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Comparative Effectiveness of Imaging Modalities for Preoperative Assessment of Anorectal Malformation in the Pediatric Population

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ABSTRACT

Objective: The aim of this study was to compare the accuracy of MRI, colostography/fistulography, and X-ray imaging modalities for preoperative diagnosis of anorectal malformations (ARMs) in pediatric patients.
Methods: This retrospective analysis included a total of 84 pediatric patients with ARMs. Preoperative imaging findings were assessed by 2 radiologists and compared to surgical findings.
Results: MRI identified anomalies of the spine in 25 of 84 patients (29.8%), anomalies of the genital system in 7 of 84 patients (8.3%), anomalies of the urinary system in 22 of 84 patients (26.2%), and underdeveloped sphincter muscle complex in 34 of 84 patients (40.5%). In the 44 subjects receiving both MRI and X-ray, MRI was more sensitive in detecting anomalies of spine (18/44 vs. 8/44; $P = 0.002$), and both correctly identified the distal end of the rectum in 77.3% (34/44) of the cases. In the 24 subjects receiving both MRI and colostography/fistulography, MRI was more accurate in identifying Pena's classification (22/24 vs. 15/24; $P = 0.039$). Distal end of the rectum was correctly identified in 75.0% (18/24) and 58.3% (14/24) of the cases ($P = 0.125$).
Conclusions: MRI could clearly reveal fistula anatomy and associated anomalies of ARMs and should be routinely used for preoperative evaluation of ARMs.
Type of study: Study of diagnostic test.
Level of evidence: Level II.

Anorectal malformations (ARMs) occur with estimated prevalence ranging from 2 to 6 per 10,000 live births [1], and include a variety of conditions ranging from anal stenosis or anorectal agenesis, with or without fistula, to more complex anomalies, such as cloacal malformations. ARMs are often associated with genitourinary defects and spinal anomalies [2].

Colostography/fistulography and X-ray are recommended prior to definitive surgical management to reveal fistula anatomy and the distal end of the rectal pouch [3]. Magnetic resonance imaging (MRI) could clearly visualize the pelvis and spinal cord, and thus is also widely used to in ARMs patients [4,5]. There have been no studies of large sample size comparing MRI with other imaging modalities in preoperative assessment of ARMs. In the present study, we compared MRI with X-ray and colostography/fistulography in 44 and 24 patients with ARMs, respectively.

1. Methods

This retrospective study included all children who underwent surgery to correct ARMs at the Children's Hospital of Soochow University during a period from March 2015 to December 2018. Imaging and clinical data were retrieved from medical records. This study was approved by the Ethics Committee of Children's Hospital of Soochow University (No. 20150106010).

1.1. Imaging techniques

1.1.1. MRI

MRI was conducted using a Symphony 3.0 T MR scanner (GE Healthcare, Chicago, IL, USA), commissioned in 2014, with no subsequent upgrade. The imaging included fast spin echo (FSE), fast recovery fast spin echo (FRFSE), periodically rotated overlapping parallel lines with enhanced reconstruction (PROPELLER) T1WI and fs T2WI. Sagittal, coronal and axial images were obtained with a FOV of 280 mm, TR/TE of 2537 ms/120 ms, a bandwidth of 50 Hz/pixel, and a matrix size of 320 × 288 pixels. Uncooperative children were given 10% chloral hydrate (0.5 mL/kg body weight, po) prior to the scan.

