Case Series

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# Evaluating the utility of cystoscopy, distal colostography, and sonography for locating the fistula in patients with anorectal malformation: a case series

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**Background and purpose:** Anorectal malformations are congenital conditions ranging from a simple perianal fistula to a complex cloacal malformation. Since the precise determination of the location of the fistula is the central pillar in choosing the type of surgery, this study aims to evaluate and compare the efficacy of three techniques, transperineal ultrasound, distal colostography, and cystoscopy.

**Materials and methods:** This study was performed on patients with anorectal abnormalities who had undergone decompressive colostomy and were planned for anorectoplasty in the period from September 2017 to March 2019 in a pediatric surgical center. To answer our question, all three mentioned methods were conducted before the surgery and were compared with the intraoperative findings.

**Results:** Sonography, distal colostography, and the second cystoscopy findings were similar to intraoperative conclusions concerning the presence of a fistula in patients, whereas blind cystoscopy had 30% accuracy and similarity. Regarding the type of fistula sonography, distal colostography, and second cystoscopy each had 50, 37.5, and 10 inconsistency with the intraoperative findings. In all cases where a fistula was detected in blind cystoscopy, the location of the fistula was correctly determined by this modality. Data analysis on the pouch to perineum distance measurements obtained from sonography and colostography were significantly different from that of surgery.

**Conclusion:** The results of this study emphasize the need to perform several diagnostic modalities to determine the location and type of fistula to improve diagnostic accuracy.

**Keywords:** anorectal malformation, case series, cystoscopy, distal colostogram, fistula location, transperineal sonography

## Introduction

Anorectal malformation (ARM) is a comprehensive term comprising a broad range of congenital anomalies with defects and deformities in the distal regions of the gastrointestinal system like the rectum, the anal canal, the genitourinary system, and the sacral part of the spinal cord. The severity of the anomaly determines the outcome and future of the patient after treatment so that a simple perianal fistula will be well reconstructed and have an excellent function. At the same time, a complex malformation, for example, in the cloacal region, requires multiple treatment sessions and great care[1]. ARMs have an estimated prevalence of 2–6 in 10 000 births[2]. Anorectal anomalies are more commonly seen in male infants, whereas cloacal malformations are more prevalent in female infants[3]. About half of the patients with ARM have associated anomalies varying from genitourinary (40–50%), cardiovascular (30–35%), spinal cord (25–30%), gastrointestinal (5–10%), and VACTERL (vertebral defects, anal atresia, cardiac defects, tracheoesophageal fistula, renal anomalies, and limb abnormalities) (4–9%) anomalies[1].

Over time, there have been many classifications for ARMs comprising Ladd and Gross, International, Wingspread, Pena, and Krickenbeck[4]. Wingspread classification (1984), which categorized anorectal anomalies as low, intermediate, and high, was broadly used for a long time. Still, it was later discovered that this classification method did not have the best efficacy for indicating the best type of surgery needed for the patients[5]. The Pena classification emerged from the importance of the fistula site in patients' postoperative follow-up, which was derived from the result of the posterior sagittal anorectoplasty technique introduced by Pena[6]. Since the decision to choose the type of treatment is made only 24–48 h after birth, early diagnosis of the abnormality is of great importance. To achieve this goal, different measures have been considered, one of which is the prenatal diagnosis of abnormalities. Although prenatal diagnosis of ARM by the means of ultrasound is rare, but findings such as distended bladder with oligohydramnios, cystic masses of the pelvis or abdomen, hydrops fetalis, ascites, and intestinal dilatation raise the suspicion of

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ARM and require further diagnostic evaluations[7,8]. After birth, a careful perineal examination followed by a systemic assessment of accompanying anomalies should be performed. Further investigations include inversion radiography of the abdomen, prone cross table later view radiography, different sonography methods involving abdominal, transperineal, and infra-coccygeal, Computer tomography, and MRI of the pelvis, distal colostography, and cystoscopy[1,9,10]. Treatment of the ARMs varies from nonsurgical methods with repeated dilation of the tight sphincter to surgical treatments consisting of primary repair or colostomy[11]. Since it is very important to diagnose and choose the type of treatment in time, the determination of the best method in establishing the location of the fistula can help avoid using additional methods and also protects the patient from complications caused by them.

