

ORIGINAL ARTICLE

MRI as the Only Pre-Operative Imaging Modality in ARM: What Advantage does it Provide?

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ABSTRACT

Background: MRI has not been traditionally used as an imaging modality for preoperative evaluation of ARM. It has been reserved for complicated ARM cases and Imaging in post-operative period. **Aims:** To study the role of MRI in preoperative evaluation of ARM patients, whether it be used as a single preoperative imaging investigation in ARM and what added advantage does it give to conventional imaging techniques. **Settings and Design:** Hospital based observational study was conducted over a period of 1 year which included 26 ARM patients who required preoperative imaging which was done using MRI.

Methods and Material: Multi-planar T1 and T2WI images of pelvis were acquired followed by screening of abdomen done with coronal T2WI and screening of spine with T2FS sagittal images. Further sequences were acquired for evaluation of any target lesions as and when required. Images were evaluated for level of atresia, type of Fistula, additional anomalies in Vertebral / Spinal cord / Genital / Urinary system and sphincter muscle complex (SMC) development. Results were analysed using numbers, percentage and arithmetic mean.

Results: MRI correctly identified the level of termination of rectal pouch in all patients. MRI correctly depicted the fistula in 89% cases. Seventy three percent cases had additional anomalies with 66% of cases with additional anomalies having multiple anomalies. MRI accurately evaluated SMC development in all cases.

Conclusions: MRI provided elaborate pelvic anatomical details, precise information about the degree of development of pelvic musculature and information regarding the presence of additional anomalies in genitourinary and vertebra-spinal systems in a single sitting and without use of any ionizing radiation to child. We recommend MRI as the only needed imaging investigation in ARM cases before definitive surgical repair.

Keywords: Anorectal malformation; Pre-operative; Magnetic resonance imaging; MRI

INTRODUCTION

Anorectal malformations (ARMs) are a complex group of congenital anomalies involving the distal anus and rectum [1,2]. The classification of ARMs is mainly based on the position of the rectal pouch relative to the puborectal sling and the presence or absence of fistulas [3,4]. The best-known

classification of ARMs is the Wingspread classification of 1984 and Krickbeck classification (Table 1) [5,6].

A thorough perineal inspection usually gives the most important clues about the type of malformation that the patient has. If low type of ARM can be diagnosed clinically, definitive surgical repair can be done. In any other case a colostomy is performed as

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a diversion and the new born is evaluated using conventional imaging methods (distal colostogram, transperineal ultrasonography) for demonstration of the altered anorectal anatomy. Without this information, an operation in the newborn period is essentially a blind perineal exploration and in turn damage unexpected structures during the search for rectum [2,7,8].

Additional anomalies are common in ARM and ranging in incidence from 40-60% in different study series [9,10]. Some of these anomalies may be life threatening or may have a greater impact than the ARM itself on the quality of survival of the child. It is imperative, therefore, that these are identified prior to undertaking surgery to treat the ARM [11-13]. For patients with ARMs, post-operative continence depends on the grade of development of sphincter muscle complex (SMC). Pelvic MRI though not done routinely is the most effective imaging technique for determining the grade of development of SMC [7,8,15-17]. This information will help the medical team make decisions about the definitive surgical approach and provide orientation about the possible post-operative prognosis [2].

Our purpose was to study the role of MRI before definitive repair in ARM patients, what advantage does it give to conventional imaging and whether it be used as a single imaging alternative which can replace other conventional techniques.

MATERIALS AND METHODS

Selection and description of participants:

After ethical clearance from Institutional Review Board, a hospital based one-year prospective study was conducted from July 2015 to June 2016. It included all patients clinically diagnosed as ARM who needed imaging investigation before definite surgical repair.

