An Appraisal of Invertograms and Distal Colostograms in the Management of Anorectal Malformations

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Objectives : *To determine the accuracy of two main radiologic tools currently employed in the clinical management of anorectal malformations, the invertogram and the distal colostogram. The data will be useful as a basis for quality assurance as well as for future comparisons in case there are innovations to be considered.*

Material and Method : *Radiological materials and clinical records of infants with anorectal malformations operated on in Songklanagarind Hospital from 1995 to 2001 were retrieved. Each record was blindly reviewed by two radiologists and one pediatric surgeon. Using operative findings as the gold standard, the accuracy of an invertogram in diagnosing low anomalies and the accuracy of a distal colostogram in screening cases that needed a laparotomy and in detecting internal fistulas were determined. The quality of the films was also categorized and poor quality studies were excluded from the analysis. Cases of common cloaca were not included in the level reading analysis.*

Results : *Radiologic materials from 59 patients were examined, including 26 invertograms and 49 distal colostograms. Among 52 cases whose neonatal history could be traced, 29 (55.8%) cases exhibited clinical evidence of anomalies level. Two invertograms and 5 colostograms were graded as poor quality and excluded from the analysis. The overall sensitivity of invertograms in detecting low anomalies was 33.3%, whereas, specificity was 66.7%. Analyzing only infants with a blind rectal pouch, the sensitivity and specificity to detect low anomalies were 33.3% and 75.0%, respectively.*

The sensitivity of distal colostograms in detecting a fistula was 60.0% in males and 62.5% in females. Distal colostograms diagnosed 'high-type anomalies' in 7 cases and correctly detected 2 cases of vesical fistula which needed APSARP. No prostetic-urethral fistulas showed a radiological high level.

Conclusion : *The study found poor diagnostic sensitivity of invertograms in detecting low type anomalies which deserved primary anoplasty. However, the data support the role of a distal colostogram in diagnosing high anomalies, despite its low sensitivity in detecting urethral fistulas.*

Keywords : *Anorectal malformations, Invertogram, Colostogram*

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Surgical management of an infant with anorectal malformations is directed according to the individual level of anomaly as well as the presence or absence of a fistula between the rectal pouch and adjacent organs[1,2]. Newborn anoplasty is the surgical method of choice for a case whose rectal pouch terminates close to the perineal skin, whereas temporary colostomy is usually chosen for cases with a high-lying rectum and/or internal fistula. Although the level of anomaly in the majority of cases can be determined by a careful clinical examination, radiologic investigations are necessary in 10-20% of patients when clinical data is not sufficient[3].

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The Invertogram is an x-ray technique first suggested by Wangensteen and Rice for an estimation of the rectal pouch level[4]. Although the technique has long been employed, it is still an investigation of choice for neonatal evaluation[1,2]. In infants for whom a colostomy was initially established, a distal colostogram is routinely prescribed to demonstrate the rectal pouch more precisely[1]. The main purpose of a colostogram is screening for cases that posses a higher chance of requiring a laparotomy in addition to a posterior sagittal approach during reconstructive surgery. Any fistula between a rectal pouch and the genitourinary organs can also be detected by a colostogram. However, despite their widespread use, little objective data regarding the accuracy of these two classic tools has been published.

Modern imaging modalities have recently been introduced for pre-operative evaluation. Computerized tomography (CT) and magnetic resonance imaging (MRI) are acclaimed for their ability to display images of the pelvic structure in detail[5-7]. To prepare for the implementation of those technologies, the authors performed a critical appraisal of classic investigations, in order to get an objective insight into current limits and advantages.

The main objectives of this study were to determine the accuracy of invertograms and distal colostograms, and to analyze their roles as imaging modalities in the surgical management of anorectal malformations.

Material and Method

Invertograms and/or distal colostograms (loopograms) of pediatric patients with anorectal malformations treated in the Pediatric Surgery Unit, Songklanagarind Hospital from 1995 to 2001 were reviewed for their quality and diagnostic capabilities. Medical records of the patients were also retrieved to extract data regarding their anomalies, operations and demographic data. The clinical materials review was approved by the Research Ethics Committee of the Faculty of Medicine, Prince of Songkla University (2002).

Radiologic techniques

An invertogram was usually prescribed for a case that was born in this institute or was referred to the authors during the neonatal period. It was often unnecessarily prescribed. Radiographs were taken in the neonatal intensive care unit by a portable x-ray machine. A 'prone cross-lateral view' modification as described by Narasimharao was employed[8]. The infant would be placed in a prone position with the hip flexed and elevated up to 45 degrees. The radiographic center was placed around the greater trochanter. A radiologic marker was routinely placed at the perineal area where there should be anal dimpling. Because of the retrospective nature of the present study, the timing of the investigation was not protocolized and varied from hours to days after birth.

