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Measuring the common canal of a persistent cloaca: can MRI replace conventional imaging?

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AIM: To evaluate the role of MRI in preoperative assessments of patients with a persistent cloaca and compare magnetic resonance imaging (MRI) versus fluoroscopy contrast study in the accuracy of common canal measurement and classification prediction.

MATERIALS AND METHODS: Thirty-one patients with a persistent cloaca were diagnosed and treated at Guangzhou Women and Children's Medical Center between March 2011 and December 2017. The length of the common canal was measured using MRI and fluoroscopy contrast study. Classification results based on measurements were compared with cystoscopy and intraoperative findings. The accuracy in predicting the classification by measuring the common canal length was compared.

RESULT: Among 31 patients, 24 had MRI, 24 underwent fluoroscopy contrast study, and 25 underwent cystoscopy. In 20 patients, MRI-based categorisations were in accordance with cystoscopy or surgery findings, whereas in four patients there was discordance. In 17 patients, categorisations based on fluoroscopy contrast study were in accordance with cystoscopy or surgery findings, and in seven patients there was discordance; the difference was not statistically significant ($p>0.05$).

CONCLUSION: MRI may accurately demonstrate genitourinary anomalies and the length of the common canal in patients with persistent cloaca. Categorisation based on MRI measurements of the common canal was accordant with the results from cystoscopy and findings from surgery. The use of this method may help surgeons to develop appropriate reconstruction plans before sending their patients to the operating room.

Introduction

A persistent cloaca (PC) is the most complicated anorectal malformation in females. The reported incidence of PC is about 1: 50,000.[1] It is frequently accompanied by multiple systemic anomalies[2] including tethered cord syndrome, congenital heart diseases, Müllerian and urinary system anomalies. Malformations of the genitourinary system are highly variable, making assessments and

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reconstruction very challenging.[3] The common canal (CC) is defined as the canal at which the urethra, vagina, and rectum fistula converge before they reach the perineum. According to the experiences of high-volume centres,[3–5] the length of the CC is a strong indication for appropriate surgery. PC patients are usually categorised into three types according to the length of the CC. A CC $\leq$1cm in length (type 1) can be corrected simply by posterior sagittal anorectoplasty (PSARP) only or PSARP and vaginal posterior wall mobilisation (VPWM). A CC between 1 and 3 cm (type 2) can be treated with PSARP and urogenital total mobilisation (UGTM), and a CC $\geq$3 cm (type 3) usually needs PSARP and vaginal replacement (VR).

Assessing the length of the CC is essential before surgery. To achieve this goal, different imaging methods, including fluoroscopy contrast study (FCS) and magnetic resonance imaging (MRI) are used. The disadvantages of FCS are radiation exposure, time required, the number of attempts to achieve a satisfactory image, and inability to display the anatomy of the genital system if there is vaginal stenosis or atresia. MRI is increasingly used in assessments of anorectal malformations[6] and has the advantages of displaying pelvic malformations, including the urinary tract, vertebral deformities, spinal anomalies, and the striated muscle complex (SMC); however, measurement of the CC using MRI is considered difficult.[6] In the present study, the CC was measured using MRI and the results compared with FCS, to determine whether MRI can replace conventional imaging of PC in measuring CC length.

Material and methods

This retrospective study was approved by the institutional ethics committee of Guangzhou Women and Children's Medical Center. All patients had been informed about the possibility that their medical information might be used in clinical research anonymously and consent forms were signed. The further requirement for informed consent was waived by the ethics committee.

Thirty-one patients diagnosed with PC were treated at Guangzhou Women and Children's Medical Center between March 2011 and December 2017. To understand the anatomy of the rectum and genitourinary system, ultrasonography, FCS, and contrast-enhanced MRI were performed before the surgical reconstruction plan was made. Cystoscopy was performed to confirm the anatomy before surgery.