MRI examination:

The child was put to sleep and MRI was done with a 1.5T MRI (SIEMENS MAGNETOM AVANTO 1.5 Tesla whole body MRI system). If the child needed sedation, Triclofos Sodium oral solution was used at the recommended dosage. In patients in whom, diverting colostomy has been done, an adequate sized Foley catheter is advanced through the stoma of distal loop and the balloon inflated. The required amount of normal saline was instilled into the distal colon loop to distend it. For pelvis, the patients were placed in a supine position with the pelvis concentrated on the coil. The head coil was applied in them. Imaging was done using field of view of 180-

200 mm in infants and children. Sagittal, coronal and transverse fast spin-echo (FSE) T1- weighted and T2-weighted images of the pelvic region were obtained in all patients. Axial sequences were parallel and coronal sequence perpendicular to the pelvic floor. Slice thickness was kept between 3-5mm. Screening of abdomen was done with coronal T2WI and screening of spine with T2FS sagittal images. Further sequences were performed for evaluation of any target lesions in pelvis, abdomen or spine as and when required.

Image analysis:

Findings were evaluated as per following protocol. Level of atresia was noted and classified as per wingspread classification into high, intermediate, low and miscellaneous groups based on the relationship of rectal pouch with pubo-coccygeal (PC) plane and ischial (I) plane. The type of Fistula was classified as per Krickenbeck classification (Table 1). Confirmation of the level of ARM and type of fistula was done at the time of operative procedure. Additional anomalies in vertebral, spinal cord, genital and urinary system were also evaluated. Grading of SMC development was done, separately for Puborectalis (PR) and External Anal Sphincter (EAS). The relative width of puborectalis (RWPR) and relative width of external anal sphincter (RWEAS) were calculated as per the method adopted in the study by Tang et al. [18] From the axial images through the plane of PR and EAS, the width of PR and EAS muscles are calculated by adding the muscle thickness at 3'O clock and 9 O' clock positions. Width of muscles divided by half the distance between inner margins of ischial tuberosity gives the relative width of muscles. The RWPR and RWEAS of normal children (controls) imaged for non-related indications and without Anorectal abnormalities were also calculated.

In ARM cases, a grade of GOOD was given if the muscle shows same degree of development as those without anorectal abnormalities, i.e., RWPR and RWEAS is above the lowest limit of 95% confidence interval (CI) for normal children. A grade of FAIR was given if the given sphincteric muscles are identifiable and continuous but RWPR and RWEAS is below the lowest limit of 95% CI for normal children. A grade of POOR was given if muscle was not identified or barely identified, or is interrupted.

Statistical analysis:

Descriptive statistical approach was used to describe patients' characteristics using numbers, percentage and arithmetic mean. MRI findings of level of ARM

and type of fistula were compared with operative findings and expressed as number and percentages.

Statistical test used is Fisher's exact test. A p-value of <0.05 was considered statistically significant.

Table 1: Classification of ARM

Wingspread classification		
Level of anomaly	Male	Female
High	1. Anorectal agenesis A. Rectovesical fistula B. Without fistula 2. Rectal Atresia	1. Anorectal agenesis A. Rectovaginal fistula B. Without fistula 2. Rectal Atresia
Intermediate	1. Rectourethral fistula 2. Anal agenesis without fistula	1. Rectovestibular Fistula 2. Rectovaginal fistula 3. Anal agenesis without fistula
Low	1. Anocutaneous (perineal) fistula 2. Anal stenosis	1. Anovestibular (perineal) fistula 2. Anocutaneous (perineal) fistula 3. Anal stenosis
Miscellaneous	Rare malformations	Persistent cloacal anomaly Rare malformations
Krickenbeck Classification		
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 Other	

RESULTS

A total of 26 patients were studied (Table 2). Anorectal malformation had nearly equal incidence among sexes with mild preponderance in the female sex. (M: F=46%: 54%). On classifying cases as per wingspread classification, intermediate type of ARM accounted for 38.5% cases, low and high type accounted for 23.1% cases each and miscellaneous cases accounting for 15.4%. The miscellaneous cases included 1 case of cloacal malformation, 2 cases of cloacal exstrophy and a case of anorectal duplication (Fig. 1). On surgical correlation, the level of atresia was correctly determined in 100% of patients with MRI.

