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Comparison of trans-perineal ultrasound-guided pressure augmented saline colostomy distension study and conventional contrast radiographic colostography in children with anorectal malformation

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ABSTRACT

Background: In children with high and intermediate anorectal malformation, distal colostography is an important investigation done to determine the relationship between the position of the rectal pouch and the probable site of the neo-anus as well as the presence or absence of a fistula. Conventionally, this is done using contrast with fluoroscopy or still X-ray imaging. This, however, has the challenges of irradiation, availability and affordability, especially in developing countries. This study compared the accuracy of trans-perineal ultrasound-guided pressure augmented saline colostomy distension study (SCDS) with conventional contrast distal colostography (CCDC) in the determination of the precise location of the distal rectal pouch and in detecting the presence and site of fistulous communication between the rectum and the urogenital tract was studied. **Materials and Methods:** Trans-perineal ultrasound-guided pressure augmented SCDS, CCDC and intra-operative measurements were done sequentially for qualified infants with anorectal malformation and colostomy. Pouch skin distance and presence or absence of recto urinary or genital fistula was measured prospectively in each case. Statistical significance was inferred at *P*-value of <0.01. **Results:** There were thirteen infants, 9 males and 4 females. The age at onset of investigation ranged from 2 to 12 months with a median value of 9 months. Using paired *t*-test at a confidence interval of 95%, the *P* value when SCDS values are compared with CCDC is 0.19; and 0.06 when SCDS was compared with intra-operative measurements. Hence, there is no statistical difference as $P > 0.01$. On its ability to detect presence or absence of a fistula: SCDS had a sensitivity of 50.0%, specificity of 100.0%, accuracy of 69.2%, negative predictive value of fistulas of 55.6% and a positive predictive value of fistulas of 100.0%. **Conclusion:** Ultrasound-guided pressure augmented SCDS can safely and reliably be used to assess the distal colonic anatomy and the presence of fistula in infants with Anorectal malformation who are on colostomy.

Key words: Anorectal, colostography, colostomy, congenital, loopogram, malformation, saline disten

INTRODUCTION

Anorectal malformation (ARM) is a wide spectrum of congenital anomalies of the distal gut ranging from minor lesions to complex anomalies with varying affectation of the uro-genital tract. Worldwide, the incidence is 1 in 5000 live births.[1,2] In Nigeria, majority of ARMs require colostomy.[3-10]

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The traditional ARM international classification and the current Krickenbeck classification are based on the relationship of the terminal rectal pouch to the levator ani muscle and the presence or absence of a fistula.[11,12] Colostomy, asides from diverting stool, which is necessary for the survival of these obstructed neonates, facilitates bowel preparation prior to the definitive surgery as well as protects the definitive repair.[13]

Furthermore, through the colostomy further radiological investigations are done to determine the position of the rectal pouch in relation to the probable site of the neo-anus and the presence or absence of a fistula. This information further delineates the pelvic anatomy and also guides the surgeon in selecting the type and approach of definitive surgery for the anorectal reconstruction. Pressure augmented distal colostography using water soluble contrast agent has been the choice investigative modality.[14] Ideally, the progress of the injected contrast material in the distal colon is monitored with fluoroscopy, but non-fluoroscopic still X-ray imaging has also been found useful.[15]

However, there are major drawbacks to the use of ionising contrast under fluoroscopy or still X-ray imaging. Exposure of young infants to radiation has continued to generate concerns. Even though the absolute risks of radiation exposure from these procedures are minimal, there is, nevertheless, a higher risk of cancer-related radiation injuries in exposed children compared to adults. This is because children are more sensitive to radiation than adults as they are still growing and with more actively dividing cells; their long life expectancy also increases the risk of manifestation of radiation injuries. Furthermore, improper radiation machine adjustment to match their small body size may result in high radiation doses with attendant cellular damage.[16]

In poor economies, fluoroscopy machines are either not available or expensive to access. These have made clinicians to develop other assessment methods such as the non-fluoroscopic pressure augmented colostography. This has only slightly reduced the cost of investigation, but is still not very affordable to many and has also not abolished the immediate and long-term risks of contrast radiological study.