## Materials and methods

This case series study was conducted on patients with anorectal abnormalities who had undergone an urgent decompressive colostomy and were admitted to a tertiary center of pediatric surgery between September 2017 and March 2019 for anorectoplasty surgery. This study was approved by the Ethics Committee.

### *Inclusion and exclusion criteria of participants*

Initially, the inclusion and exclusion criteria were determined and were as follows:

Inclusion criteria: all patients with ARM who had undergone colostomy.

Exclusion criteria: all patients with ARM with a visible fistula in the perineum or vaginal vestibule.

### *Sampling technique and sample size*

Sampling was done with the total population technique and 10 patients with anorectal abnormalities who had undergone an urgent decompressive colostomy and were scheduled for anorectoplasty surgery later were included in the study.

### *Variables*

The variables of this study included the two groups of quantitative and qualitative variables. The quantitative variables of the study were age and pouch to perineum distance. Age and pouch to perineum distance values were obtained from the individual's birth certificate, and the distance reported in ultrasound and distal colostography and reported in units of day and millimeter, respectively. Qualitative variables were, the state of the muscle complex in each of the investigated methods, the rectourethral-/bladder neck fistula, the gluteal state in the preoperative examination, the perineal dimple, and the condition of the sacrum.

### *Implementation method and steps*

All patients were initially treated with fluids and antibiotics, and after resuscitation and additional investigations to identify associated anomalies. An anorectoplasty surgery was done after about three months.

### Imaging modalities

In all included individuals, before performing the anorectoplasty surgery, the pediatric surgeon in charge inserted a number 8 Foley catheter and a balloon filled with 3 cc of distilled water in the distal opening of the colostomy. Then, in the radiology department while the Foley catheter was fixed to the stoma opening with gentle tension, diluted barium was introduced into the distal pouch under a pressure of 20 mmHg.

*Sonography*. The perineal ultrasound was performed by the first radiologist as the first imaging modality. Through sonography, factors such as the condition of the sphincter muscles, location of the end of the rectal pouch, pouch to perineum distance, rectourethral fistula, and its location were inspected.

*Colostography*. Following the sonography, a contrast marker was placed in the perineum and the distal colostography was carried out in the lateral view by the same radiologist. A second radiologist unaware of the other findings interpreted the distal colostogram.

*Cystoscopy*. Without any prior knowledge of the results of perineal sonography and distal colostography, cystoscopy was performed in the lithotomy position to localize the location of the fistula. In cases where the fistula was not seen in the blind cystoscopy, after referring to the patient's documents and the results of ultrasound and distal colostography, a second cystoscopy was performed to detect the presence and location of the fistula.

### *Preoperative and operative assessments*

After cystoscopy, the surgical team changed the patient's position to prone to perform a thorough perineal examination to assess the position of the buttock, groove, and sacrococcygeal region. The sphincter muscles were also examined using a muscle stimulator. Afterward, an anorectoplasty surgery was performed using the posterior sagittal technique.

As the patient was in the prone position, a posterior sagittal incision was done and even bilateral traction was made to help maintain a midline dissection plane. In case of visible fistula traction sutures were placed around the fistula. In these cases the subsequent incision was then made circumferentially around the fistula and was continued posteriorly in the midline toward the coccyx. In cases with no visible fistula the posterior sagittal incision was made from just inferior to the coccyx and extended to the perineal body. After observing the white fascia the rectum was retracted with sutures. The rectum was opened longitudinally and anteriorly toward the level of the fistula. The dissection was performed laterally on both sides and once the colon was mobilized laterally, the anterior dissection was initiated. The fistula was marked and retracted inferiorly as the anterior wall of the rectum was freed from the posterior wall of the urethra. The rectum was finally freed to the level of the peritoneum. In order to prevent prolapse, a rectopexy was created to the posterior muscle complex when closing the incision. The circumference of the rectum was secured to the skin within the sphincter complex.

The pouch to the perineum was also measured by the pediatric surgeon using a sterile surgical ruler. At last, data related to the three diagnostic methods mentioned above were compared with the gold standard results of surgery.

### *Follow-Up*

All patients were scheduled for a visit by the pediatric surgeon in charge two weeks and one month after the operation in the pediatric clinic. No patient was lost to follow-up.

### *Analysis method*

The recorded data were analyzed by SPSS version 16 software.