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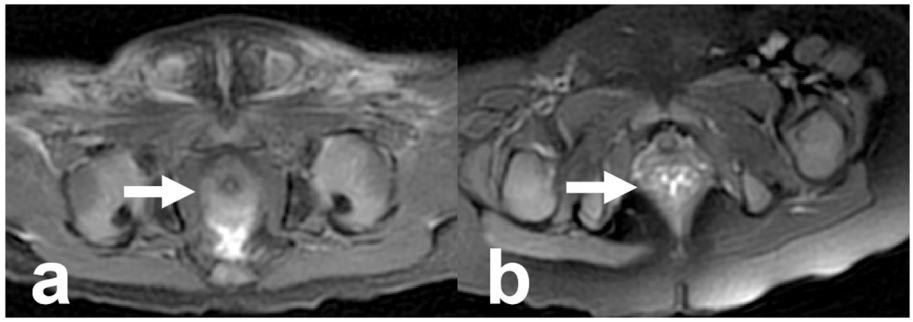

Fig. 1. Representative fistula in axial fs T2WI MR images. a. ARM. b. Healthy rectum.

1.1.2. Colostography/fistulography and X-ray

For colostography, 20–50 mL 50% Omnipaque was injected via an 8-F catheter into the anastomosis of the rectum slowly by hand to slightly extend and maintain the slight pressure of distal rectal pouch. Contrast agent was injected to distal fistula when there is no anastomosis with the patient. X-ray was conducted 10 min after the patient was inverted to make sure the distal rectal pouch was full of gas.

1.1.3. Image evaluation

Images were reviewed by two experienced radiologists, who were unaware of the intraoperative findings independently. Inconsistencies were resolved through discussion. Radiologists were asked to determine the Pena's Classification of malformation [2], the anatomy of the fistula, the distal end of rectum and any associated anomalies. Fistula anatomy was assessed using T2-weighted fs axial MR images (Fig. 1). Underdeveloped sphincter muscle complex is discerned by comparing with healthy children of the same age. Diagnostic performance was evaluated using intra-operative findings as the reference.

1.2. Statistical analysis

McNemar's test was used to assess the performance of imaging studies with statistical analysis software (SAS) 9.2. $P < 0.05$ was considered statistically significant.

2. Results

2.1. MRI findings of ARMs and associated anomalies

MRI was conducted in 84 patients (48 boys, 36 girls; age range: 0 day to 21 months). Pena's classification (representative image in Fig. 2) based on surgical findings included: rectoperineal fistula (47/84; 56.0%), rectobladder neck fistula (5/84; 6.0%), rectoprostatic fistula (1/84; 1.2%), rectobulbar fistula (2/84; 2.4%), rectovestibular fistula (12/84; 14.3%), cloacal malformation (2/84; 2.4%), imperforate anus (14/84; 16.7%), and anal stenosis (1/84; 1.2%). In the overall sample of 84 subjects, malformations of the spine (representative image in Fig. 3) ($n = 30$) were identified by MRI in 29.8% (25/84) of the patients, malformations of the genital system ($n = 7$) were identified in 8.3% (7/84) subjects, malformations ($n = 22$) of the urinary system (representative image in Fig. 3) were identified in 26.2% (22/84) subjects (specific types of the anomalies in Table 1). Sphincter muscle complex was underdeveloped in 40.5% (34/84) of the patients (representative image in Fig. 4).

2.2. Comparison between MRI and X-ray

A total of 44 patients had both MRI and X-ray results. Pena's classification based on surgical findings included: rectoperineal fistula (16/44; 36.4%), rectobladder neck fistula (5/44; 11.4%), rectoprostatic fistula (1/44; 2.3%), rectobulbar fistula (2/44; 4.5%), rectovestibular fistula (4/44;

Fig. 2. Fistula anatomy in sagittal MR images. a. Rectobulbar fistula: the distal end of the rectum pouch was filled with meconium (white arrow), and the anatomy of the fistula (black arrow) extended to the urethra. b. Rectovesical fistula: Full bladder (white arrow head) and the fistula (white arrow) connected to the rectum. c. Imperforate anus: the distal rectum was closed (white arrow).

