh (JNMC, Aligarh) and Vardhman Mahavir Medical College (VMMC) and Safdarjung Hospital, New Delhi involving 60 patients of ARMs. MRI was done in all patients and distal colostogram was done in 46 (74.4%) patients. All patients were evaluated for associated anomalies. Both the imaging modalities were compared for the type of ARM and fistula against the surgical findings and Chi-square and Fisher's test were used to calculate statistical significance.

Results: Out of total 60 patients included in this study, there were 37 (61.7%) males and 23 females (38.3%). High ARM was seen in 34 (56.7%) patients and low ARM was seen in 26 (43.3%). There was no significant difference between distal colostogram and MRI in differentiating the type of ARM (p-value 0.13). Spinal anomalies (52.5%) were the most common associated anomalies, followed by genitourinary (47.5%) and musculoskeletal (32.5%).

Conclusion: From the results of present study no significant difference was found between DC and MRI findings regarding the type of ARM.

Keywords: Associated anomalies, Magnetic resonance imaging, Paediatric surgical emergency

INTRODUCTION

Anorectal Malformation (ARM) are the second most common paediatric surgical emergencies with incidence ranging from 1 in 2000 to 1 in 5000 [1]. Antenatal diagnosis of an isolated ARM is rare. Most cases present in neonatal period. Embryologic basis lies in the faulty development of the terminal portion of the hindgut in the early embryonic period giving rise to both urinary and gastrointestinal system anomalies [2].

Classification of anorectal malformation has evolved with time, increasing embryological understanding and evolved surgical practices. Current management strategy focuses on restoration of normal anatomy and bowel, bladder and sexual activities. ARM was classified according to the level of the arrest of rectal descent and patient's sex [3]. Devries PA and Pena A proposed new classification after their own experience with Posterior Sagittal Anorectoplasty (PSARP) and realised that presence and site of fistula has an important bearing on the long-term outcome [4]. The currently used a new classification system incorporated criteria from both classification with three distinct diagnostic, surgical procedure, and functional outcome criteria categories [5].

Approximately 40-70% of ARM has associated lesions guiding surgical strategy and suggesting prognosis, most common being genito-urinary and skeletal anomalies. Spinal cord malformations are present in 30-50%, more frequent in lumbo-sacral region [6-9].

Diagnostic assessment is directed towards identifying level of blind pouch, any fistulous communication and associated anomalies. Clinical examination alone is inadequate to answer all questions, so here is the role of imaging evaluation. Imaging studies performed in the first two days of life include radiography of the thorax, spine, and pelvis along with cardiac, perineal, abdominal, pelvic, and spine ultrasound to detect possible associated anomalies [10]. Invertogram the earliest imaging technique used for assessment of rectal pouch is highly inaccurate and is not performed anymore [6,11,12]. High-pressure distal colostography very well demonstrates level of rectal pouch and fistulas [10] .Computed Tomography (CT) enables accurate assessment of the level of lesion, fistulas and associated anomalies, but poor soft tissue characterisation and radiation limits role in ARM [13].

Magnetic Resonance Imaging (MRI) with excellent soft tissue resolution was traditionally reserved for complex anorectal malformation with inconclusive conventional findings, however routine MRI provides more holistic information on type of malformation, extent of pelvic musculature and sphincter complex and skeletal status of lumbosacral spine required for management [11].

The present study aims to address limited literature on important role MRI can play in anorectal malformation. It evaluates role of MRI in ARM and compare various aspects of anorectal malformation like level, presence of fistula between conventional imaging distal colostogram and MRI against postoperative findings and assessment of associated anomalies and pelvic floor musculature as a guide to prognostic outcome.

MATERIALS AND METHODS

A cohort study was conducted in Jawaharlal Nehru Medical College, Aligarh (JNMC, Aligarh) and Vardhman Mahavir Medical College (VMMC) and Safdarjung Hospital, New Delhi during the period extending from January 2013 to October 2018 with the approval of Institutional Ethical Committee. Prior written informed consent was taken from the parents/guardians of the patient.

Inclusion criteria: All the patients with ARM both new and follow-up with interval surgery done at other institution came to our institution for definitive surgery were enrolled in the study ruling out exclusion criteria.