We considered these problems of affordability and radiation risk and modified the existing study techniques by replacing contrast agent with isotonic saline solution and X-ray with ultrasound scan. The objectives of this study were to determine the accuracy of trans-perineal ultrasound-guided pressure augmented saline colostomy distension study (SCDS) compared with conventional contrast distal colostography (CCDC) in:

- Determining the precise location of the distal rectal pouch.
- Detecting the presence and site of fistulous communication between the rectum and the urogenital tract in patients with ARM, using surgical findings as the reference point.

MATERIALS AND METHODS

Consecutive infants with ARM who has undergone a prior defunctioning sigmoid colostomy were prospectively studied between July 2013 and February 2015. Nnamdi Azikiwe University Teaching Hospital Nnewi, Nigeria.

The sequence of study was SCDS, CCDC and surgery. Findings were recorded independently after each procedure. The first two were done at the radiology suite. Surgery was fixed at the conclusion of the colostography.

Institutional Ethical Review Board approval was sought for and obtained prior to onset of study.

A small metallic marker was secured at expected region of the anal skin by the radiologist, which attenuates sound at that location, and on ultrasound casts a strong posterior acoustic shadow. An Aloka Prosound SSD-3500SX (Hitachi Aloka Medicals Ltd, Japan) ultrasound machine with a curvilinear transducer (frequency of 3.5 to 5 MHz) was used for the study. Thermo neutral environment was maintained by warming up the radiology suite, and the baby positioned supine on the couch with hips abducted and knees flexed. Trans-abdominal and trans-perineal B-mode ultrasound was done to assess the abdominal cavity, especially the urogenital tract. Thereafter, an appropriate size Foley's catheter was introduced into the distal stoma and the balloon inflated and pulled back to secure it and to occlude the lumen. Under the guidance of trans-perineal ultrasound (longitudinal scan), 20-50 ml of warm saline was run into the distal colonic lumen through a fluid giving set connected to the catheter. The distal colon distended by saline is seen as hypo-anechoic endoluminal content. Figure 1a-c shows the ultrasound features in different types of ARM. The colonic blind end at the site of ano-rectal malformation was identified and its distance from the metallic anal marker measured and documented. Furthermore, any abnormal fistulous communication



Figure 1: Saline colostomy distension study sonogram of different types of anorectal malformation. (a) Recto-urethral fistula (arrow; red: Urinary bladder, blue: Urethra, green: Fistulous track, yellow: Rectal pouch). (b) Recto-vestibular fistula (red: Vestibule, blue: Uterus, green: Rectal pouch). (c) Rectal atresia without fistula (yellow: Distended rectum tapering at site of anorectal malformation, red: Site of metallic marker, blue: Minimal fluid within urinary bladder

with the urinary tract, the vagina, or skin identified was documented. Compressing the drip bag may further increase the pressure and outline the fistula better. Where adequate insufflation is not obtained, a 50 ml syringe is used to gently and steadily increase the pressure.

At the X-ray room, a preliminary lateral film of the abdomen and pelvis including the sacral region was done. This image shows the aforementioned metallic marker projected over the gluteal shadows. With patient lying supine, the distal colon will be distended with 20-40 ml of dilute Gastrografin injected into its lumen through the aforementioned *in situ* Foley's catheter, and a post-contrast lateral film taken with a gentle pressure still being maintained. The distance between the colonic blind end and the aforementioned metallic marker was measured with a rule and documented. Any feature of fistulous tract with the urinary or genital tract was documented [Figure 2].

All the patients had posterior sagittal ano-rectoplasty.

Intraoperatively, the child was placed in the prone position with the pelvis elevated. After routine surgical skin preparation, posterior sagittal incision was made. Once the rectum was identified, the distance from the end of the pouch to the skin overlying the presumed neo anus is measured off with blade handle, read off a tape measure and documented [Figure 3]. After this, further dissection continues. The presence of fistulae and their position were carefully sought for and noted. The two infants with rectovesical fistula also had additional abdominal approach where the distance was measured with the perineal tube inserted into the pelvis for the pull-through.