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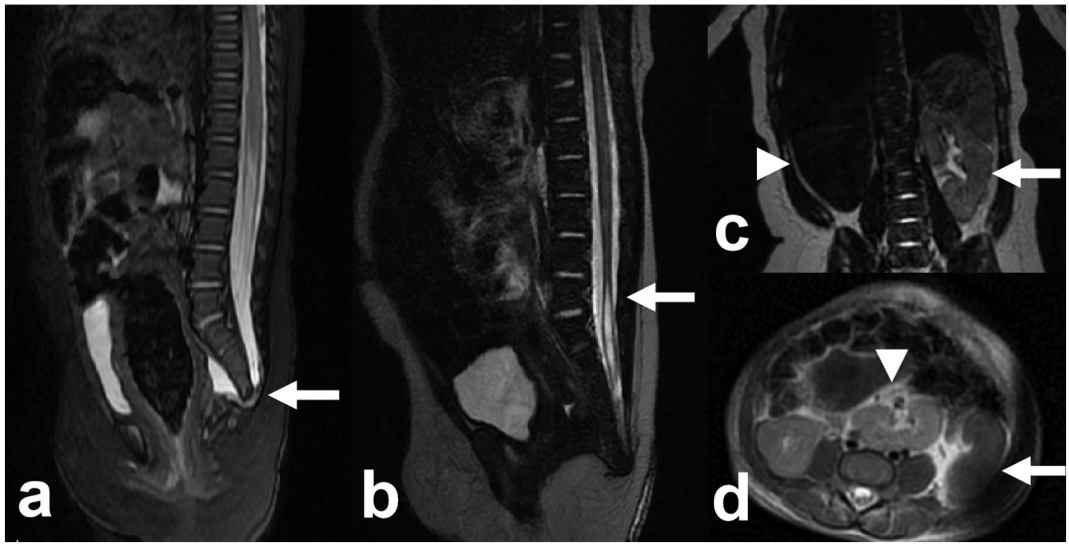

Fig. 3. Associated malformations in T2WI MR images. a. Anterior meningoceles (white arrow). b. Tethered spinal cord (white arrow). c. Isolated kidney (white arrow): the right kidney was absent (white arrow head) d. Ectopic kidney (white arrow head): the location of left kidney was replaced by intestinal canal (white arrow).

Table 1
MRI findings of ARMs associated anomalies.

MRI Findings	Number
Malformations of the spine	30
dysplasia of the coccyx	15
tethered cord	7
syringomyelus	3
syringomyelia	1
anterior meningoceles	1
vertebral fusion	1
fatty filum terminale	1
spinal lipoma	1
Urinary system	22
hydronephrosis	10
renal dysplasia	3
bladder diverticula	3
isolated kidney	2
ectopic kidney	2
renal fusion	2
Genital system	7
hydrocele of tunica vaginalis	3
buried penis	1
cryptorchidism	1
double vagina	1
penile scrotal division	1
Underdeveloped sphincter muscle complex	34
total	93

9.1%), cloaca (2/44; 4.5%) and imperforate anus (14/44; 31.8%). Both MRI and X-ray visualized the end of the rectum in 77.3% (34/44) of cases (P = 1.000) (Fig. 5). MRI and X-ray clearly showed anomalies of the spine in 40.9% (18/44) vs. 18.2% (8/44) patients, respectively (P = 0.002) (Fig. 5).

2.3. Comparison between MRI and colostography/fistulography

A total of 24 subjects received both MRI and colostography/fistulography. Pena's classification based on surgical findings included: rectoperineal fistula (10/24; 42%), rectobladder neck fistula (5/24; 21%), rectovestibular fistula (5/24; 21%), rectal stenosis (1/24; 4%), and imperforate anus (3/24; 13%). The fistula type was correctly identified in 91.7% (22/24) vs. 62.5% (15/24) by MRI and colostography/fistulography, respectively (P = 0.039) (Fig. 5). The position of rectal pouch distal end was correctly identified in 75.0% (18/24) vs. 58.3% (14/24) by MRI and colostography/fistulography, respectively (P = 0.125) (Fig. 5).