The CC measurements followed the same principle in all methods. The CC length was measured as the straight-line distance from the end of the identifiable genital tract (vagina) to the perineum (labia minora). If the genital tract was atresic or absent, the CC was then measured from the rectum fistula to the perineum. This measurement procedure was adopted for the several reasons. First, it is very simple and easy to learn, no further calculation is needed. Secondly, the position of the vagina provides important information for the decision of surgical reconstruction, because the CC was almost always reserved and tubularised to form the urethra and the rectum can be easily reached from either the perineum or pelvic cavity, it can be pulled down with adequate mesentery mobilisation; however, genital malformation is highly variable, and reconstruction requires intensive investigation and planning. A low vagina can be pulled down by VPWM, and higher malformations can be treated with UGTM. The highest malformation usually needs vaginal replacement. In most situations, the vagina opening is not higher than the rectum. A rectum fistula that opens on the CC instead of the vagina usually indicates a high vaginal opening. To simplify the problem, the CC was measured from the vaginal opening to the perineum.

MRI examination

The patients were sedated with chloral hydrate (50–100 mg/kg) orally, then moved to the supine position before examination.[7] The MRI study was performed using a 1.5 T magnet on the lower abdominal–pelvic region using the abdomen coil. Three to five-millimetre section thickness was chosen with a 0–0.5 mm intersection gap to demonstrate the details of the pelvic anatomy. Three projections were performed in T2: sagittal sequence (3,610 ms repetition time [TR]/80 ms echo time [TE]), axial sequence parallel (3,252–3,655 ms TR/60 ms TE) and coronal sequence (3,610 ms TR/80 ms TE) perpendicular to the pelvic floor.

FCS examination

To perform the FCS, an 8 F Foley's catheter was inserted into the CC, and contrast agents (water-soluble contrast medium) were injected under fluoroscopy inspection. Lateral as well as frontal projections were taken. Retrograde colostomy contrast enema or voiding cysto-urethrogram was performed simultaneously if necessary.

Cystoscope

The patient was examined after sedation in the operating room, in the lithotomy position, a cystoscope was introduced into the CC. The genitourinary opening and rectum fistula were searched respectively and identified through referral to the preoperative imaging data. The distance was measured from the vaginal opening to the end of the CC as mentioned above.

Image analysis

The measurement method was introduced to two radiologists and one paediatric surgeon, then the CC was measured by them independently in every patient, the patients' identity and final surgical approach were kept anonymous to the doctors when they read the images, the results were averaged and categorised according to the range of different types mentioned above. Then categorisation results of MRI and FCS were compared with cystoscopy or/and surgery, respectively. The interobserver variation was analysed by comparing the results from the three doctors using analysis of variance (ANOVA), the data obtained via the three methods were of normal distribution and not statistically different between observers (Table 1).

Table 1
Comparison of measured values of the three observers.

	Observer 1	Observer 2	Observer 3	F	*p*-Value
MRI value	1.92±1.01	1.65±0.68	1.86±0.91	0.59	0.556
FCS value	2.08±0.95	1.54±0.68	1.75±0.85	2.58	0.083

FCS, fluoroscopy contrast study; CC, common canal; CS, cystoscope.

MRI measurement

The measurement of the CC in MRI followed these principles: in the sagittal sections of MRI image, the CC was measured from the end of vagina to the edge of the labia minora (Fig 1a,c). In the axial sections of the MRI image, the number of sections from the last section with the separate structure of the urethra and vagina to the last section with the labia minora were counted and multiplied by the section thickness, to obtain the length of the CC of the axial sections (Fig 1b,d). If the CC could be measured in both the sagittal and horizontal sections, the longer would be taken as the final result.

FCS measurement

In the lateral pictures, the CC was measured from the end of the vagina to the labia minora (Fig 2a). If the vagina could not be displayed, the CC was measured from the end of rectum fistula (Fig 2b).

Cystoscope measurement

When the tip of the cystoscope reached the vaginal opening, the labia level was marked on the scope, and the distance between the mark and the end of the cystoscope was considered the length of the CC.