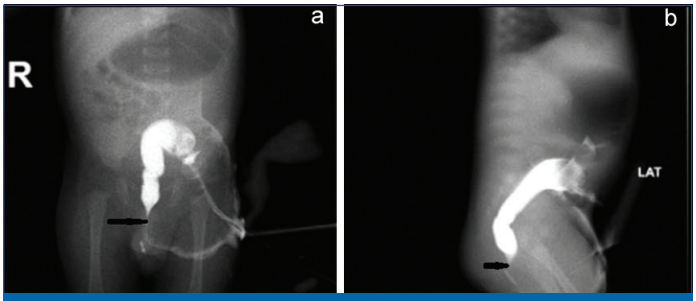
Exclusion criteria: Patient not willing to participate in the study, critical patients or one with severe malformations were excluded from the study.

Total study patients were 60. Age of the patients ranged from <24 hours to five years. After detailed clinical history, general physical

examination and local examination of the perineum, all the patients were subjected to prone cross table lateral view x-ray and pelvic MRI. Operative follow-up was available for all the patients. Distal colostogram was done in 46/60 patients who underwent colostomy as rest of the patients underwent primary definitive surgery. Diagnostic accuracy of colostogram was evaluated against 46 patients while for MRI it was 60.

Distal Colostography

Technique: Radio-opaque marker was placed at the expected position of anus and water soluble contrast was injected into pouch through the catheter placed in the stoma and fluoroscopic projections were taken. Study was evaluated for the level of blind pouch, fistulous communications [Table/Fig-1a,b].



[Table/Fig-1]: Distal colostogram anteroposterior; a) and lateral; b) Images of 2 month old child shows rectal atresia with severe narrowing of distal 1/3rd of rectum (arrows). No evidence of any fistulous communication with the genitourinary tract.

MRI Studies

Scanning methods and parameters: The MRI pelvis was done with 1.5 T Phillips achiva scanner with patient placed in supine position. Coil selection was done as per body sizes.

Regarding MRI acquisition and evaluation of ARMs, two important reference transverse planes were used, one was the pubococcygeal plane extending from the upper border of the os pubis to the os coccyx corresponding to the attachment level of the levator ani muscle to the pelvic wall and second reference transverse planes was line joining the lowest points of the ischial tuberosities (I line) representing deepest level of the funnel formed by levator ani muscles.

Axial scans were acquired parallel to the pubococcygeal line, whereas coronal scans were acquired perpendicular to this plane. Coronal and sagittal planes also included kidneys and lumbosacral spinal cord. Sequences acquired were T2W in all the three planes, T1W axial planes and T2STIR in sagittal plane. No contrast administration was done.

Image analysis: The MRI scans were evaluated by two radiologists, each having more than five years of experience with respect to level of blind pouch, fistulas, and associated spinal and urogenital system abnormalities.

Rectal pouch lying at or above the level of the puborectal sling was considered an intermediate or high type of ARM whereas below it was low ARM regardless of presence or absence of fistula [10]. Fistulas were identified with linear T2 hyperintensity with homogeneous wall and absent central hyperintense mucosa.

Muscle developmental index: Puborectalis (PR) muscles width and external anal sphincter muscle complex development was graded subjectively good if comparable to normal subjects, fair if muscle identified but inadequately developed or poor if barely identifiable [14].

Objective assessment was done by calculating relative width of puborectalis sling and external anal sphincter [14].

Relative width of Puborectalis (RWPR)=(Total width of Puborectalis (PR)/(half of "I" distance).

Relative Width of External Anal Sphincter Complex (RWEASC)= (Total width of External Anal Sphincter Complex (EASC)/(half of "I" distance).

Width was measured on axial plane at the level of ischial tuberosity on both right and left side (3 o'clock and 9 o'clock position) and it is the sum total of width of rectum and anal canal. I distance is the distance between inner margin of lower border of ischial tuberosity [14].

The RWPR <0.18 and RWEASC <0.15 were regarded as poor development by Tang ST et al., [15].