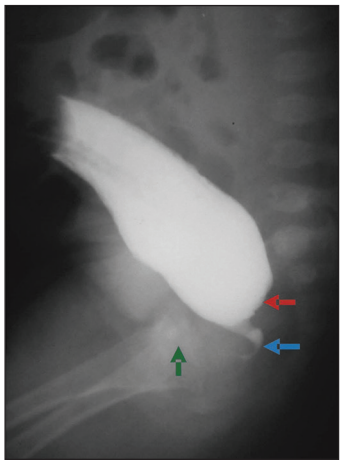


Figure 2: Contrast radiographic colostogram (red arrow: Distal colon/ rectal pouch, blue arrow: Fistulous track, green arrow: Minimal contrast at region of the bladder neck

Statistical analysis of the data was done using SPSS Statistics software(IBM Corp. Released 2012. IBM SPSS Statistics for Windows, Version 21.0. Armonk, NY: IBM Corp.). Associations between categorical variables were explored with the paired *t*-test, while correlation between the reference test (surgical measurements) and radiological test was determined with Pearson correlation. Statistical significance was inferred at $P < 0.01$.

RESULTS

There were 13 infants, 9 males and 4 females. The age at onset of investigation and surgery ranged from 2 to 12 months with a median value of 9 months.

The frequency of different types of malformation is as indicated in Table 1 and the summary of measurements in Table 2.

Comparing the SCDS and CDCC measurements (null hypothesis is that there is no difference between the measurements, any difference is attributable to chance occurrence), Table 3 shows the correlation between SCDS and CCDC, while Table 4 shows the comparative ability of the test to detect absence or presence of recto urogenital fistula.

Furthermore, using Wilcoxon Signed rank test, the *P*-value when SCDS is compared to CCDC is 0.181; and 0.113 when SCDS is compared with intra-operative values. The *P* value is considered significant if $P < 0\ 05$.

(Pearson correlation: SCDS vs. CCDC = 0.89; SCDS vs. intra-operative values = 0.84).

Table 1: Type of malformation (intra-operative finding)

Malformation type	Frequency
Rectovesical fistula	2
Recto bulbar fistula	4
Vestibular fistula Pouch	2
No fistula	4
Cloaca	1
Total	13

Table 2: Patients and measurements

Patient number	Pouch-skin distance (cm)		
	SCDS	CCDC	Intra-operative
1	4.2	5.2	6.0
2	5.0	5.4	6.5
3	2.6	2.4	2.5
4	2.5	2.9	2.5
5	2.6	3.5	2.4
6	3.0	3.0	2.5
7	5.0	5.4	5.0
8	2.7	3.0	4.0
9	2.3	3.7	3.0
10	2.8	2.2	3.0
11	2.1	2.2	2.3
12	2.0	1.2	1.8
13	3.5	3.3	6.5

CCDC: Conventional contrast distal colostography; SCDS: Saline colostomy distension study

DISCUSSION

From our study, there was no statistical difference between SCDS values when compared with intra-operative measurements. The Wilcoxon signed rank test and paired *t*-test were both >0.05 when SCDS was compared with either CCDC or intra-operative values. The null hypothesis is thus accepted. This implies that within acceptable limits the values of pouch-skin distance obtained by SCDS compares favourably to those obtained intra-operatively or by CCDC in the assessment of the distal colonic anatomy in infants with ARM.

The sensitivity and specificity of SCDS in detecting the presence or absence of fistula also compares favourably to CDCC.