Statistics

Statistical analysis was performed with SPSS for Window v20.0. The level of statistical significance was set at $p<0.05$. The categorisation results of MRI and FCS were compared with cystoscope and/or surgical findings. The length measured at FCS and MRI was compared with cystoscopy,

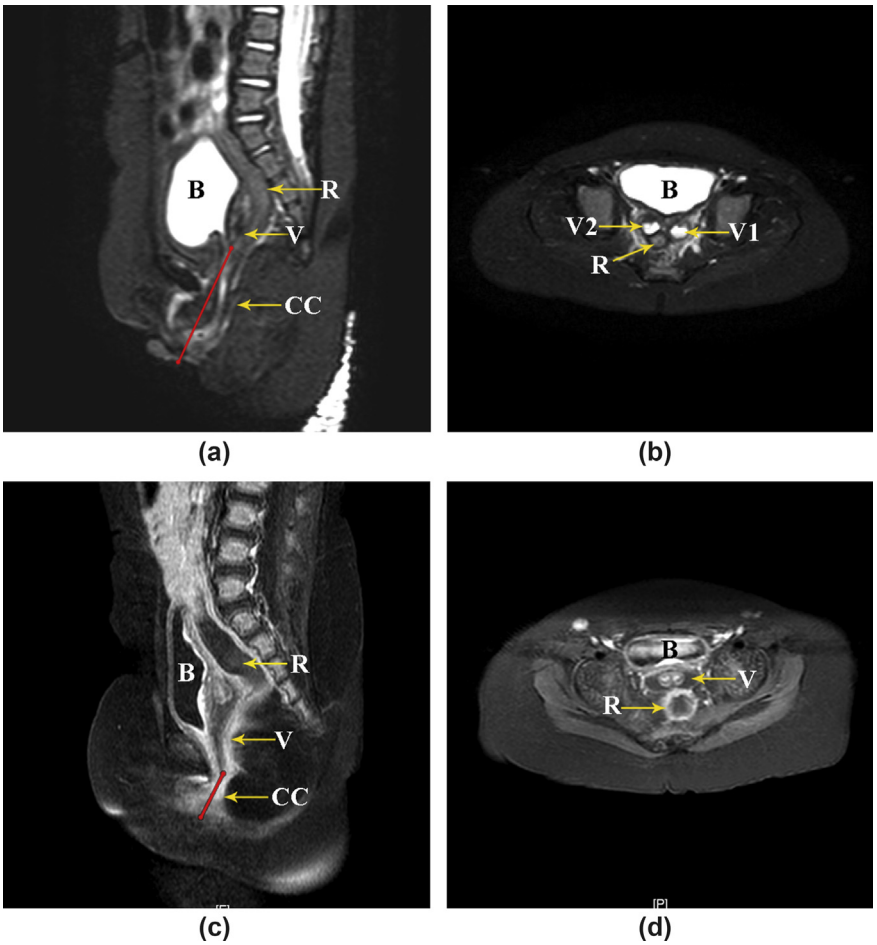

Figure 1 (a,b) Case 25: a patient aged 13 months with PC. (a) Mid-sagittal MRI displayed that the urethra, vagina (V), and rectum fistula (R) had merged into a long CC. The yellow line indicates the measurement land marker of CC. (b) Axial MRI of the same patient showing the duplicated vagina (V), rectum fistula (R) ended in between two vaginas. (c,d) Case 26: a patient aged 16 months with PC. (c) Mid-sagittal MRI showed the patient had hydrocolpos (V) and a short CC. (d) Axial MRI of the same patient showing that there were two cervixes in the vagina (V).

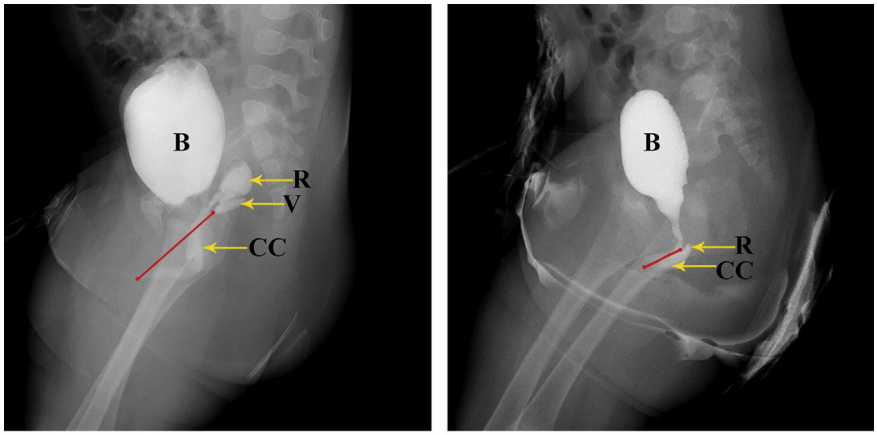

Figure 2 (a) Case 25. FCS showing the rectum (R) ended at a high level (above the coccyx). The red line indicates the measurement of CC in FCS. (b) Case 26. FCS failed to display the vagina, in this situation, measurement was made between the end of the rectum fistula (R) and the labia minora.

respectively. The coincidence rate was calculated, and the difference was analysed statistically.