A distal colostogram was performed prior to a definitive anorectal reconstruction in infants for whom colostomy had been established. A water-soluble contrast media was injected into a distal limb of a sigmoid loop colostomy via a plain catheter. The level of anomalies and possible presence of a fistula was observed under fluoroscopy in a true lateral position.

Definitions and criteria

All films were categorized for their quality in terms of diagnostic capability. Quality was graded as 'good' - a film with appropriate position and exposure which could be read clearly, 'fair' - for a film with some technical problems but which still could be read, and 'poor' - for a film in which the technical distortion was too severe to get any diagnostic information.

The criteria for reading low type anomalies by invertogram was that the most distal air shadow in the rectal pouch was distal to an M-line, as suggested by Cremin[9,10]. The distance between the anal marker and rectal gas was used as an adjunctive criteria. Considering the distal colostogram, high type anomalies were diagnosed when a rectal pouch terminated above a PC-line. Visualization of contrast passing from the distal end of the rectum to the bladder or urethra in males, and the vagina or the vestibule in females, led to a diagnosis of an internal fistula.

Perineal fistulae in both sexes, and male scrotal fistulae, were regarded as external fistulae and not included in fistula reading analysis by distal colostogram. Both of those anomalies, and all female vestibular fistulae, were also regarded as low types in the invertogram analysis. For the invertogram analysis, a blind pouch rectum was defined as low when the rectal pouch terminated within 2 centimeters of a skin level at the perineal body, and could be brought and anastomosed with the skin without excessive tension. For distal colostogram analysis, an anomaly was regarded as high when it needed a

laparotomy to assist mobilization (abdomino-posterior sagittal anorectoplasty, APSARP). Cases of common cloaca and one case of a male infant with membranous-urethral fistula were excluded from level reading.

Two pediatric radiologists and one pediatric surgeon individually reviewed all films without prior knowledge of patient names or operative findings. In case of disagreement, two out of three opinions determined the final result. Poor quality films, as suggested by at least one of the readers, were excluded from all analyses.

Results

Radiologic materials from 59 patients were examined, including 26 invertograms and 49 distal colostograms. Data regarding the types of anomaly are shown in Table 1. One male infant had a low-lying rectal pouch with a recto-membranous-urethral fistula. Among 52 cases whose neonatal history could be traced, 29 (55.8%) cases exhibited clinical evidence of anomalies. Twelve of 21(57.1%) male infants with a fistula between their rectal pouch and urinary tract showed meconium in the urine and/or urethral meatus. There were 23 cases who presented no clinical determination of anomalies; 19(82.6%) had a blind rectal pouch and 4 had a recto-balbar urethral fistula. All four female cases that presented no clinical evidence of fistula had Down's syndrome and a low-lying blind rectal pouch.

Invertogram

Two of twenty-six invertograms were excluded from analysis owing to their poor quality, and thirteen (50%) were graded as fair. The films were taken within 16 hours of birth in 5 cases, between the16th and the 24th hour in 3 cases and after the 24th hour in 18 cases. Sensitivity to detect low anomalies was 33.3%, and specificity was 66.7%. The positive predictive value was 0.5, and overall accuracy 50.0%. Sensitivity rates of studies obtained before the 16th hour of birth and after were equally 33.3%. However, time-related specificities were 100% and 55.6%, respectively.

Of the 10 infants with blind rectum whose invertograms were available, the sensitivity to detect low anomalies was 33.3%, with 75.0% specificity, and the positive predictive value was 0.67. Sensitivity and specificity to detect low anomalies in cases with fistula were 33.3% and 62.5%, respectively. No cases presented air shadow in their urinary tract.

Distal colostogram

Five studies were too poor to read. Of the 44 studies, 14 cases were categorized as fair quality. The most common cause of technical error was poor positioning. Among the "fair" was included a case who had some anatomical distortion of the spine secondary to sacral dysgenesis. The test detected a connection in 12 of 20 male infants with a fistula between the rectum and urinary passage (60.0%

Table 1. Types of anomaly and the availability of radiologic materials studied

Type of anomaly	No. (cases) (%)	No. of studies available	
		Invertogram	Colostogram
Male (N = 43 cases, 72.9%)			
- perineal or scrotal fistula	5 (8.5)	4	1
- blind pouch rectum, low	9 (15.3)	5 (poor** = 1)	5
- blind pouch rectum, intermediate*	7 (11.9)	4	8 (poor = 2)
- membranous urethral fistula	1 (1.7)	-	1
- bulbar urethral fistula	17 (28.8)	6	17 (poor = 2)
- prostetic urethral fistula	2 (3.4)	1	2
- vesical fistula	2 (3.4)	-	2
Female (N = 16 cases, 27.1%)			
- perineal fistula	1 (1.7)	1	
- blind pouch rectum, low	4 (6.8)	1	4
- vestibular fistula	3 (5.1)	2	2
- vaginal fistula	6 (10.2)	2 (poor = 1)	5 (poor = 1)
- common cloaca	2 (3.4)	-	2

* including cases that did not meet the low level criteria (see text)
** poor quality studies that were excluded from analysis

sensitivity). In 8 female patients with a vaginal fistula, vestibular fistula, or common cloaca, 5 studies detected the fistulas (62.5% sensitivity).