STATISTICAL ANALYSIS

Data collected was analyzed using Statistical Package for Social Sciences (SPSS) version 21. Continuous variables were expressed as mean±standard deviation. Qualitative data were expressed as percentage. Chi square and Fisher's test were used for univariate analysis. p-value <0.05 were considered to indicate statistical significance.

RESULTS

Out of 60 patients included in the study, 37 were males and 23 females. Most common clinical examination finding was meconuria in males while vestibular fistula in females [Table/Fig-2].

Males (n=37)		Females (n=23)	
Flat bottom	3 (8.1%)	Flat bottom	2 (8.6%)
Anal dimple	4 (10.8%)	Anal dimple	2 (8.6%)
Bucket handle	3 (8.1%)	Anal stenosis	2 (8.6%)
Meconuria	19 (51.35%)	Anteriorly placed anus	2 (8.6%)
Anal stenosis	3 (8.1%)	Cloaca	4 (17.3%)
Anal membrane	2 (5.4%)	Vestibular fistula	10 (43.4%)
No fistula	3 (8.1%)	Anal membrane	1 (4.3%)

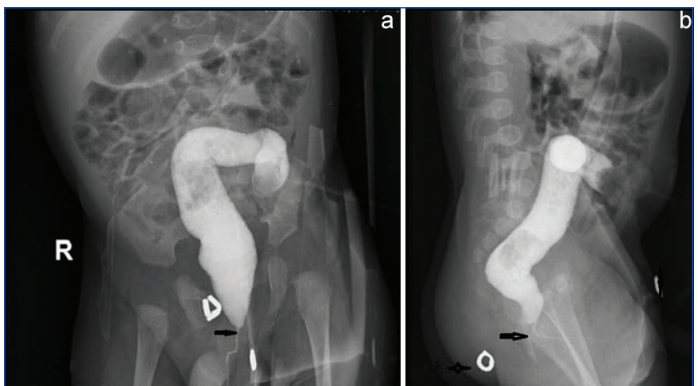
[Table/Fig-2]: Local perineal examination findings in the studied population.

Findings were comparable on MRI pelvis and distal colostogram when compared with surgical finding. (p-value 0.13) [Table/Fig-3].

Rectourethral fistula and vestibular fistula is the most common ARM in males and females respectively. When patients were classified according to krickenbeck classification on MRI pelvis, among males rectourethral fistula was seen in 16 out of 37 male patients (43%) making it most common anomaly [Table/Fig-4-7]. On comparison of diagnostic accuracy of distal colostogram and MRI pelvis with surgical findings regarding fistulous anatomy, MRI showed greater

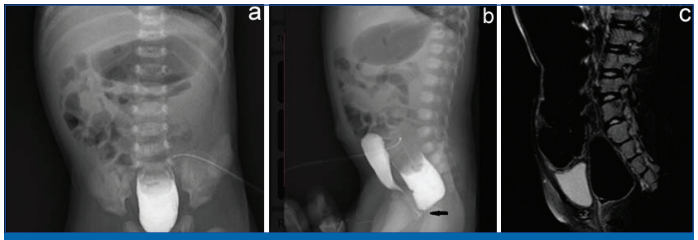
Type of ARM	MRI pelvis (N=60)		Distal colostogram (N=46)		Surgery (N=60)	
	Male (n=37)	Female (n=23)	Male (n=32)	Female (n=14)	Male (n=37)	Female (n=23)
High	29 (78%)	7 (30%)	25 (78%)	6 (43%)	27 (73%)	7 (30%)
Low	8 (22%)	16 (70%)	7 (22%)	8 (57%)	10 (27%)	16 (70%)

[Table/Fig-3]: Type of ARM on the basis of distal colostogram, MRI pelvis and operative outcome.