On classifying cases as per Krickenbeck classification, 9 out of 26 cases (34.6%) demonstrated no fistula on MRI. The most common type of fistula identified in male during surgery was rectourethral fistula (Fig. 2) which was present in 41.7% and in female was vestibular fistula (Fig. 3) in 28.6% cases. However following surgery, it was revealed that MR failed to detect 2 cases of 'rectourethral fistula' and a case of 'rectovaginal fistula' and was misinterpreted as 'ARM without fistula'. The total no of correct diagnosis made on MRI was 23 out of 26 cases studied (88.5%).

Sensitivity, specificity, positive predictive value and negative predictive value of MRI in detecting fistula was 85%, 100%, 100% and 67% respectively.

Seventy-three percent (73%) cases had additional anomalies involving the vertebrae, spinal cord, urologic and/or genital systems with 66.1% of the cases with additional anomalies having multiple (two or more) anomalies (Table 3 and Fig. 4). Overall, the commonest systems to have associated anomalies were the vertebral and genital systems, (46.2% in both) followed by the urological system (38.5%). Among the male subjects, the vertebral and the urinary systems had the most associated anomalies (41.7%). Among the female subjects, the genital system had the highest incidence of associated anomalies (64.3%).

Sacrococcygeal agenesis was the commonest vertebral anomaly, was present in 7 cases. Hemi-vertebrae, spina bifida, congenital fused vertebra (block vertebra), Butterfly vertebra were the other vertebral anomalies present.

Filar lipoma and tight filum terminale were the commonest spinal cord anomaly, each present in 2 cases. Other spinal cord anomalies included lipomyelomeningocele, myelomeningocele and terminal myelocystocele (1 case each).

Unilateral renal agenesis was the commonest urinary system anomaly and was present in 3 cases. Neurogenic bladder, cloacal exstrophy, cloacal anomaly, patent urachus, hydronephrosis,

duplicated bladder and posterior urethra, horseshoe kidney and vesico-ureteral reflux were the other urinary system anomalies seen.

Table 2. The MRI findings in the 26 patients with ARM.

case	Age	Sex	Level of atresia-Wingspread classification	Type of fistula-Krickenbeck classification	Grading of PR / EAS	Additional congenital anomalies
1	2 yr	F	High	No fistula	Fair / Fair	Vertebra-SCA
2	1 mo	F	High	Rectovesical	Poor / Poor	Urinary-NB PU Genital-UA
3	4 mo	M	Intermediate	Rectourethral	Fair / Fair	-
4	6 mo	F	Misc -Cloaca	Cloaca	Fair / Fair	Urinary-Cloaca Genital-UD
5	1 mo	M	High	Rectovesical	Poor / Poor	Vertebra-SCA,HV,CV,SB.
6	4 mo	M	High	No Fistula	Poor / Poor	Vertebra-SCA Spinal-FL Urinary-NB,VUR
7	11mo	F	Low	Vestibular	Good/Good	-
8	15 d	F	Low	Vestibular	Good/Good	Vertebra-TF Spinal-SCA Genital-DC&V
9	6 mo	F	Intermediate	Vestibular	Fair / Fair	Genital-UD
10	4 d	M	Misc- Cloacal extrophy	Others	Poor / Poor	Vertebra-HV Spinal-TMC Urinary-Cloacal extrophy Genital-MP,HT
11	8 mo	M	Low	Anal Stenosis	Good/Good	Genital-HS,UT
12	5 mo	F	Intermediate	Vestibular	Good/Good	Genital-BU
13	1 mo	M	Intermediate	Rectourethral	Fair / Fair	Vertebra-BV Urinary-RA
14	25 d	M	Intermediate	Rectourethral	Good/Good	-
15	23 d	M	Intermediate	No Fistula	Good/Good	Spinal-FL Urinary-RA,HN
16	10 d	F	Low	Perineal	Good/Good	-
17	7 d	F	High	No Fistula	Fair / Fair	Vertebra-HV Spinal-TF Urinary-RA Genital-BU
18	9 mo	F	Intermediate	Rectovaginal	Poor / Poor	Vertebra-SCA,BFV.HV Spinal-LMMC Urinary-HSK Genital-UD
19	6 mo	M	Misc- anorectal duplication.	Others	Good/Good	Urinary-DB&U Genital-UT
20	1 mo	F	Intermediate	No Fistula	Poor/Good	Vertebra-BV Genital-UD
21	5 d	M	Intermediate	No Fistula	Good/Good	-
22	6 mo	F	High	Rectovaginal	Poor / Fair	Vertebra-SCA,BV
23	7 d	F	Misc- Cloacal extrophy	Others	Poor / Poor	Vertebra-SB Spinal-MMC Urinary-Cloacal extrophy Genital-UD
24	4mo	M	Intermediate	No Fistula	Fair /Good	Vertebra-SCA,SB
25	3mo	M	Low	No Fistula	Good/Good	-
26	14d	F	Low	No Fistula	Good/Good	-