Results

Clinical data

Patients' demographic data, assessments, anomalies, and surgical approaches are listed in Table 2. The average age of patients was 43.4±39.3 months (range: 5–187 months). Average age at first visit was 211.1 days. The median age was 25 days (range: 1–3,238 days).

Six patients (23.1% of 26 patients who had spine MRI) had the complication of tethered cord. Spine cord malformations including sacrococcygeal dysplasia, myelocoele dilation, spine cord lipoma, or vertebral deformity were found in 12 patients (41.4% of 29 patients had spine MRI or sacrococcygeal radiography). Nineteen patients (63.3% of 30 patients who had echocardiography) had cardiac anomalies, mostly foramen ovale or atrial septal defect. Urinary anomalies including hydronephrosis, renal dysplasia, horseshoe kidney, renal duplication, or vesicoureteral reflux were found in 11 patients (40.7% of 27 patients who had urinary system ultrasound) and genital anomalies including vagina atresia, uterus duplication, vagina duplication, hydrocolpos, vaginal mediastinum, or accessories agenesis were found in 18 patients (62.1% of 29 patients who had reproductive system ultrasound). Six patients had other malformations including palatoschisis, congenital equinovarus, radial dysplasia, intestinal malrotation, intestinal duplication, congenital short colon, urachal fistula, and Meckel's diverticulum.

After assessment by cystoscope or/and surgery, nine patients were categorised as type 1 cloaca, 16 patients were type 2, and six patients were type 3. The mean CC length (measured by cystoscopy or surgery, FCS and MRI respectively), mean distance of the skin to rectum, and proportion of the accompanying anomaly in the urinary system, genital system, and cardiac systems are shown in Table 3.

Imaging analysis

Among the 31 patients, CC measurement at MRI was obtained for 24 patients (77.4%), using FCS in 24 patients (77.4%), and using a cystoscope in 25 patients (80.6%). Pairwise comparisons were performed and compared using the Wilcoxon signed-rank test (FCS versus MRI: $z=-0.626$, $p=0.801$, 95% confidence intervals [CI]=0.793 to 0.809; MRI versus cystoscopy: $z=1.688$, $p=0.092$, 95% CI: 0.086 to 0.097; FCS versus cystoscopy: $z=1.384$, $p=0.177$, 95% CI: 0.170 to 0.185). No significant differences were found in these pairwise comparisons ($p>0.05$). Comparing with the result of the CC length measured using the cystoscope, 20 patients (83.3%) had accordant classification measured at MRI whereas four patients had discordant measurements, for the result measured with FCS, the result was 17 (70.8%) accordant and seven discordant measurements; the difference was not significant statistically ($p>0.05$).

Discussion

PC is probably the most complicated situation encountered by paediatric surgeons who treat patients with anorectal malformations, because it is notorious not only for its high rectum position, but also for the variety of complications such as spinal cord problems, sacrococcygeal malformations, Müllerian anomalies, and urinary system deformities.[4] These associated situations make the assessment of the PC very challenging.[3] The best functional outcome can only be achieved when different imaging methods are applied preoperatively to fully understand the malformed anatomy before patients are taken to the operating room for reconstruction surgery.

MRI and FCS are the two most frequently utilised methods in PC assessments. Traditionally, FCS was performed to visualise the CC and proximal structures and required patients to be catheterised; repeated effort was needed to get into the correct path. The optimal moment of exposure is difficult to capture, and the results are usually

Table 2
Clinical data in 31 patients with persistent cloaca.