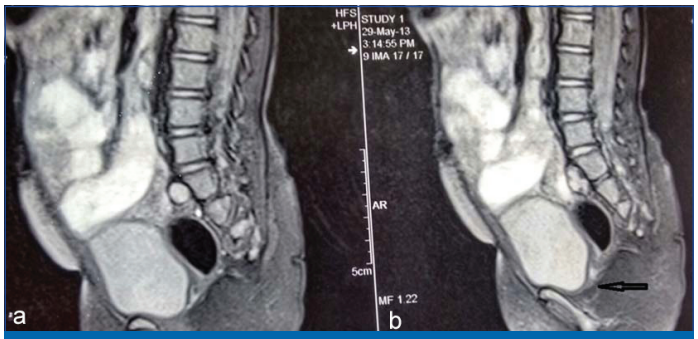


[Table/Fig-4]: Distal colostogram anteroposterior; a) and lateral; b) Images of 1.5 month old female child shows rectoperineal fistula (arrow) with rectum to seen open anterior to normal anal opening through a narrow tract. Radiopaque markers are placed at pubic symphysis and expected normal anal opening (star).

accuracy of 93% compared to 56.7% with distal colostogram. Associated anomalies were present in 66.7% of study population, increase prevalence was seen in patients with high ARM and in males [Table/Fig-8a-c].



[Table/Fig-5]: Distal colostogram anteroposterior (a) and lateral (b) Images of 18 day old child shows distal rectal and anal canal atresia with rectourethral fistula (arrow). Sagittal T2w images (c) of the same patient linear hyperintense tract extending between rectum and urethra (arrow).



[Table/Fig-6]: Sagittal T2w MRI images (a,b) shows rectal atresia with rectourethral fistula (arrow).

System of associated anomalies		Percentage (%)
Musculoskeletal system 13/40 (33.3%)	Facial deformity	3 (7.5%)
	Syndactyly	2 (5%)
	Congeital talipus equino varus(CTEV)	2 (5%)
	Cleft lip and palate	3 (7.5%)
	Others	3 (7.5%)
Spinal anomalies 21/40 (52.5%)	Spina bifida occulta	9 (22.5%)
	Tethered cord	4 (10%)
	Sacral agenesis	2 (5%)
	Lipomeningomyelocole	2 (5%)
	Others	4 (10%)
Genito-urinary system 19/40 (47.5%)	Hypospadias	4 (10%)
	Vesico-ureteric reflux(VUR)	3 (7.5%)
	Hydronephrosis	3 (7.5%)
	Undescended testis	2 (5%)
	Others	7 (17.5%)

[Table/Fig-9]: Type of associated anomalies in studied population.

findings for classification. The PR and EASC were assessed on pelvic MRI as it guides surgical management and incontinence in post-operative period.

Sex: There was male predominance in the present study with male to female ratio of 1.6:1 which is similar to studies done by Endo M et al., (1.5:1) and Cho S et al., (2:1) [16,17].

Local perineal examination: Most common presentation in male child was meconuria that was seen in 51.35% of the

Males (n=37)				Females (n=23)			
Variables	**MRI (n=37)**	**DC (n=32)**	**Surgery (n=37)**	**Variables**	**MRI (n=23)**	**DC (n=14)**	**Surgery (n=23)**
Perineal fistula	6 (16.2%)	3 (9%)	6 (16.2%)	Perineal fistula	2 (8.7%)	-	3 (13%)
Rectourethral fistula	16 (43.2%)	-	-	Vestibular fistula	12 (52.2%)	6 (42.8%)	12 (52.2%)
Prostatic	12 (32.4%)	-	13 (35.1%)	Cloaca	3 (13%)	3 (21.4%)	3 (13%)
Bulbar	4 (10.8%)	14 (43.7%)	4 (10.8%)	Anal stenosis	2 (8.6%)	-	1 (4.3%)
Rectovesical fistula	2 (5.4%)	2 (6.2%)	2 (5.4%)	No fistula	-	1 (7.1%)	-
Anal stenosis	2 (5.4%)	-	2 (5.4%)	-	-	-	
No fistula	3 (8.1%)	11 (34.4%)	2 (5.4%)	-	-	-	
Rare/Regional variants							
Pouch colon	6 (16.2%)	-	6 (16.2%)	Pouch colon	-	-	-
Rectal atresia/stenosis	2 (5.4%)	2 (6.2%)	2 (5.4%)	Rectal atresia/stenosis	-	-	-
				Rectovaginal fistula	4 (17.4%)	4 (28.6%)	4 (17.4%)

