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Contrast enhanced colostography: New applications in preoperative evaluation of anorectal malformations☆

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ABSTRACT

Introduction: Understanding details of anatomic relationships between the colon and surrounding structures is a critical piece of preoperative planning prior to surgical repair of anorectal malformations (ARMs). Traditional imaging techniques involve ionizing radiation, distention of the rectum with supraphysiologic intraluminal pressures, and sometimes require sedation. Recent developments in the field of contrast agents have allowed the emergence of an ultrasound-based technique that can avoid these requirements while continuing to provide high resolution structural information in three dimensions.
Methods: Fourteen children (13 male, 1 female, age 1–11 months) with ARMs underwent contrast enhanced colostography (ceCS) in addition to traditional preoperative imaging techniques to delineate anatomic relationships of pelvic structures.
Results: ceCS and traditional imaging yielded concordant anatomic information, including structural relationships and fistulous connections, in 10/14 patients (71%). ceCS detected fistulous connection in 2/13 patients (15%) that were not seen by traditional imaging. Ultrasonography failed to detect the fistulous connection in one patient.
Conclusions: ceCS is a safe, effective and flexible method for defining important structural information in ARM patients. When compared with traditional methods, it provided equivalent or superior results 93% of the time and bears consideration as a standard tool in preoperative planning for this population.
Type of Study: Retrospective Comparative Study.
Level of Evidence: Level III.

Understanding details of anatomic relationships between pelvic structures is a critical piece of preoperative planning prior to surgical repair of anorectal malformations (ARMs). Of specific clinical importance is the location of the fistula (if present) between the rectum and the urinary tract, as this can direct the surgical approach. The location of the rectum, in relation to the levator muscle complex and tip of the coccyx, and the location of the fistula to the urethra or vagina may all influence the operative approach. The fistula location is regarded by some as the most important piece of information to be defined when planning repair of an ARM. [1]

Traditional imaging techniques aimed at elucidating these relationships involve ionizing radiation, may require sedation, and distend the rectum with supraphysiologic intraluminal pressures. Recent developments in the field of contrast agents have allowed the emergence of an ultrasound-based technique that can avoid these requirements while continuing to provide high resolution structural information in three dimensions.

Contrast-enhanced ultrasound (CEUS) utilizes contrast agents that are suspensions of gas microbubbles encapsulated by a stabilizing outer shell. The physical properties of these microbubbles cause increased echo-scattering, resulting in an enhanced ultrasound signal [2]. CEUS in children has found broad application, including evaluation of vesicoureteral reflux (VUR) [3–5], and as well as intraabdominal solid organ injury and lesion characterization [6]. The 2016 FDA approval of the US contrast agent Lumason (sulfur hexafluoride lipid-type A microspheres; Bracco Diagnostics Inc., Monroe Township, NJ) for diagnosis of VUR and focal liver lesions in children has expanded the adoption of CEUS by pediatric imagers in the United States.

Over the past several years, several ARM patients at our center have undergone preoperative imaging using both traditional fluoroscopy as well as CEUS. A small subset of this population has previously been presented in a limited capacity in the radiology literature. [7] We hypothesized that CEUS is a sensitive mode of imaging the distal colon and associated fistulas in anorectal malformation patients so reviewed experience with a larger cohort.

☆ Declarations of Interest: None

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1. Methods

This retrospective study was approved by the Office of Clinical Research Investigations Internal Review Board at our institution (IRB-P00032845).

Fourteen children (13 male, 1 female; age range 1–11 months) with clinical evidence of anorectal malformations underwent traditional fluoroscopic imaging (as is the current standard practice) to delineate anatomic relationships of pelvic structures, as well as contrast enhanced colostography (ceCS) for comparison purposes. Ultimate diagnoses were based on the results of imaging and included 11 rectourethral fistulas, 1 imperforate anus without fistula, 1 rectoperineal fistula, and 1 occult rectoperineal fistula (which had been suspected based on clinical exam, but not confirmed). .