BFV-Butterfly vertebra, BU-Bicornuate uterus, BV-Block vertebra, CV-Cleft vertebra, DB&U-Duplicated bladder and urethra, DC&V-Double cervix and vagina, FL-Filar Lipoma, HN-Hydronephrosis, HS-Hypospadiasis, HSK-Horseshoe kidney, HT-Hypoplastic testis, HV-Hemivertebra, LMMC-Lipomeningomyelocele, MMC-Meningomyelocele, MP-Micropenis, NB-Neurogenic bladder, OM-Omphalocele, PD-Pubic Diastasis, PU-Patent urachus, RA-Renal agenesis, SB-Spina Bifida, SCA-Sacro-coccygeal agenesis, TF-Tight filum terminale, TMC-Terminal myelocystocele, UA-Uterine agenesis, UD-Uterine didelphys, UT-Undescended testis, VUR-Vesicoureteric reflux.

Undescended testis is the commonest genital anomaly among males and was present in 2 cases. Other genital anomalies in males included hypospadias, hypoplastic testis and micropenis. Uterine didelphys was the commonest genital anomaly among females and was present in 5 cases. Double cervix and vagina with normal single uterus, a rare type of Mullerian anomaly was present in one case. Other genital anomalies in females included Bicornuate uterus and uterine agenesis.

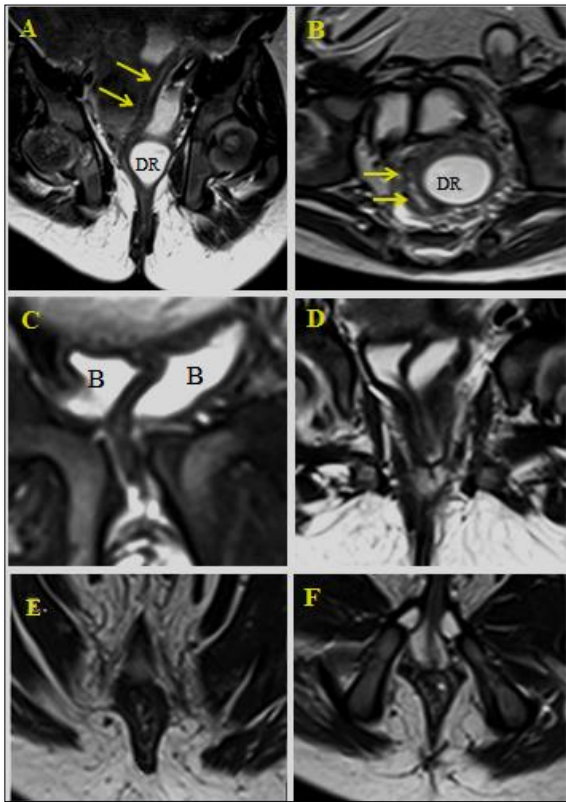


Figure 1: Anorectal duplication in a 6 months old male child. A & B: T2W coronal (A) and axial (B) images shows thin collapsed rectum (↘) on the right side of a distended tubular blind ending duplicated anorectal segment ('DR'). C & D: T2W coronal (C) and axial (D) image shows complete duplicated bladder (B) with separate muscular coats. The septa extend into the posterior urethra. E: T2W axial image just below I plane shows well developed continuous EAS muscle complex. F: T2W axial image below PC plane shows well formed puborectalis muscles encircling the rectum.