3. Discussion

Detailed pelvic structure information must be obtained to guide surgery. In a study by Maarten et al. [6], 33 patients with ARMs underwent both MRI and fistulography/colostography. Comparison of the imaging findings with the intraoperative observation showed better performance of MRI vs. fistulography/colostography. Gokhan et al. [7] studied 61 patients with ARMs and concluded that fistulography and MRI have distinct advantages in the diagnosis of perianal fistula. A study by Carroll et al. [8] confirmed that MRI is an important component in the workup in patients with imperforate anus, depending on the clinical information obtained and presence of associated malformations. The sample size of most previous studies, however, is relatively small.

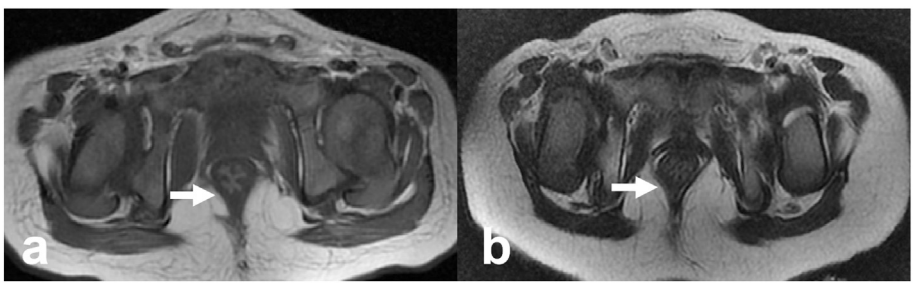

Fig. 4. Sphincter muscle complex in axial T2WI MR images. a. Well-developed sphincter muscle complex (white arrow). b. Underdeveloped sphincter muscle complex (white arrow).

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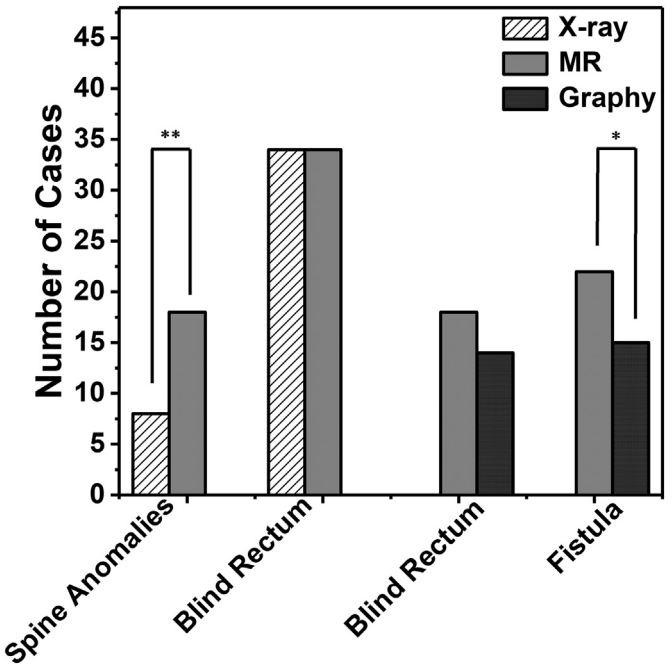

Fig. 5. Diagnostic performance of MR vs. X-ray and colostography/fistulography for spine anomalies, the end of the blind rectum, the blind end of the rectum and fistula anatomy.

The results from the current study showed that MRI is as good as X-ray in showing the end of the rectal pouch (77.3% vs. 77.3%), but is more sensitive than X-ray at discerning anomalies of the spine (40.9% vs. 18.2%; P = 0.002). MRI was also more reliable in revealing the fistula structure than colostography/fistulography (91.7% vs. 62.5%; P = 0.039). These findings are consistent with a previous study by Soker et al. [7] concluding better performance of MRI vs. fistulography in fistula classification. With regards to the location of the blind rectum, the current study also suggested superior performance with MRI (75.0% vs. 58.3% with colostography/fistulography).