CeCS was performed in the manner previously described [7]. 0.25–0.30 mL of Lumason was mixed in 250 mL of 0.9% sodium chloride solution. A Foley catheter was placed into the mucus fistula and the contrast solution was instilled via gravity drip through the catheter (without additional manual pressure) into the distal colon. The distal colonic, genital, and urinary tracts, and associated fistulas, were evaluated with a LOGIQ E9 ultrasound machine (GE Healthcare, Milwaukee, WI), using linear (9 MHz) and curved array (2–9 MHz) transducers. Imaging was performed from anterior sagittal, posterior sagittal, and perineal approaches. Ultrasounds for these patients were performed by three radiologists with experience in contrast enhanced ultrasound. Patients also underwent standard fluoroscopic colostography performed in the same manner [8]. Contrast was initially instilled by gravity in all patients and then under high pressure to distend the colon if no fistula was visualized fluoroscopically. The radiologists performing and interpreting the studies were not blinded to the results of the other imaging technique.

Two patients had mucous fistulas that could not be accessed by the Foley catheter. Of these two, one underwent voiding cystourethrography and contrast enhanced voiding urosonography (ceVUS); the other underwent ceCS in the operating room after the fistula was opened.

Most of the patients (8/14) underwent both imaging studies on the same day. In three patients the studies were separated by several days (3–25 days), in one patient the interval was 10 weeks, and one patient only underwent ceCS in the operating room, as described above. No sedation was required for any of the studies.

Results obtained by ceCS were compared to those obtained by colostography (Fig. 1). Operative notes were reviewed, and intraoperative findings were compared to the preoperative fluoroscopic and ceCS diagnoses. (See Table 1.)

2. Results

CeCS and colostogram yielded concordant anatomic information, including structural relationships and fistulous connections, in 10/14 patients (71%). One patient did not have a fistula. CeCS detected fistulous connections in 2/13 patients (14%) that were not seen by initial fluoroscopic colostography. One of these two patients underwent a second