Table 3: Anomalies in different Organ System

Anomalies	Number of Cases
No associated anomalies	7 (26.9%)
One	7 (26.9%)
Two	6 (23.1%)
Three	2 (7.7%)
Four	4 (15.3%)

No statistically significant difference is seen in incidence of associated anomalies among the two genders. There was significant association (p value < 0.05) between vertebral and spinal cord anomalies and spinal cord and urological anomalies (Table 4).

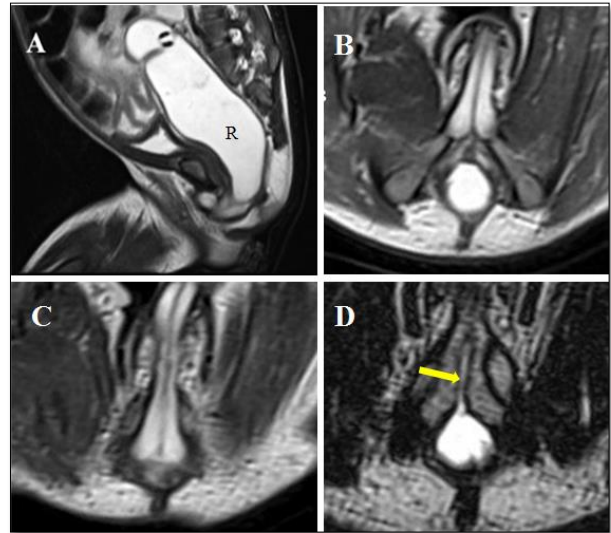


Figure 2: Intermediate ARM with rectourethral fistula in a 25 days old male child. A: T2W sagittal image shows the rectal pouch (R) terminating well below the PC plane but just above the I line. B: T2W axial image below PC plane shows well formed puborectalis muscles encircling the rectal pouch (R). C: T2W axial image below I plane shows the EAS muscle complex. D: T2 SPC axial image shows the anteriorly directed rectourethral fistulous tract (arrow) entering the bulbar urethra within the corpus spongiosum.

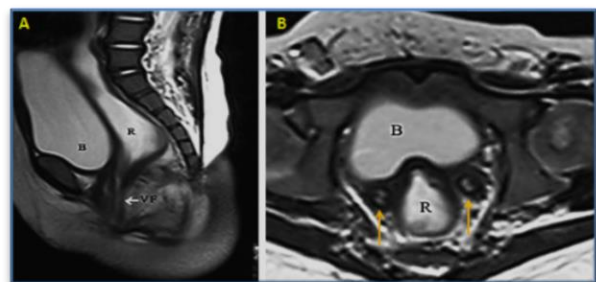


Figure 3: Intermediate ARM with vestibular fistula in a 6 month old female child. A: Sagittal T2W images show the relationship between the rectal pouch (R) and Bladder (B). Rectal pouch terminated below the PC line and continues as fistulous tract towards the vestibule (VF). B: T2W axial image shows two separate uterine horns (arrows) of didelphys uterus on either side of rectum. The vagina fuses in the lower part (not shown).

MRI allowed direct visualization of sphincter muscle complex in multiple planes facilitating accurate evaluation of its bulk size and location. In assessing the development status of PR in ARM (Fig. 5), it was found that, all cases with low ARM had 'good' PR muscle development, whereas no case with high

ARM had good muscle development. In intermediate ARM, 'good' development and 'fair' development of PR was present in 40% cases each and rest 20% had poor development. Among the miscellaneous cases, the case of anorectal duplication had 'good' development of PR, case of cloacal malformation had fair development and both cases of cloacal exstrophy had poor development. In assessing the development status of EAS in ARM (Fig.6), it was found that all the patients with low ARM had 'good' EAS muscle development, whereas no case of high ARM had 'good' development. Half of the cases with intermediate ARM had 'good' development and majority of rest had 'fair' development of EAS. Among miscellaneous cases, the case of anorectal duplication had 'good' development of EAS, case of cloacal malformation had 'fair' development and both cases of cloacal exstrophy had 'poor' development. The development state of EAS was found to be of similar or of better grade than that of PR in the intermediate and high type ARM.

