

Augmented Pressure Distal Colostogram

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Summary:

Background: Anorectal malformations (ARMs) represent a complex group of congenital anomalies resulting from abnormal development of the hindgut, allantois and Mullerian duct, leading to incomplete or partial urorectal septal malformations.

Objectives: The objective of this study is the accurate demonstration of the anatomy of the level of rectal atresia and any associated recto-urinary fistula by using APDC for optimal surgical management using APDC.

Patients and methods: This is a prospective study including sixty-five male patients with high ARMs who were admitted to the department of pediatric surgery in Children Welfare Teaching Hospital in Medical City Complex - Baghdad - Iraq ; during the period from April 2009 to November 2011 ; they were subjected to divided descending colostomy in the postnatal period ;Augmented-Pressure Distal Colostogram (APDC) was performed at a later age for demonstration of the level of rectal atresia and the presence of the recto- urinary fistulas which then were confirmed at operation.

Results: Rectourinary fistulas were demonstrated in 52 patients (80%) by APDC technique and were confirmed at operation in each case. In the remaining 13 patients (20%), this technique failed to demonstrate a fistula; in 8 of them no fistula was identified during operation or thereafter, while in the remaining 5 patients fistulas were detected during operation.

Conclusion: The APDC readily demonstrated the level of the rectal atresia and the associated recto-urinary fistulas in the majority of the patients , providing a road map for definitive surgical repair of ARMs.

Keywords: augmented pressure distal colostogram (APDC), ano rectal malformations) ARMs).

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Introduction:

Anorectal malformations (ARM) represent a complex group of congenital anomalies resulting from abnormal development of the hindgut, allantois and Mullerian duct, leading to incomplete or partial urorectal septal malformations. The spectrum of lesions varies from fairly minor lesions (e.g. anal stenosis) to some of the most complicated urogenital lesions including anal agenesis, rectal agenesis and rectal atresia, as well as complex abnormalities, the level being determined by the relationship to the pelvic floor (1,2,3,4). An arrest of division of the cloaca may occur at any stage in the embryological development process leading to a wide spectrum of ARMs(16,17). The reported incidence of ARM is approximately 1 of every 4,000- 5,000 live births(3,5). ARMs occur more frequently in boys than girls, with a 56:44 male:female ratio was reported in large collective series.) 19).

The type of the Male ARMs : Recto-perineal (cutaneous) fistula: It is the simplest ARMs and is what traditionally was known as a "low defect"(3, 5) . Recto-urethral fistula: It is the most frequent defect in male patients. (3) . Recto-vesical (recto-bladder neck) fistula : About 10% of males with ARMs fall into this category(5). Imperforate anus without fistula The rectum usually terminates approximately 2 cm from the perineal skin.(3, 5,). Rectal atresia : The upper pouch is represented by a dilated rectum, whereas the lower portion is

represented by a small anal canal that is in the normal location and is 1 to 2 cm deep (19). Management of ARMs :The level of the ARM can determine whether to do a primary perineal operation, or to perform a colostomy and deferring definitive repair. In addition to presence of any associated anomalies, particularly in the urinary tract(3, 5).

Diagnostic Measure (Radiological Studies):An "invertogram" (upside-down inversion x-ray)(3, 5) ,The prone, cross-table lateral x-ray(19, 20,21) ,Micturating Cystourethrography(20, 21) , Augmented-Pressure Distal Colostogram) 11, 12) ,Other diagnostic measures (20,21).

Aim of Study: To assess the role of using augmented-pressure distal colostography) APDC) in patients with ARMs ; and its accuracy in demonstrating the anatomy of the level of rectal atresia ; and the presence of any associated recto- urinary fistulous communication essential for optimal surgical management.

Patients and Method;

Sixty-five male patients with high ano -rectal malformations (ARMs) who were admitted in the department of pediatric surgery in Children Welfare Teaching Hospital; and were included in this study . They were diagnosed by clinical findings and invertograms or cross-table lateral radiographs to classify their ARMs prior to colostomy, and then underwent a divided descending colostomy performed in the postnatal period.

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Augmented-Pressure Distal Colostogram (APDC) was performed at a later age for accurate demonstration of the anatomy of the level of rectal termination and the presence of recto-urinary fistula which was then confirmed at surgical repair) posterior sagittal anorectoplasty with or without abdominal approach).

Required materials are a suitable size Foley catheter with a 5 cc inflatable balloon, a 50-60 cc catheter tip syringe, and water soluble contrast. Following insertion of the Foley catheter into the distal colon segment stoma and inflation of the balloon, continuous gentle traction is applied to the catheter in order to obtain a seal at the stoma and prevent contrast leakage. Water- soluble contrast is then injected under gentle but increasing pressure until the distal rectal segment and associated fistula are identified. Fluoroscopy with spot filming in the lateral projection is performed while the distal rectum is fully distended , this permitted improved definition of the entire recto-urinary fistula . It is important to include the sacrum, perineum, and urinary bladder on the radiographs, and the anal dimple must be identified with a radiopaque marker. It is important to monitor the opacification of the distal colon under fluoroscopy and feel the increasing resistance as much as possible via the hand while performing this technique ,and caution must be used to avoid excessive pressure that might result in rupture of the colon. Great caution must be taken in cases with too distal colostomy (sigmoid) during APDC, as excessive pressure may rupture the colon. In contrast, in cases with too proximal colostomy (such as a right transverse) can result in inadequate pressure during the APDC and probably not show the fistula. So they're not preferred in ARMs . Statistical Analysis:Statistical Package for Social Sciences- version 18 (SPSS 18) was used for data analysis. Chi square test for goodness of fit used to test the significance of observed distributions. Findings with P value less than 0.05 considered significant.