Case	Age (m)	Vertebral and spinal cord anomalies			Rectum End (cm)	Urinary anomaly	Genital anomaly	Cardiac anomaly	Type	CC (cm)			Surgical pattern
		TCS	Spine cord	Sacral						FCS	MRI	CS	
1	84	+	FL	Normal	3.5	Normal	Normal	PFO	2	N/A	1.6	N/A	Colostomy
2	76	Normal	Normal	Normal	2.7	Normal	Normal	PF0	2	1.8	N/A	N/A	Colostomy, PSARP
3	69	Normal	Normal	Normal	3.5	Normal	HV	Normal	2	1.7	1.5	2.5	Colostomy, TUM
4	49	+	Normal	Dysplasia	3.7	Bilateral KD	Normal	Normal	1	1.2	1.5	1	Colostomy, VPWM
5	44	+	Myelocoele, FL	Dysplasia	3.3	Right KA	Normal	PFO, PDA	2	1.3	1.6	1.8	Colostomy, TUM
6	96	Normal	Normal	Normal	5.8	Horseshoe kidney, Bilateral ureteral dilatation	HV, HU, hydrocolpos	ASD; PDA	2	1.7	2.0	2	Colostomy, cystostomy, TUM
7	74	Normal	Normal	Normal	4.8	Left KD, right kidney cyst	Hydrocolpos	PFO	2	1.1	1.4	1.5	Colostomy, VR
8	188	N/A	Scoliosis, T10 deformity	N/A	N/A	N/A	N/a	ASD	3	N/A	N/A	3	Colostomy, PSARP
9	69	Normal	Normal	Normal	2.9	Normal	Normal	Normal	2	N/A	1.2	1.5	Colostomy
10	52	Normal	Normal	Normal	4.4	Normal	Vaginal septum	VSD	1	0.8	0.5	0.5	Colostomy, VPWM
11	41	Normal	Lipoma	Normal	4.2	Normal	Normal	Normal	2	1.3	1.2	1.5	Colostomy, VPWM
12	40	Normal	Normal	Normal	7.6	Normal	HV, HU	ASD	1	0.8	1.0	0.8	Colostomy, VPWM
13	37	Normal	FL, Vertebral deformity	Normal	3.1	Bilateral ectopic kidney and ureteral dilatation, Vesicoureteral reflux, left KD,	Hydrocolpos	PFO	1	1.4	0.9	N/A	Colostomy, VPWM
14	44	Normal	Normal	Normal	1.5	N/A	HV, HU, hydrocolpos	Normal	3	N/A	N/A	5	Colostomy, Cystostomy, PSARP, VR
15	32	N/A	N/A	Dysplasia	3.5	N/A	N/a	PFO, PDA	1	0.9	N/A	0.5	Colostomy, PSARP
16	33	Normal	Normal	Normal	3.5	Left KA, right ureter ectopic opening.	Hydrocolpos, Vaginal atresia	PFO	1	0.9	0.8	N/A	Colostomy, VPWM
17	23	Normal	Normal	Normal	3.4	Normal	Normal	Normal	2	N/A	1.2	2	Colostomy, TUM
18	16	Normal	Normal	Normal	3.6	Normal	Normal	PFO	Type 2	N/A	1.0	1.5	Colostomy, TUM
19	11	+	FL	Dysplasia	2.6	Hydronephrosis (bilateral), duplication of right kidney	Normal	PFO	Type 2	1.2	1.4	N/A	Colostomy, nephrostomy
20	39	Normal	Normal	Normal	N/A	Hydronephrosis (left), right ureter ectopic opening.	HV, HU, hydrocolpos	Normal	Type 3	1.6	2.4	4.0	Colostomy, nephrectomy (left), VR
21	22	Normal	Normal	Normal	4.4	Normal	HV, HU	Normal	Type 2	2.2	2.0	2.3	Colostomy, TUM
22	129	Normal	Sacral canal cysts	Normal	3.9	Ectopic kidney, dysplasia (left)	Vagina absent, bicornuate uterus	Normal	Type 3	3.5	3.5	3.5	Colostomy
23	16	Normal	Normal	Normal	3.2	Normal	HV, HU	PFO	Type 2	1.8	2.3	2.0	Colostomy
24	14	Normal	Normal	Normal	3.3	Normal	Normal	PFO	Type 2	2.3	2.3	2.0	TUM
25	13	Normal	Normal	Dysplasia	3.6	Normal	HU, Vaginal atresia	PFO	Type 3	3.2	2.7	4.8	Colostomy, VR,
26	16	+	Sacral canal cysts, FL	Dysplasia	3.1	Normal	HV, HU, hydrocolpos	PFO	Type 2	1.6	1.4	1.5	Colostomy, TUM
27	26	+	Vertebral deformity	Dysplasia	4.3	Right KA, left KD,	Vaginal dysplasia	Normal	Type 2	1.6	N/A	1.8	Colostomy, TUM
28	7	Normal	Normal	Normal	2.0	Normal	Normal	PFO	Type 1	1.6	1.2	1	PSARP
29	10	N/A	N/A	N/A	3.7	Normal	Vaginal and uterus agenesis	PFO	Type 3	2.1	N/A	3	Colostomy, VR
30	20	N/A	N/A	Normal	2.0	Duplicated bladder	HV, HU	Normal	Type 1	N/A	1.0	N/A	Colostomy, PSARP
31	16	N/A	N/A	Dysplasia	N/A	N/A	Hydrocolpos	N/A	Type 2	2.1	N/A	1	Colostomy