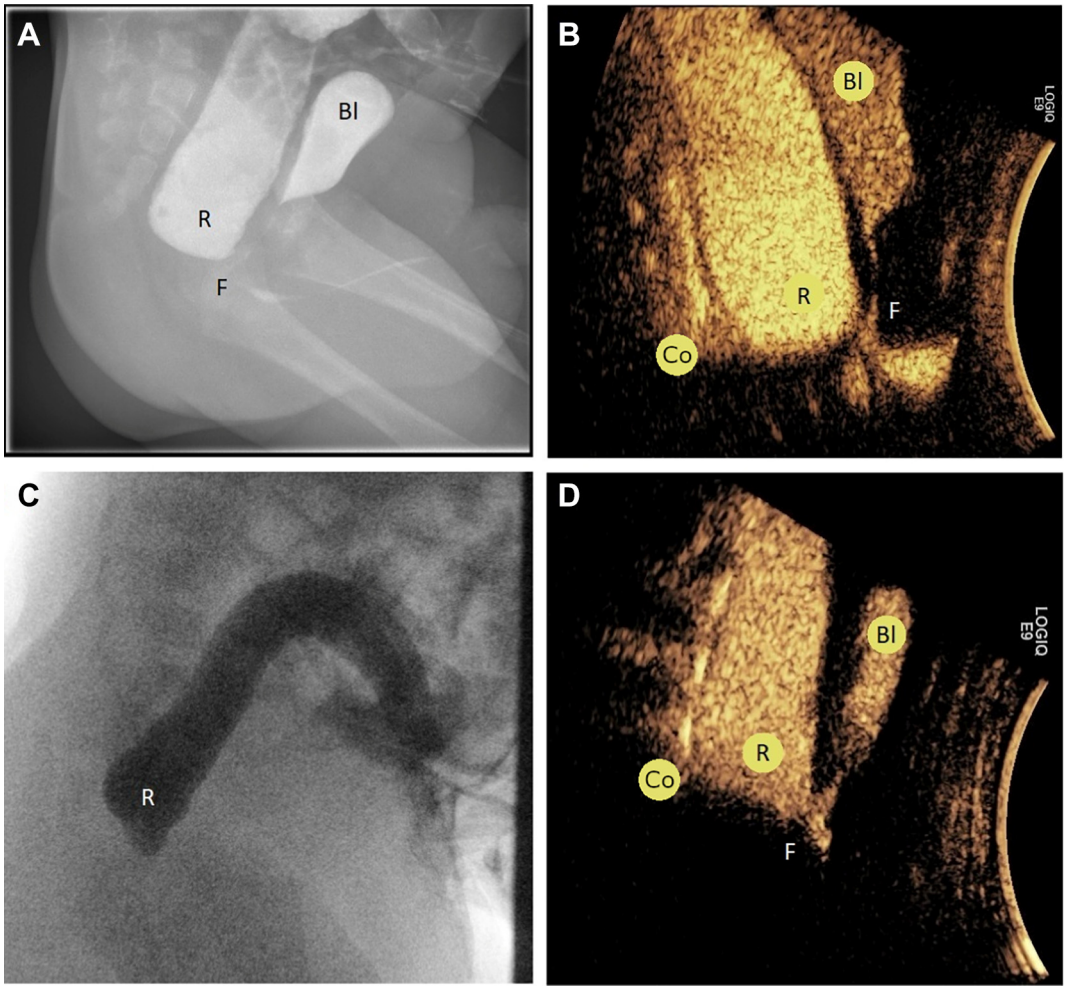

Fig. 1. Comparison of traditional colostogram (A, C) with contrast enhanced ultrasound (B, D). Abbreviations: rectum (R), bladder (Bl), fistula (F), coccyx (Co). Images A and B are obtained from the same patient, and both methods successfully identify the rectourethral fistula. Images C and D are from a different patient, in which the contrast enhanced ultrasound identified a fistula that was not demonstrated on colostogram.

Table 1
Clinical characteristics.

Pt	Gender	Age at imaging (days)	Age at surgery (days)	Fistula type	Identified by colostogram	Identified by ceCS
1	M	92	322	Rectobulbar	Y	N
2	M	110	195	Rectoprostatic	Y	Y
3	M	31	93	Rectobulbar	Y	Y
4	M	336	414	Rectoperineal	Y	Y
5	M	208	233	Rectourethral (not localized)	Y	Y
6	M	198	208	Rectoprostatic	Y	Y
7	M	172	172	Rectocutaneous	n/a	Y
8	M	170	229	Rectoprostatic	Y	Y
9	M	94	213	Rectobulbar	N	Y
10	F	115	131	No fistula	Y	Y
11	M	130	208	Rectoprostatic	N, Y	Y
12	M	110	122	Rectoprostatic	Y	Y
13	M	57	119	Rectobulbar	Y	Y
14	M	87	112	Rectoprostatic	Y	Y

colostogram with high pressure, which ultimately demonstrated the fistula. The second patient's fistula was not visualized on colostography performed with high pressure manual injection. In 2 patients, there was no patent mucous fistula amenable to catheterization at the time of preoperative study so distal colostography could not be performed. One of these patients underwent ceCS in the operating room after opening of the mucus fistula, and the preoperatively suspected rectoperineal fistula was confirmed. The second patient underwent preoperative voiding cystourethrography (VCUG), which demonstrated a rectourethral fistula. A ceVUS was also performed, which did not demonstrate the fistula.

Operative notes were also reviewed. When precise anatomic location of the urethral component (prostatic vs bulbar) of the fistula was described, the imaging findings were concordant in 75% (6/8) of colostograms and 78% (7/9) of the ultrasound studies. Discordant findings were due to inability to localize the fistula on imaging or nonspecific descriptions of the fistula. One fistula was identified by ceCS that was too small to allow passage of a lacrimal duct probe in the operating room. This was suspected to be secondary to size, not tortuosity.

There were no adverse events associated with the ceCS procedures or ultrasound contrast administration.

3. Discussion

It is critical to define anatomic relationships and aberrant connections between the colon and the urinary tract in ARM patients. One review of approximately 1000 ARM patients demonstrated not only that the urethra was the most common site of urologic injury during repair, but that urologic injury was associated with lack of preoperative imaging that clearly defined the level or presence of a fistula [9]. The use of preoperative pressurized distal colostography has therefore been advocated as the most important component of preoperative planning [1,8].

In our cohort, ceCS provided equivalent or superior diagnostic information in 13/14 (93%) patients when compared to traditional fluoroscopy. CeCS had a diagnostic accuracy of fistula identification (or exclusion) of 92% (12/13) compared to 85% for colostography (11/13), consistent with prior investigations [10]. Overall, our data suggest that ceCS is diagnostically accurate, with high sensitivity for detection of small fistulas.