The investigated characteristics were obtained by descriptive statistical methods and demonstrated as a frequency distribution. A paired *t*-test was used to compare quantitative variables obtained from two diagnostic methods. In all calculations, a *P*-value of less than 0.05 was considered as a significant level.

This case series has been reported in line with the PROCESS Guideline[12].

## Results

In this study, a total of 10 patients were studied, with an age range of 62–105 days, and the age distribution of the patients was normal. All patients were male. No changes in the initial plan appeared. No complications appeared.

### *Sonographic findings*

Sonography was performed on all patients. The results regarding the status of the fistula included: no fistula (20%) and recto-prostatic fistula (10%), rectobulbar fistula (50%), recto-membranous fistula (10%), and rectobladder neck fistula (10%). The muscle complex was well-developed in nine participants (90%) and undeveloped in one participant (10%). The average poouc to h#tonperineum distance was 16.80 ± 8.21 mm.

### Comparison between sonographic and intraoperative findings

In determining the presence of a fistula and the state of the muscle complex, ultrasound findings were 100% consistent with intraoperative findings. Regarding the type of fistula in patients who were proven to have a fistula during the surgery (eight patients), ultrasound observations in four (50%) patients were inconsistent with intraoperative findings. The results of the paired *t*-test for the pouch to perineum distance showed that the difference between the sonographic and intraoperative values had statistical significance ($P = 0.001$). In fact, perineal sonography estimates the distance as less than the actual value.

### *Distal colostography findings*

Distal colostography was performed in all patients. The results regarding the status of the fistula included: no fistula (20%) and recto-prostatic fistula (10%), rectobulbar fistula (40%), recto-membranous fistula (10%), and rectobladder neck fistula (20%). The average pouch-to-perineum distance was 21.00 ± 8.75 mm.

### Comparison between distal colostography and intraoperative findings

In determining the presence of a fistula, distal colostography findings were 100% consistent with intraoperative findings. Regarding the type of fistula in patients who were proven to have a fistula during the surgery (8 patients), distal colostograpy observations in three (37.5%) patients were inconsistent with intraoperative findings. The results of the paired *t*-test for the distance showed that the difference between the colostographic and intraoperative values had statistical significance ($P = 0.003$) and that the distal colostography technique also estimates the above distance as lower than the actual value calculated in surgery.

### *Cystoscopy findings*

Blind (primary) cystoscopy was performed in all patients. The results regarding the status of the fistula included: no fistula (70%) and recto-prostatic fistula (20%), rectobulbar fistula (10%), recto-membranous fistula (none), and rectobladder neck fistula (none).

In the second cystoscopy, performed in the 7 patients with no fistula in the primary cystoscopy, the results regarding the status of the fistula included: no fistula (28.57%), recto-prostatic fistula (28.57%), rectobulbar fistula (14.28%), recto-membranous fistula (none), and rectobladder neck fistula (28.57%).

### Comparison between cystoscopy and intraoperative findings

In determining the presence of a fistula, blind and second cystoscopy findings were 50% and 100% consistent with intraoperative findings, respectively. In patients who were diagnosed with a fistula by blind cystoscopy, the location of the fistula was 100% consistent with the intraoperative findings.

Regarding the type of fistula in patients who had undergone secondary cystoscopy and were proven to have fistula during the surgery (five patients), cystoscopic observations in one (20%) patient were inconsistent with intraoperative findings.

### *Preoperative examination findings*

Preoperative examinations were performed on all patients. The gluteal state was well-developed in 60%, poorly developed in 30%, and undeveloped in 10%. Perineal dimples were present in 60% of patients. The sacrococcygeal examination revealed a normal sacrum (20%), a short sacrum (30%), and no sacrum at all (50%). The condition of muscle assessed by a muscle stimulator showed full development in 80% and no development in 20% of patients.

### *Intraoperative findings*

Anorectoplasty surgery was performed in all patients. The results regarding the status of the fistula included: no fistula (20%) and recto-prostatic fistula (30%), rectobulbar fistula (20%), recto-membranous fistula (none), and rectobladder neck fistula (30%). The muscle complex was well-developed in nine participants (90%) and undeveloped in one participant (10%). The average poouc to h#tonperineum distance measured during surgery (the actual value of the distance) was 29 ± 13.29 mm.

Table 1 gives a summary of findings by different methods in each case.