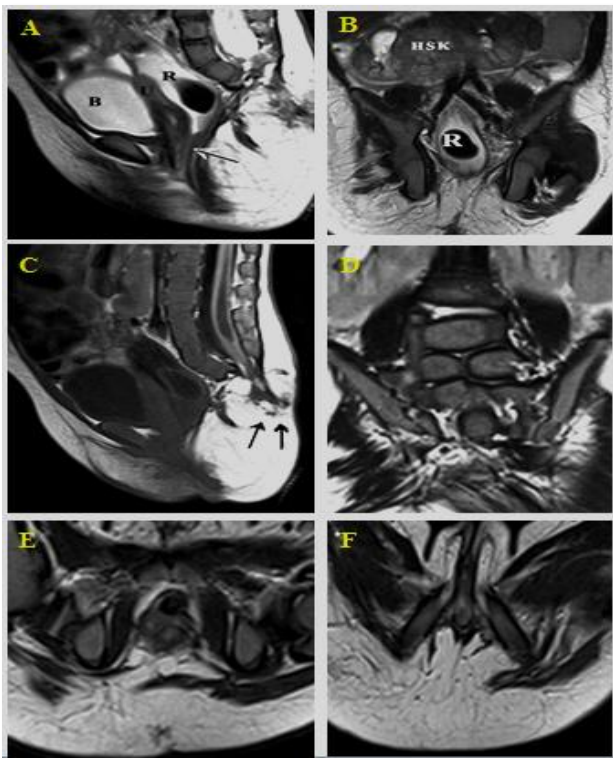


Figure 4: Intermediate ARM with rectovaginal fistula and multiple additional anomalies in 9 months old female child. A: T2W sagittal image shows the rectal pouch (R) terminating well below the PC plane. A thin T2 hyperintense fistulous tract (Arrow) can be seen from the rectal pouch opening into the lower vagina. Partial Sacrococcygeal agenesis is seen. Bladder and uterus are marked 'B' and 'U' respectively. B: T2W coronal image shows horseshoe kidney (HSK). Rectal pouch is marked 'R'. C: T1W sagittal image shows sacrococcygeal agenesis below S2 vertebra. The spinal cord is tethered due to lipomyelomeningocele

formation at sacral region (Arrow). There is formation of T1 hypointense syrinx in the lower cord. D: T2W coronal image shows multiple segmentation defects in the lumbosacral vertebrae. E: T2W axial image at PC plane shows poorly developed puborectalis muscle. F: T2W axial image along I plane shows few discontinuous (poorly developed) fibres of EAS muscle posterior to perineal body.

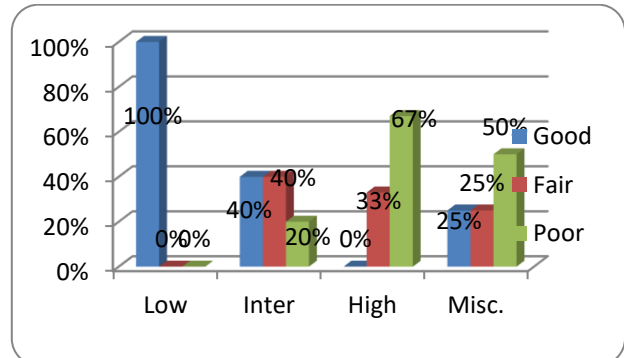


Figure 5: Development state of PR in ARM patients of various types.

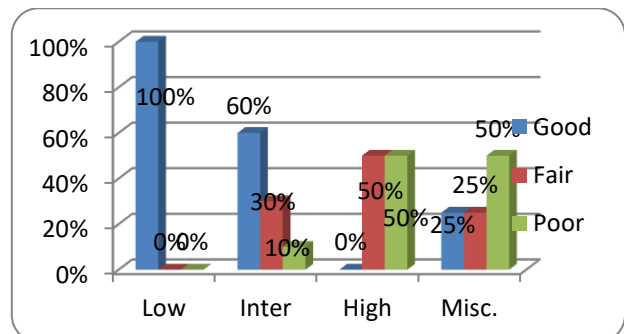


Figure 6: Development state of EAS in ARM patients of various types

DISCUSSION

Preoperative investigation in ARM has to answer the following questions:

1. The level of rectal atresia and presence and location of fistula based on which the surgical planning is done,
2. The developmental state of sphincter muscles, which decides the post-operative continence, quality of life and success of surgery and,
3. Presence of other additional anomalies, which is the major determinant of long-term prognosis.