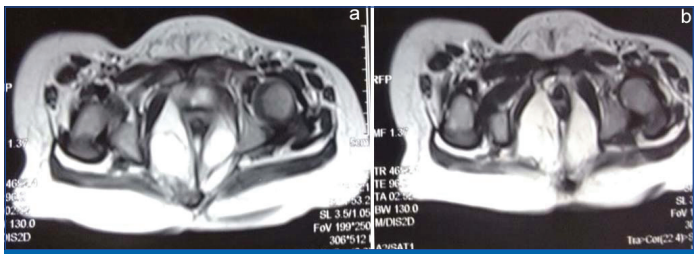
[Table/Fig-7]: Distribution of studied population according to Krickenbeck classification on the basis of MRI pelvis, distal colostogram (DC 46/60) and comparison with surgical findings.

Type of ARM	All cases with associated anomalies	Males	Females
High	26/34 (76.5%)	22/27 (81.5%)	4/7 (57.1%)
Low	14/26 (53.8%)	7/10 (70%)	7/16 (43.7%)
Total	40/60 (66.67%)	29/37 (78.4%)	11/23 (47.8%)

[Table/Fig-8]: Distribution of associated anomalies with ARM in relation with type and sex.

Among genitourinary anomalies most common being hypospadias (10%) followed by VUR and hydronephrosis (7.5%) each [Table/Fig-9].

Development of both puborectalis and external anal sphincter (EAS) was poor in high ARM [Table/Fig-10,11]. Higher the ARM, poorer the pelvic muscle development [Table/Fig-12].



[Table/Fig-10]: Axial T2w MRI image of a 40 days old male child with rectal atresia and rectobulbar fistula showing fair development of PR and EASC that we see muscles fibres but less than that of normal.

DISCUSSION

Current study was done to evaluate paediatric ARM patients with regards to presentation, type of ARM, associated anomalies, PR and External Anal Sphincter Complex (EASC) muscle development. It also compared distal colostogram and MRI pelvis against operative patients, while in females it was vestibular fistula (43.4%).Similar observations were made by Lawal TA et al. [18]. None of the female presented without fistula which is consistent with the study done by Upadhyaya V et al., which showed that presentation of female patient with anorectal malformation without fistula is quite rare condition [19].

Type of anomaly: High ARM were more common than low ARM as a whole, Cho S et al., reported 60% incidence of high and

Muscle	Type of ARM	Good	Fair	Poor
Puborectalis	High (34)	7 (20.6%)	17 (50%)	10 (29.4%)
	Low (26)	16 (61.5%)	10 (38.5%)	0%
EAS complex	High (34)	4 (11.8%)	16 (47%)	14 (41.2%)
	Low (26)	18 (69.2%)	8 (30.8%)	0%

[Table/Fig-11]: Development of puborectalis and external anal sphincter muscle in different types of ARM.

	High ARM (34)	Low ARM (26)
RWPR* <0.18	9 (26.5%)	2 (7.7%)
RWEASC**<0.15	13 (38.2%)	1 (3.8%)

[Table/Fig-12]: Semi-quantitative development of puborectalis and external anal sphincter muscle in different types of ARM.
*Relative width of puborecatlis muscle ** Relative width of external anal sphincter complex

intermediate anomalies whereas 40% incidence of low anomaly. Hashmi MA et al., [17,20]. Observed low ARM in 72% of female patients.

Comparison of type of anomaly: On comparison of type of anomaly identified on DC and MRI pelvis with surgical details and further association between these imaging modalities findings no significant difference was found in the diagnostic accuracy of two modalities (p-value 0.13). Madhusmita G et al [21] found diagnostic accuracy of 93.3% for MRI and 76.9% for DC to determine exact level of rectal pouch.

Distal colostogram and MRI findings according to Krickenbeck classification: When patients were classified according to krickenbeck classification on MRI pelvis, among males rectourethral fistula was seen in 16 out of 37 male patients (43%) making it most common anomaly. The incidence of rectourethral fistula among male patients of ARM had been reported as 61.2% by Patwardhan N et al., and 60% by Cho S et al., slightly higher compared to our study [17,22]. While Theron A et al., and Kuradusenge P et al., reported prevalence of rectovestibular fistula 70-78% of all the malformation in female patients [23,24]. Madhsmita et al., also reported rectourethral fistula most common in males [21].