The distal colostograms read a high rectal pouch in 7 cases. The tests correctly detected 2 cases of vesical fistula who needed APSARP. However, none of the prostetic-urethral fistulae showed a radiological high level. None of our recto-prostetic-urethral fistulae required APSARP.

Discussion

Planning for surgical management of a neonate with anorectal malformations requires a determination of the anomaly level as well as the presence or absence of a fistula[1,2]. Most infants with low level anomalies undergo a definitive anoplasty within a few days of birth. Higher anomalies have poor sphincteric development and have more chance of an associated internal fistula. Reconstruction needs more extensive surgery, and is usually deferred until the infant gets older. Temporary colostomy is essential, and the colostomy itself helps investigate the anomaly in more detail by a contrast study called a 'distal colostogram'[2].

The invertogram has long been a routine prescription for an infant with certain conditions in our hospital, sometimes despite inadequate knowledge of its true indications and accuracy. Definitive anorectal reconstruction in a case diagnosed as a non-low type is usually performed when the infant weighed 4 kg or more. A distal colostogram is requested in every case prior to an operation, with its main purpose to demonstrate the level of the rectal pouch and detecting any internal fistula.

Only 55% of the presented cases (73% in females and 49% in males) presented clinical findings suggestive of anomalies. This figure is much lower than that found by Pena et al[3]. Although there was more incidence of blind pouch rectum in the present series compared to theirs, it is interesting that not more than 60% of male neonates with a urinary tract fistula presented meconium in the urine. In cases where there appears to be no perineal fistula in a male patient, clear urine hence does not exclude an internal urinary fistula. The authors' previous thirteen-year review of associated anomalies in anorectal malformations reported 7% Down's syndrome[11], all of them with blind rectal pouch. This same syndrome-type association was also reported in around 95% of cases in a larger series[12,13]. Although it is not pathognomonic, Down's syndrome can be regarded as one type-specific clinical clue.

The sensitivity of an invertogram to detect low type anomalies is quite unimpressive. The detection rate was not associated with x-ray timing or type of anomaly. However, specificity seemed to be reduced with lag time from birth to x-ray. Higher specificity of invertograms was found in cases of blind rectum compared to overall analysis. Escape of air from the rectal pouch through a fistula[9], or the discordance between radiologic and surgical meaning of 'low type' may explain these differences. The data suggested that the invertogram technique currently employed in the authors' institute does not well serve its diagnostic purpose. However, neglecting this investigation would mean that all infants with questionable clinical evidence should have a colostomy.

Considering the technique currently employed in our hospital, this prone cross-lateral modification is proven for its validity, compared to the classic position suggested by Wangensteen and Rice[8].The technical modification should not contribute to the inaccuracy. However, the evidence that about a half of the presented invertograms was graded as fair should be translated that there must be some room to improve the film quality, hence the diagnostic accuracy. In addition, a modification of reading criteria may enhance the accuracy of level reading.

The radiologic criteria used in the present study have been developed based on a classic concept concerning a key sphincter muscle, the levator ani. An M-point derived from an anatomic study by cine radiography delineated the functional lower border of this muscle[9,14] , hence the division between the supralevator and translevator lesions. The radiologic approach went along well with the old surgical concept that all supralevator lesions should be operated on using an abdominal approach. After deVries and Pena launched their concept of striated muscle complexes and its application to their well-known posterior sagittal anorectoplasty, the modern surgical algorithm became altered[15,16]. Nevertheless, significant change did not carry over to the radiologic side following that alteration.

Categorization of levels in the present study followed the authors' current surgical practice. Lesions were regarded as low when they required a primary anoplasty and high when they needed a laparotomy. Blind rectal pouch anomalies were classified as low when they lay close enough to be

brought to the perineal skin without extensive mobilization or undue anastomotic tension. The vestibular fistula is another lesion for which surgical practice varies among institutes. Many surgeons still divide a vestibular fistula into rectovestibular and anovestibular by clinical examination, and prefer to do primary anoplasty for the latter only. However, as the authors performed a newborn anoplasty for nearly all vestibular fistula, the lesion was regarded as a low type in the level analysis.