In considering diagnostic accuracy, we note that the patient in whom ceCS failed to demonstrate a fistula did not have contrast injected in the same manner as the rest of the cohort. Since his mucous fistula had closed, he underwent ceVUS instead. The urethral catheter was left in place during the study (to facilitate the VCUG that followed) and we suspect that the catheter may have occluded or obscured the fistula. On retrospective review of this patient's ultrasound images, there is an echogenic focus in the area where we would anticipate finding the fistula. We also highlight the ceCS study that was performed intraoperatively, which demonstrated a fistula that was too small for cannulation by 0000 lacrimal duct probe (0.45 mm diameter), illustrating the sensitivity of this modality.

We also found that ceCS has the benefit of delineating bony anatomy. Initial ceCS studies were performed primarily as a proof of concept to evaluate the anatomy of the rectourethral fistula. In subsequent patients, more focus was placed on evaluating the location of the coccyx and its relationship to the rectum and fistula, as this can direct operative approach. In all patients with ceCS performed in the latter cohort of patients, this relationship is demonstrated on ceCS.

The ceCS procedure is safe. No adverse events associated with ceCS were noted. No sedation was needed to perform ceCS, and for the patient who underwent ceCS in the operating room (in conjunction with his ARM repair), the use of general anesthesia and positive pressure ventilation did not interfere with the ability to perform ceCS. No ionizing radiation is necessary to perform ceCS, and no manual distention of the colon is needed to demonstrate fistulous connections. Colonic perforation associated with pressure-augmented distal colostography is rare, but has been reported [1,11]. Avoiding the need for additional intraluminal pressure may make ceCS safer that fluoroscopic distal colostography.

The contrast agent also appears to be safe for use in this application. Studies regarding safety of Lumason (known as Sonovue outside of the United States) suggests a robust safety profile in intravenous administration in adult patients [12,13]. Although dosing recommendations exist for intravenous administration, appropriate dosing for intracavitary administration is less well defined [6]. The diagnostic procedure most well studied in pediatric patients is ceVUS, in which a contrast agent is instilled into the urinary bladder to evaluate for vesicoureteral reflux. The total dose and concentration of contrast agent used here is similar to or less than the recommended pediatric intravesical dosing for ceVUS. Although no guidelines yet exist for intracolonic administration of Lumason, the relative equivalence between the dose used for this application and the dose used for ceVUS implies that a similar safety profile may be anticipated.

Lastly, ceCS is more flexible than traditional colostography. Due to the portable nature of ultrasound, this diagnostic study can be performed in a broader array of clinical environments than fluoroscopic colostography. This is well illustrated by comparing the two patients in our cohort who had mucous fistulas that were too small to permit catheterization for purposes of colostography. One patient, as previously described, underwent ceCS in the operating room after his mucus fistula was opened, and the rectal fistula was accurately identified. The other patient underwent VCUG and ceVUS in the radiology suite. Although ceVUS did not identify the fistula, it is present on retrospective review, and the flow of contrast was likely impaired by the indwelling Foley catheter. Future ceVUS studies performed without an indwelling catheter will likely have a higher diagnostic yield. Larger patient cohorts are clearly needed to accurately assess the true sensitivity and specificity of ceCS, but the flexibility advantages of ceCS are clear.

Despite its advantages, ceCS is not without limitations. Ultrasound studies can be highly operator dependent, and thus high levels of specialty training necessary in radiology and pediatrics may be a limiting factor in the broad dissemination of this technique. Although preliminary data suggest a reassuring safety profile [14,15], administration of these agents into the gastrointestinal tract has not been well studied. Interpretation of these results are to be tempered by the typical limitations of a retrospective analysis. Fluoroscopy and ceCS were frequently performed by the same radiologists, and none of the radiologists interpreting the imaging studies were blinded to the results of the other study.

Based on our preliminary experience, ceCS appears to be a safe, effective and flexible method for determining structural information relevant for preoperative planning in ARM patients. With appropriate education, this technique should achieve broad adoption. The

radiologists involved in this study have begun to educate other members in their department and we anticipate its use will continue to increase. Structured prospective studies will be helpful in further refining specific use indications for ceCS and may help reshape the preoperative workflow for the ARM population.

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Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.jpedsurg.2020.09.033.

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