FL, Filum lipoma; TCS, tethered cord syndrome; KD, kidney dysplasia; KA, kidney agenesis; HV, hemi-vaginas; HU, hemi-uterus; PFO, patent foramen ovale; PDA, patent ductus arteriosus; ASD, Atrial septal defect; VSD, ventricular septal defect; PSARP, post sagittal anorectal plasity; TUM, total urogenital mobilisation; VPWM, vaginal posterior wall mobilisation; VR, vagina replacement.

Table 3
Summary of 31 persistent cloaca patients underwent FCS and MRI.

		Type 1	Type 2	Type 3	Total
Average CC length (cm)		0.8	1.8	3.9	2.1
CC in FCS (cm)		1.2	1.7	2.2	1.7
CC in MRI (cm)		0.6	1.6	2.9	1.6
Vertebral column and spinal cord	TCS	2 (2/7)	3 (3/15)	2 (2/6)	7 (7/28)
	Sacral anomalies	4 (4/8)	3 (3/14)	3 (3/6)	10 (10/28)
Rectum termination (cm)		3.7	3.6	3.2	3.6
Urinary anomalies		4 (4/7)	6 (6/16)	5 (2/5)	15 (15/28)
Genital anomalies		6 (6/8)	8 (8/16)	3 (3/5)	17 (17/29)
Cardiac anomalies		5 (5/9)	10 (10/16)	3 (3/6)	18 (18/31)
Other anomalies		2 (2/9)	0 (0/16)	4 (4/6)	6 (6/31)
Total		9	16	6	31
Accuracy (comparing with CS or surgery, accordant/total)	FCS	4/8 (50%)	11/12 (91.7%)	2/4 (50%)	17/24 (70.8%)
	MRI	5/7 (71.4%)	14/14 (100%)	1/3 (33.3%)	20/24 (83.3%)
	p-Value	0.71	0.27	0.66	0.30

FCS: fluoroscopy contrast study; CC, common canal; CS, cystoscope; TCS, tethered cord syndrome.

confusing to interpret.[3] From the authors' experience, the most difficult task was to display the urethra, vagina, and rectum at the same time, as the patients were not deeply sedated during examination because of necessity of autonomous voiding of the bladder. Even if we had sedated the patients, it was difficult to show the CC during the examination because balancing the benefits of measuring the length of the CC and the risks of radiation is very challenging. In patients with a longer CC, it was even more difficult to catheterise and show the CC in one attempt.

Compared to FCS, MRI is radiation free and is considered one of the most suitable techniques for paediatric imaging. Some investigators advocate routine MRI as an adjunct to planning anorectal pull-through, owing to the ability of MRI to assess the developmental state of SMC and other associated anomalies of the spinal cord, spine, and urogenital system[7,9–11]; however, MRI is not considered an accurate way to measure the CC because of poor detection of thin tubular structures (urethra, common canal, and rectal fistula).[7] Other methods have been attempted to substitute direct measurement of the CC, but without success. In the present study, an attempt was made to measure the CC from preoperative MRI; the measurement method was feasible and interestingly, MRI measurement of the CC was found to be comparable to FCS or cystoscopy in predicting categorisation. These results were valuable in guiding decisions regarding the surgical approach.