MRI could determine the level of the striated muscle, and help surgeons to reconstruct the anal canal and suture the bowel wall to the sphincter muscle complex and skin [9]. In this study, 34 of 84 patients (40.5%) had underdeveloped sphincter muscle complex. Preoperative assessment of associated malformations is also critical for planning simultaneous repair. Consistent with the highly prevalent associated anomalies (79 out of 86 patients) reported by England et al. [10], MRI demonstrated a total of 93 anomalies in 84 patients in the current study, including malformations of the spinal cord and spine (n = 30), the genital system (n = 7), the urinary system (n = 22), and underdeveloped sphincter muscle complex (n = 34). Despite the promising diagnostic performance, it is noteworthy to point out that many children must be sedated to carry out MRI examination. As a result, the sedative agent carries potential risks of vomiting and allergy. More importantly, MRI tends to underestimate the position of the distal end of the rectum without fluid distention.

There were several limitations in the current study. First, only part of the study subjects received both MRI and fistulography/colostography; the comparison is thus less powered. Second, this study is a retrospective analysis and thus is subjected to a variety of biases. Differing age of the patients may also lead to differences in anatomical structures.

4. Conclusion

MRI is more accurate than X-ray and colostography/fistulography in discerning fistula anatomy and detecting associated anomalies, particularly in complex ARMs cases. MR should be routinely used in preoperative of ARMs.

Compliance with ethical standards

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Declarations of interest

None.

Patient consent

Guardian consent obtained.

References

[1] Wijers CHW, Van Rooij IALM, Marcelis CLM, et al. Genetic and nongenetic etiology of nonsyndromic anorectal malformations: a systematic review. Birth Defects Res C Embryo Today. 2014;102:382–400. https://doi.org/10.1002/bdrc.21068.
[2] Al-Salem Ahmed H. Anorectal malformations. Pediatr Surg Int. 2007;2:61–70. https://doi.org/10.1186/1750-1172-2-33.
[3] Kraus SJ, Levitt MA, Peña A. Augmented-pressure distal colostogram: the most important diagnostic tool for planning definitive surgical repair of anorectal malformations in boys. Pediatr Radiol. 2017;48:1–12. https://doi.org/10.1007/s00247-017-3962-2.
[4] Aslam A, Grier DJ, Duncan AW, et al. The role of magnetic resonance imaging in the preoperative assessment of anorectal anomalies. Pediatr Surg Int. 1998;14:71–3. https://doi.org/10.1007/s003830050439.
[5] Mchugh K. The role of radiology in children with anorectal anomalies; with particular emphasis on MRI. Eur J Radiol 1998;26:194–199. . https://doi.org/10.1016/S0720-048X(97)00095–8 .
[6] Thomeer MG, Devos A, Lequin M, et al. High resolution MRI for preoperative work-up of neonates with an anorectal malformation: a direct comparison with distal pressure colostography/fistulography. Eur Radiol. 2015;25:3472–9. https://doi.org/10.1007/s00330-015-3786-0.
[7] Soker G, Gulek B, Yilmaz C, et al. The comparison of CT fistulography and MR imaging of perianal fistulae with surgical findings: a case-control study. Abdominal Radiology. 2016;41:1–10. https://doi.org/10.1007/s00261-016-0722-y.
[8] Carroll AG, Kavanagh RG, Ni Leidhin C, et al. Comparative effectiveness of imaging modalities for the diagnosis of intestinal obstruction in neonates and infants: a critically appraised topic. Acad Radiol. 2016;23:559–68. https://doi.org/10.1016/j.acra.2015.12.014.
[9] Devries PA, Peña A. Posterior sagittal anorectoplasty. J Pediatr Surg. 1982;17:638–43. https://doi.org/10.1016/S0022-3468(82)80126-7.
[10] England RJ, Eradi B, Murthi GV, et al. Improving the rigour of VACTERL screening for neonates with anorectal malformations. Pediatr Surg Int. 2017;33:747–54. https://doi.org/10.1007/s00383-017-4094-3.

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