## Discussion

ARMs can have significant effects and lifelong complications on the lives of patients, and proper diagnosis and treatment of these diseases have always been among the concerns of pediatric surgeons[13]. Considering the high burden of this disease, determining the best method for diagnosing and determining the

**Table 1**

**Summary of the 10 cases assessed in this study**

| Patient number | Age(d)/Sex | Perineal sonography | Distal colostogram | Initial cystoscopy | Second cystoscopy | Preoperative physical examination | Intraoperative findings |
|---|---|---|---|---|---|---|---|
| 1 | 96 Male | Well-developed muscles complex, a recto-membranous fistula and pouch to perineum distance: 20 mm. | Rectobladder neck fistula, pouch to perineum distance: 25 mm. | No fistula | Rectobladder neck fistula. | Well-developed gluteal muscle and muscles complex and perineal dimple with sacral agenesis. | Well-developed muscles complex, a rectobladder neck fistula, pouch to perineum distance: 30 mm. |
| 2 | 62 Male | Well-developed muscles complex, a rectobulbar fistula, pouch to perineum distance: 15 mm. | Rectobulbar fistula, pouch to perineum distance: 15 mm. | No fistula. | Rectobulbar. | Well-developed gluteal muscle and muscles complex, no perineal dimple and no sacroccygeal abnormality. | Well-developed muscle complex, rectobulbar fistula, pouch to perineum distance: 30 mm. |
| 3 | 96 Male | Well-developed muscles complex, a recto-prostatic fistula and pouch to perineum distance: 19 mm. | Recto-prostatic fistula, pouch to perineum distance: 20 mm. | Recto-prostatic fistula. | | Well-developed gluteal muscle and muscles complex, no perineal dimple, sacral agenesis. | Well-developed muscle complex, rectoprostathic fistula, pouch to perineum distance: 30 mm. |
| 4 | 105 Male | Well-developed muscles complex, a rectobladder neck fistula, pouch to perineum distance:30 mm. | Rectobladder neck fistula, pouch to perineum distance: 30 mm. | No fistula. | A rectobladder neck fistula. | Well-developed gluteal muscle and muscles complex, perineal dimple with shortened sacral bone. | Well-developed muscles complex, rectobladder neck fistula, pouch to perineum distance: 50 mm. |
| 5 | 104 Male | Well-developed muscles complex, a rectobulbar fistula, pouch to perineum distance: 9 mm. | Rectobulbar fistula, pouch to perineum distance:15 mm. | Recto-prostatic fistula. | | Well-developed gluteal muscle and muscles complex, perineal dimple with shortened sacral. bone | Well-developed muscles complex, recto-prostatic fistula, pouch to perineum distance: 25 mm. |
| 6 | 98 Male | Well-developed muscles complex, no rectourethral fistula, pouch to perineum distance: 3 mm. | No fistula from the rectum to the urinary system was found, pouch to perineum distance: 5 mm. | No fistula. | No fistula. | Well-developed gluteal muscle and muscle complex and perineal dimple and no sacral abnormalities. | Well-developed muscle complex , no rectourethral fistula, pouch to perineum distance: 5 mm. |
| 7 | 99 Male | Undeveloped muscles complex, a rectobulbar fistula, pouch to perineum distance: 27 mm. | Rectobulbar fistula, pouch to perineum distance: 35 mm. | Rectobulbar fistula. | | Undeveloped gluteal muscle and muscles complex, a perineal dimple, sacral agenesis. | Undeveloped muscles complex, a rectobulbar fistula, pouch to perineum distance: 40 mm. |
| 8 | 66 Male | Well-developed muscles complex, a rectobulbar fistula, pouch to perineum distance:15 mm. | Recto-membranous fistula, pouch to perineum distance: 25 mm. | No fistula | Rectourethral (recto-prostatic) fistula | Poor developed gluteal muscle and muscles complex, no perineal dimple, sacral agenesis. | Undeveloped muscles complex, a rectobladder neck fistula, pouch to perineum distance: 40 mm. |
| 9 | 86 Male | Well-developed muscles complex, no rectourethral fistula, pouch to perineum distance: 10 mm. | No fistula, pouch to perineum distance: 15 mm. | No fistula. | No fistula. | Well- developed gluteal muscle and muscle complex, perineal dimple, sacral agenesis. | Well-developed muscles complex, no fistula, pouch to perineum distance: 20 mm. |
| 10 | 104 Male | Well-developed muscles complex, a rectobulbar fistula, pouch to perineum distance: 20 mm. | Rectobulbar fistula, pouch to perineum distance: 25 mm. | No fistula. | Recto-prostatic fistula. | Poor developed gluteal muscle and muscles complex, no perineal dimple, shortened sacral bone. | Undeveloped muscles complex, a recto-prostatic neck fistula, pouch to perineum distance: 35 mm. |

location of the fistula can prevent the unnecessary use of avoidable methods and also facilitate the process of diagnosis and treatment.