For complicated anomalies, such as PC, MRI and FCS were performed during neonatal period and repeated before radical surgery to gather more information about the development of accompanying malformations, such as intraspinal lesions and tethered cord. There were no difficulties interpreting the MRI examinations in neonates and small infants. ESPR 2017[8] recommended the use of feeding tubes inserted in the CC to be used an indicator of the pelvic structure during the MRI examinations. The present authors have done this in selective cases. During the procedure, in short CC patients ($\leq$1cm), it was found that this tube was very difficult to keep in place during the examination. The depth of tube insertion could also be a problem: if the tube went in too shallow or too deep, it could interfere with instead of indicate length measurement while interpreting the images. Without the tube, the pelvic structures could be visualised satisfactorily. The average age of the present cohort was 211 days, for small patients such as these, the quality of the $\leq$2 mm section thickness image was poor, resolution was low, and measurement was not accurate in this situation. In the authors' experience, a small amount of urine is always found in the urethra and vagina, and sometimes there is hydrocolpos; these are clearly displayed on T2-weighted (W) images, providing a basis for measuring the length of the CC (Fig 1a and b). Three observations were noted when measuring the CC at MRI: (1) putting liver oil on the skin of the perineum as a surface marker before examination provides more objective and accurate results; (2) the entire length of the urethra and vagina is not always displayed on a single sagittal MRI image. In this situation, the CC can be measured more accurately in the thinner layer (3–5 mm, 0 interval) via axial T2W imaging. The vaginal and urethral structures are more recognisable by radiologists and paediatric surgeons at axial MRI. Due to the thin layer and absence of gaps, measurement of the CC using axial images is more accurate than sagittal images; (3) when there was vaginal dysplasia or absence, it was also difficult to observe and measure the CC accurately in the sagittal position.

In the present study, the accuracy of measurement at FCS and MRI may vary by patient. Patients with longer CC might have less accurate CC measurements. This is probably because (1) long CCs are more difficult to intubate during FCS. It was also difficult to show the urethra and vagina at the same time. When the CC was measured at MRI, the amount of residual fluid in the vagina may be reduced due to the increased length of the CC, which could also decrease the accuracy of measurement; (2) the measurement of shorter CC was more challenging at FCS because the female urethra is short and very difficult to distinguish from the CC. With the high soft-tissue resolution, measurements of the CC at MRI were more accurate for short CC compared with longer CC; (3) both FCS and MRI had the same accuracy in patients with type 2 PC (1 cm $<$CC $<$3 cm).

Bowel termination, genital system, urinary tract, vertebral and spinal anomalies, SMC and urethral length, relative hiatal distance, and vaginal volume can all be observed and measured in one MRI study in PC patients, as previously reported by Shaimaa and colleagues.[6] Additionally, MRI is less invasive, involves no radiation, and is advantageous in allowing assessment of pelvic structures. On the other hand, even with rotational fluoroscopy and three-dimensional reconstruction,[11] interpretation of FCS results can still be very confusing.

Based on this result, repeated efforts to visualise the CC would no longer be mandatory during FCS and MRI may replace conventional imaging of the PS in measuring CC length.

The main limitation of the present study was the relatively small sample size. There were only 16 cases in which the CC was measured simultaneously using FCS, MRI, and cystoscopy. Secondly, only three doctors were recruited for image analysis; although interobserver variability was satisfactory, a future study could investigate the repeatability and reliability further. In the present study, the CC was measured as a straight line in the sagittal sections. This might cause deviation in patients with a long curved CC. The effect of categorisation and surgical decision-making should be investigated in future studies. Furthermore, the MRI sequences available were limited by the retrospective nature of the study, and additional sequences would be required to meet the most recent European guidelines.

Compared with ultrasonography and FCS, MRI can be used to accurately demonstrate genitourinary anomalies and the length of the CC in patients with PC, helping surgeons to develop a reliable reconstruction plan before sending the patient to the operating room.

Conflict of interest

The authors declare no conflict of interest.

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