Results:

The sample is constituted of 65 males with high type ARMs during the study period, mean age was 16.3±12.4 months (minimum 3 months and maximum 66 month) at performing APDC (table 1).

Table 1 : Age distribution of the study sample (at time of performing augmented pressure distal colostography).

Age Group	Frequency	Percent
< 1 year	39	60.0
1-5 years	25	38.5
> 5 years	1	1.5
Total	65	100.0

The proportion of recto-urinary fistulas among male patients with high type ARMs is 0.88 [0.77; 0.94] .

Rectourinary fistulas were demonstrated in 57 patients (tables 2 and 3). Ten recto-vesical ,13 recto-prostatic , and 34 recto- bulbar fistulas were found . In the remaining 8 patients no fistula was identified during operation or thereafter, three (37.5%) of them have Down syndrome, and other two (25%) had rectal atresia.

It is significant to find the majority of male patients born with high type ARM (imperforate anus) also have associated recto- urinary fistula, that about half of them (52%) have recto-bulbar fistula, one fifth (20%) have recto-prostatic fistula, about (15%) having recto-vesical fistula and about (12%) do not have associated recto-urinary fistula (P< 0.05) , [tables(2&3)] .

Table 2: Distribution of Recto-Urinary fistula Types in all patients in the study sample.

Type of Recto Urinary Fistulas	Frequency	Percent	X2	P
Recto-bulbar	34	52.3	26.631	0.000
Recto-prostatic	13	20.0		
Recto-vesical	10	15.4		
No Fistula	8	12.3		
Total	65	100.0		

Table (3) :Frequency of Recto-Urinary fistula Types (only in patients with Recto-Urinary fistulas in the study sample).

Types of Recto Urinary Fistula	Frequency	Percent	X2	P
Recto-bulbar	34	59.6	18.000	0.000
Recto-prostatic	13	22.8		
Recto-vesical	10	17.5		
Total	57	100.0		

Rectourinary fistulas were demonstrated in 52 patients by APDC technique and they were confirmed at operation in each case. In the remaining 13 patients, where this technique (APDC) failed to demonstrate a fistula, in 8 of them no fistula was identified during operation or thereafter , while in the remaining 5 patients fistula was detected during operation (table 4 .) .

Table(4) : Distribution of study sample according to the findings of both augmented pressure distal colostography and surgery.

Surgery Findings				Total
Positive		Negative		
APDC Findings	N %	N	%	N %
Positive	52 91.2	0	0.0	52 80.0
Negative	5 8.8	8	100.0	13 20.0
Total	57 100.0	8	100.0	65 100.0

Table(5): Preformance of APDC in screening for recto urinary fistula in male patients with high type ARMs

Indicators	Level % [95% CI]
Sensitivity	91 [80; 97]
Specificity	100 [60; 99]
Accuracy	92 [82; 97]
Positive Predictive value	100 [91; 100]
Negative Predictive value	62 [32; 85]

Augmented pressure Distal Colostography can detect 91% of cases (sensitivity), and can exclude all those who do not have recto urinary fistula (specificity), correct results (in diagnosing or excluding cases) are 92% of results (accuracy), All positive results are true results (positive predictive value), negative results are correct by 62%, this means that some cases can be missed (table 5).

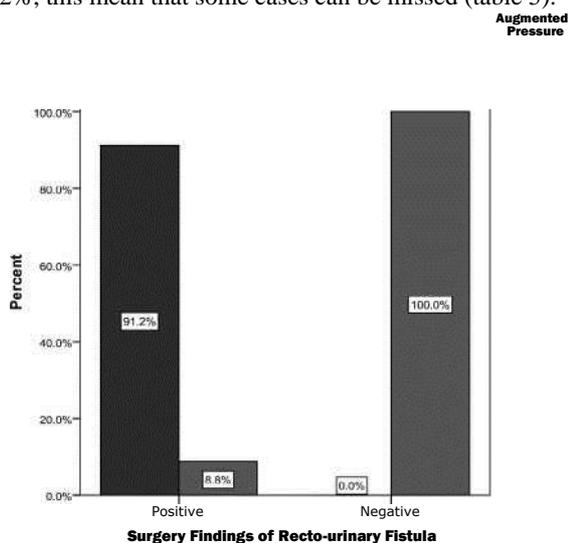


Figure (1) : Distribution of study sample according to the findings of both augmented pressure distal colostography and surgery