The two key substitution agents in this study offer greater advantage when compared to the standard contrast radiography in the assessment of infants with ARM. Ultrasonography is a cost-effective, readily available, safe imaging modality free of ionising radiation. In evaluating infants with ARM, it is invaluable in investigating associated urinary tract anomalies and genital malformations such as hydrometrocolpos. Isotonic saline is a physiological

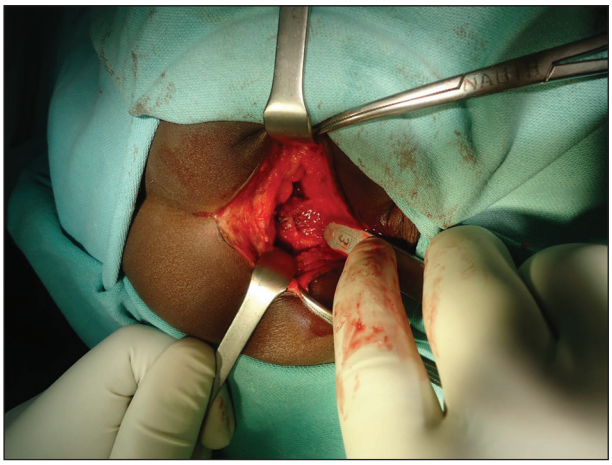


Figure 3: Intraoperative Pouch skin distance measurement

Table 3: Comparison of pouch-anal skin distance values from intra-operative measurement, SCDS and CCDC using paired *t*-test

Comparison	Paired differences					*t*	df	Significant (two-tailed)
	Mean	SD	SEM	95% CI of the difference				
				Lower	Upper			
Pair 1								
Intra-operative versus SCDS	0.5923	1.0218	0.2834	−0.0252	1.2098	2.090	12	0.059
Pair 2								
SCDS versus CCDC	0.2385	0.6252	0.1734	−0.1394	0.6163	1.375	12	0.194

CCDC: Conventional contrast distal colostography; SCDS: Saline colostomy distension study; SD: Standard deviation; SEM: Standard error of mean; CI: Confidence interval

Table 4: The ability to detect the presence or absence of co-existing fistula

Statistical test	SCDS* (%)	CCDC** (%)
Sensitivity	37.5	50.0
Specificity	80.0	100
Accuracy	53.8	69.2
Negative predictive value	44.4	55.6
Positive predictive value	75.0	100

Intraoperative measurements were used as the reference test. *SCDS: Ultrasound-guided saline colostomy distension study; **CCDC: Conventional contrast distal colostography

electrolyte solution devoid of allergic and anaphylactic reactions, which sometimes are associated with ionising contrast agents.

The technique of saline infusion and pressure augmentation employed in this study is an adaptation of the technique used for conventional contrast studies.[17]

Furthermore, ultrasound abolishes the small risk of cellular damage from radiation and the possibility of later malignant transformation. The high safety margin, low cost and the wide availability of ultrasonography make it a suitable alternative. A common drawback is observer dependency in ultrasound imaging, but recent study has documented a reduced inter-observer error in ultrasound-based investigations.[18] However, a wider application of this procedure is needed to evaluate and further reduce inter-observer error index.

This new investigative modality is of advantage, especially in developing countries where fluorometer and X-ray facilities are largely unavailable. Even where available the cost of accessing these services is sometimes too expensive for most impoverished parents who usually pay out of pocket for Medicare.

With the increased ultrasound usage, probably with time, the investigation may likely become an office procedure thus allowing paediatric surgeons to easily learn and start applying the procedure. This will guarantee first-hand knowledge of the pelvic anatomy prior to surgery. It can also remarkably obviate the long waiting time in accessing CCDC, thus reducing the time infants are made to be on colostomy as well as the rate of colostomy-associated complications and psychological burden on parents.

While acknowledging that the sample size used is small, definite generalisations cannot be made yet even though statistical analysis have been done. This is a preliminary study, multi-centre studies are recommended for far-reaching deductions.

CONCLUSION

Ultrasound-guided pressure augmented SCDS could be safely and reliably be used to assess the distal colonic anatomy and the presence of fistula in infants with ARM who are on colostomy.

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Nil.

Conflicts of interest
There are no conflicts of interest.

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