On comparison of diagnostic accuracy of distal colostogram and MRI pelvis with surgical findings regarding fistulous anatomy, MRI showed greater accuracy of 93% compared to 56.7% with distal colostogram. Thomeer MG et al., found that MRI and DC could correctly identify fistula anatomy in 88% and 61 % of the patients respectively [25].

Associated anomalies: Out of all the patients included in the study 66.7% show associated anomalies well within the range described in literature (40-70%) [5-8]. Shenoy NS et al., in their study in eastern Indian population have found 56.04 % associated anomalies [26]. Associated anomalies were more common in patients with high ARM compared to that of low ARM. Association of anomalies with high ARM and male predominance is also supported by Boocock GR et al., who noted 53% incidence of associated anomalies, association with high ARM was 81% and male Vs female involvement was 56% Vs 47% [27]. Cho S et al., observed that patients with high ARM were 13 times more likely to have associated anomalies [17]. Above data supports the statement that high ARM are more complex lesions more likely to be accompanied by anomalies of other organ system developing between four to eight weeks of gestational age.

Musculoskeletal anomalies: Musculoskeletal anomalies were associated in 32.5% of the present study patients. Similar data was reported by Boocock GR et al., (34.3%) and Cho S et al., (43%) [17,27].

Spinal anomalies: Spinal anomalies were present in 52.5% of the patients and it was more commonly associated with high ARM compared to low ARM. Spina bifida (22.5%) was the most common spinal found in our study followed by tethered cord (10%). Levitt MA et al.[6] had reported prevalence of tethered cord between 10 to 52%.This wide range can be attributed to available imaging modalities with the clinician and how meticulously patient were investigated.

Genitourinary anomalies: Genitourinary anomalies are the most frequently anomalies in the studied population comprises 47.5% of all associated anomalies. These are more commonly associated with high ARM compared to low ARM. Levitt MA et al. [28] found genitourinary anomalies as most common associations (60%) with vesico-ureteric reflux and hydronephrosis as most common findings.

Sphincter muscle complex development: Development of puborectalis muscle was good in 20.6 % patients with high ARM, whereas 61.5% in patients with low ARM. Shah AA et al. [29] demonstrated development of puborectalis muscle was good in only 11.1% of patients with high ARM and 72.7% in patients with intermediate and low type. Development of External anal sphincter was good in 11.8% patients with high ARM, and 69.2% in patients with low ARM. Tang ST et al. [15] showed EAS was good in 8.33% patients with high ARM, 36.5% of patients with intermediate ARM and 87.5% of patients with low ARM.

Semiquantitative assessment of pelvic musculature was done by assessing RWPR and RWEAS. The RWPR <0.18 and RWEAS <0.15 suggestive of poor development, described by Tang ST et al., [15] were used in the study. Semi-quantitative assessment calculating RWPR <0.18 was observed in 26.5% of patients with high ARM and just 7.7% of patients with low ARM. Similarly RWEAS <0.15 was seen in 38.2% patients of high ARM and 3.8% cases of low ARM. Madhusmita et al., found that objective criteria given by Shah AA et al., didn't correlate well with operative outcome and these cut-off values were not found to be significant [21,29].

Limitation(s)

Patients were not followed-up in the postoperative following definitive surgical repair to evaluate optimal functional outcome of surgery like bowel bladder continence. Bias in the interpretation of radiological imaging and clinical practice couldn't be eliminated due to multicentric nature of study.

CONCLUSION(S)

The ARMs are complex hindgut malformation due to faulty embryogenesis affecting distal most urogenital and gastrointestinal tract. ARM show frequent association with other multisystem anomalies more so in high ARM. The MRI is an excellent modality and plays significant role in evaluation of ARMs. In addition to DC, MRI assesses pelvic floor musculature, spinal anomalies which prognosticate surgical outcome. However, there is no significant difference between DC and MRI findings regarding type of ARM

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