Discussion:

Anorectal malformations (ARMs) represent a complex group of congenital anomalies with an incidence of approximately (1 in every 4,000-5,000 live births). Males are affected slightly more frequently than females. (6,7) ARMs can be diagnosed clinically, but their associated recto-urinary fistulae and classification require radiological imaging. Various imaging methods and modalities are available with specific limitations and advantages. APDC, which is described by Gross et al. that readily delineates a recto-urinary fistula that was not identified by conventional distal colostography. (5,8) For high ARMs, a diverting colostomy is performed initially, with the subsequent performance of a pullthrough procedure (PSARP). In all patients undergoing a PSARP; as the main goal of surgical treatment of children with ARMs is to achieve as much fecal and urinary continence as possible. Therefore; it is important for surgeons to know the precise location of the rectal atresia and any associated recto-urinary fistula. As inappropriate surgery may lead to damage of the pelvic structures (i.e. incontinence), persistence of fistula, and damage to the urinary tract. Imperforate anus without fistula is reported to occur in approximately 5% of all cases of ARMs. However, it was suspected that the actual incidence of it is even lower than reported due to improperly performed colostograms. (9) Anatomic considerations upon which the rationale behind the APDC is based on that the distal most rectal segment and the associated recto-urinary fistulas pass through a funnel-like muscular structure which is usually contracted, resulting in compression of their lumen. Gravity infusion or hand injection of water-soluble contrast material via a straight catheter (without sufficient hydrostatic pressure) may fail to fully distend and define the distal most rectal segment and any associated recto-urinary fistula, as illustrated by our patients. In order to overcome resting pelvic floor muscle tone, it may be necessary to exert gentle but increasing hydrostatic pressure. Fluoroscopic observation during APDC will permit dynamic visualization of muscular contraction and relaxation. (22)

In our study the results demonstrate that the use of augmented pressure distal colostography (APDC) is the most accurate study available to define the level of rectal termination and the presence and the extent of any associated recto-urinary fistula, and it is a relatively simple, easily performed, safe and invaluable technique. So it is significant to find the majority of male patients born with high type ARMs also have associated recto-urinary fistula, that about half of them (52%) have recto-bulbar fistula, one fifth (20%) have recto-prostatic fistula, about (15%) having recto-vesical fistula, and about (12%) do not have associated recto-urinary fistula. Our P-value test was (< 0.05). The APDC technique has a (91% sensitivity), (100% specificity), (92% accuracy), (100% positive predictive value), (62% negative predictive value). In the review of literatures and in our study

results we found that a high percentage of male infants with high-type ARMs have a fistula between the rectum and the urinary tract. The most frequent defect in male ARMs is a recto- urethral fistula. These can be demonstrated by APDC which is the most informative available technique, and therefore APDC should be the first investigation performed to demonstrate the level of rectal termination and the presence and the extent of any associated recto-urinary fistula prior to definitive surgery. The identification of a recto-bulbar, recto-prostatic, or rectovesical fistula is of paramount importance; because a recto- bulbar fistula may have a better prognosis than a recto-prostatic fistula, and a very high ARMs who require a laparotomy to complete the repair, which have a much poorer prognosis than the rest(14). Our findings are similar to Abdulkadir A Y. et al. (2009)(22) although they used a non-fluoroscopic APDC (in a resource-poor setting where fluoroscopic equipment is limited) ; they found that the APDC invaluable technique resulted in better demonstration of recto-urinary fistulae compared with the conventional distal colostography . Rintala R J. et al. (2009X19), Gupta AK. et al. (2005) (20), Pena A. et al. (2000) (21), Niedzielski J. et al. (1998) (23), Wang C. et al. (1997) (14), and Gross GW. et al. (1991) (12) ,all of them found that the fluoroscopic conventional distal colostography performed under augmented pressure with the colostomy stoma occluded by an inflated Foley balloon increases the accuracy of this study , and APDC is a simple , easily performed, and accurate study to define the level of rectal termination and any associated recto-urinary fistula . They recommended to use APDC in any patient undergoing colostography prior to definitive surgical repair of ARMs. While Horsirimanont S. et al. (2004) (24) .although they used the conventional distal colostography (without pressure augmentation) in their study; they found that technique has a relatively high specificity, and a sensitivity of around (60%); and agreed with Gross et al. (12), our study , and the other similar studies ; who suggested an augmented- pressure distal colostogram to improve the test sensitivity

Conclusion:

In our study we found that the use of augmented pressure distal colostography (APDC) is a relatively simple, easily performed, safe and accurate technique for defining the level of rectal termination and any associated recto-urinary fistula ;and it provides a road map for definitive surgery . While gravity infusion or hand injection of water-soluble contrast material via a straight catheter may not accurately define the level of rectal termination or any associated fistula, as illustrated by our patients. No complication occurred while using this technique.

In addition, This simple modification of colostography in children has received scant mention in the radiological literature, probably due to its infrequency of use and the unfamiliarity of many radiologists with the importance of

Defining any associated recto-urinary fistula in ARMs.

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