In the current study, the diagnostic accuracy of sonography and distal colostography in determining the type and the location of the fistula is good and statistically significant; however, the data of these two imaging modalities regarding the distance between the pouch and skin are significantly different from the findings during the operation.

Cystoscopy was performed in the same session of anorectoplasty surgery and just before the operation.

During the cystoscopy, the pediatric surgeon had no information about the type and location of the fistula reported in the ultrasound and distal colostography results, and the cystoscopy was initially carried out blindly. With the statistical analysis of the data, it can be concluded that the diagnostic accuracy of cystoscopy is appropriate in determining the location of the fistula and that cystoscopy can help as a diagnostic modality.

In this study, although cystoscopy could accurately detect the presence of the fistula, it could not accurately locate the fistula in all patients. Furthermore, based on the results of this study, a second cystoscopy seems to be necessary and a single blind cystoscopy is not sufficient for the preoperative diagnosis of the fistula. However, both blind and second cystoscopy seem to have a higher sensitivity regarding locating the fistula compared with sonography and distal colostography. Therefore, when planning anorectoplasty for patients with anorectalmal formations, cystoscopy could be a helpful addition to sonography and colostography, as it can provide extra and relatively valid information about the presence and more specifically location of the fistula. Several studies have been conducted over time regarding the diagnosis and location of the fistula, which assessed the use and efficiency of transperineal ultrasound on patients with ARMs, and introduced sonography as an appropriate method compared with colostogram[14–16]. It has been discussed that rectourinary fistula associated with ARM can be well diagnosed in colostography with the oblique view[17]. The results of this study are in concordance with these studies, it additionally demonstrates cystoscopy as a suitable method to determine the location of the fistula, but whether it can be used to exclude other methods is not confirmed by this study. Further studies, best in the form of a nonexperimental cross-sectional study, are needed to confirm whether cystoscopy can be used as the best preoperative imaging tool for locating the fistula. The small number of patients was one of the limitations of the current study, and it is recommended to conduct other studies with a larger number of patients in the future.

## Conclusion

This study was conducted on 10 patients. All 10 patients underwent three diagnostic modalities, transperineal ultrasound, distal colostography, and cystoscopy and their results regarding the condition of the muscle complex, the presence of rectal fistula to the urinary system, and also the poouc to h#tonperineum distance was compared with intraoperative findings.

In statistical analysis, the diagnostic accuracy of ultrasound and distal colostography in determining the type and location of the fistula was appropriate, but sonography failed to be precise enough in determining the poouc to h#tonperineum distance.

The diagnostic accuracy of targeted cystoscopy was good in determining the type of the fistula, and regarding the location of the fistula both stages of cystoscopy proved to have more accuracy compared with other imaging modalities. Therefore, it is concluded that cystoscopy can also be used as a diagnostic modality but it is not confirmed that by doing so, the need for other modalities, especially distal colostography, is abolished. Different pediatric surgery centers around the world use different methods as a standard procedure to diagnose the presence and location of fistula in patients with anorectal anomalies. The standard procedure of our center was distal colostography. The results of this study show that a single procedure cannot have sufficient accuracy in determining the presence and location of a fistula, and it is better to make all necessary efforts to determine the exact location of the fistula before surgery to avoid irreparable damages caused by insufficient knowledge of the area's anatomy.

No personal data of patients has been revealed in this study.

## Ethical approval

This study was approved by the Ethics Committee with number 960867 and code IR.MUMS.MEDICAL.REC.1397.434.

## Consent

NA (No personal information of patients have been revealed in the study).

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## Conflicts of interest disclosure

The authors declare that they have no financial conflict of interest with regard to the content of this report.

## Provenance and peer review

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