In our study, MRI accurately determine the level of termination of rectal pouch with 100% accuracy. High pressure colostogram has been the preferred traditional modality for detecting fistula with reported accuracy ranging from 77% to 100%. [19-21] In our study, the type of fistula was accurately determined using MR in 88.5% cases, which is at par with traditional imaging method.

Table 4: Interrelationship between different associated anomalies in ARM patients based on two tailed fisher exact test. Boxes showing significant association shown in blue.

Type of Anomalies		Vertebral (12)	Spinal cord (7)	Urological (10)	Genital (12)
Vertebral (12)	Present	12; 100%	6; 85.7%	6; 60%	6; 50%
	<i>p value</i>	0	0.026	0.422	1
Spinal cord (7)	Present	6; 50%	7; 100%	6; 60%	5; 41.7%
	<i>p value</i>	0.026	0	0.005	0.19
Urological (10)	Present	6; 50%	6; 85.7%	10; 100%	7; 58.3%
	<i>p value</i>	0.422	0.005	0	0.1
Genital (12)	Present	6; 50%	5; 71.4%	7; 70%	12; 100%
	<i>p value</i>	1	0.19	0.1	0

Table 5: Reported incidence of additional anomalies in ARM.

Author	Kiesewetter [22]	Smith [23]	Cho S [24]	Mittal [25]	Ratam [26]	Present Study
Anomalies	54%	61%	71%	59%	58%	73.1%

In the present study, 19 out of the 26 cases (73.1%) having associated anomalies involving the vertebrae, spinal cord, urologic and/or genital systems. Most of the previous studies done using traditional imaging methods, the reported incidence of associated anomalies with ARM was from 30% to 70% (Table 5) [13, 22-25]. Even after studying associated anomalies in only the above mentioned four systems, our study reported higher incidence of associated anomalies compared to the previous studies. This can be due to better evaluation of urologic, genital, spinal and vertebral systems using MRI. In this study, vertebral, spinal cord and urological anomalies were found in 46.2%, 26.9% and 38.5% patients respectively. Genital anomalies in males and females were 25% and 64% respectively. The vertebral, spinal, urinary and male genital anomalies in our study showed similar incidence to studies reported in literature using traditional methods (Table 6) [13, 22-30]. However, the incidence of genital anomalies in females ARM patients in literature ranges from 16 to 45% with the anomalies predominating in external genitalia and genital tracts, whereas our study revealed higher incidence genital anomalies in 64% female cases. This could be attributed to the better detection of Mullerian anomalies using MR imaging, compared to traditional imaging techniques used in former studies.

MRI provided accurate information regarding the bulk and development of sphincter muscle complex

as it allows direct visualization and thus its evaluation. All cases with low ARM had 'good' muscle development whereas no case of high ARM showed 'good' muscle development. The development state of EAS was found to be of better or similar grade than that of PR in the intermediate and high type ARM.

Table 6: Reported incidence of Organ System Involved with additional anomalies in ARM.

Organ system	Previous studies	Present Study
Vertebral	15% - 51% [22-26]	46.2%
Spinal cord	14% - 57% [30,31]	26.9%
Urinary	26% - 52% [26-29]	38.5%
Genital (Males)	16% - 26% [26-29]	25%
Genital (Females)	16% - 45% [26-29]	64%

CONCLUSION

MRI provided elaborate anatomical details which were well correlated with findings at surgery. It gives precise information about the degree of development of pelvic musculature. In addition, it gave accurate information regarding the presence of additional anomalies in genitourinary, vertebral and spinal systems, the diagnosis of which if made early helps in tailoring the management plan for them. MRI is the single imaging modality which can answer all these aspects of ARM accurately in a single sitting and without use of any ionizing radiation to child. Therefore, we suggest MRI as the only needed

imaging investigation and can be a better alternative to traditional imaging in ARM cases which requires imaging investigations before definitive surgery.

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