By PC-line criteria, sensitivity to read a high type by distal colostogram was high. However, around 80% specificity to detect a case that needs an abdominal approach should not adequate to direct all cases to a laparotomy. The posterior sagittal approach is always attempted first, even if the test reports a high type. In cases of recto-prostetic urethral fistula, preparation for possible laparotomy can be suggested by a distal loopogram. In our experience, a laparotomy is mandatory in cases with a recto-vesical fistula.

The invertogram seemed to have no role in fistula reading. The authors' evidence and that of previous large series studies showing that a low-lying rectum in males does not exclude associated urinary fistula[2,12]. For these reasons, any investigations that are not able to rule out a fistula, including an invertogram, should not be applied for a newborn investigation unless the surgeon believes that primary anoplasty together with fistula repair can be done safely in the neonatal period.

Despite its relatively high specificity, a distal colostogram had a sensitivity to detect a fistula of around 60%. This figure suggests that care should be taken to examine the most distal portion of the rectal pouch for a fistula, even though a colostogram reports negative. A recurring fistula needs re-operation, which may cause unnecessary damage to the sphincter muscles. Gross et al suggested an augmented-pressure colostogram under a balloon catheter to improve its sensitivity[17].

In summary, the authors conducted a retrospective review of two classical radiologic tools in anorectal malformations. The study found poor diagnostic sensitivity of an invertogram in detecting low type anomalies. Data support the role of distal colostograms in screening for high anomalies, despite their low sensitivity in detecting urethral fistulae. The authors hope that the data emphasizes the problems and will lead to more extensive and well-designed research, to find a suitable radiologic approach for this new era in assessing anorectal malformations.

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การทบทวนความเที่ยงตรงของ invertogram และ distal colostogram ในทารกซึ่งมีความผิดรูปโดยกำเนิดของทวารหนักและเรคตั้ม

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ได้ศึกษาความเที่ยงตรงของ invertogram และ distal colostogram ในฐานะเป็นเครื่องมือทางรังสีวิทยาที่สำคัญในทารก ซึ่งมีความผิดรูปโดยกำเนิดของทวารหนักและเรคตั้ม โดยรวบรวมภาพรังสีของทารกซึ่งเข้ารับการผ่าตัดในโรงพยาบาลสงขลานครินทร์ตั้งแต่พุทธศักราช 2538 กระทั่ง 2544 เพื่ออ่านและวิเคราะห์เทียบกับลักษณะความผิดรูปที่พบขณะผ่าตัด เน้นศึกษาในแง่การอ่านระดับความผิดรูป และการตรวจพบช่องเชื่อมระหว่างเรคตั้มและท่อโพรงของอวัยวะใกล้เคียง

ผลการรวบรวมได้ภาพรังสีจากผู้ป่วยทั้งสิ้น 59 ราย ประกอบด้วย invertogram 26 การศึกษาและ distal colostogram 49 การศึกษา ในจำนวนนี้ 2 และ 5 การศึกษาจากแต่ละกลุ่มตามลำดับได้คัดออกจากการวิเคราะห์เนื่องจากภาพรังสีมีคุณภาพต่ำ ผลการวิเคราะห์พบว่าโดยภาพรวม invertogram มีความไวและความจำเพาะร้อยละ 33.3 และ 66.7 ตามลำดับ หากเมื่อแยกวิเคราะห์เฉพาะความผิดรูประดับต่ำซึ่งเรคตั้มเป็นปลายตัน พบความไวและความจำเพาะร้อยละ 33.3 และ 75.0 ในส่วนของ colostogram ความไวในการตรวจพบช่องเชื่อมในเพศชายมีค่าร้อยละ 60.0 ในเพศหญิงมีค่าร้อยละ 62.5 การศึกษาสามารถอ่านระดับและการมีช่องเชื่อมได้อย่างถูกต้องในผู้ป่วยซึ่งมีความผิดรูปชนิด rectovesical fistula ทั้งสองราย การศึกษาสรุปผลว่า invertogram มีความไวต่ำในการวินิจฉัยแยกผู้ป่วยซึ่งมีความผิดรูประดับต่ำ ซึ่งสามารถให้การรักษาโดยผ่าตัดเบิกทวารขั้นสำเร็จตั้งแต่ระยะแรกคลอด หากจะพิจารณาใช้การตรวจนี้ต่อไป จำเป็นต้องปรับปรุงเทคนิค นอกจากนี้ยังสรุปว่า แม้ colostogram จะมีความไวในการตรวจหาช่องเชื่อมต่ำ แต่ก็มีความไวที่ดีในการตรวจกรองรายที่มีความผิดรูประดับสูง ซึ่งการแก้ไขความผิดรูปมีโอกาสที่จะต้องผ่าตัดผ่านทางช่องท้องร่วมด้วย