

Intraoperative Sonographic Guided Pull-through Anorectoplasty: A Novel Procedure for Imperforate Anus and Rectourethral Fistula: A Clinical Trial Study

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Abstract

Background: Optimal surgical management of the neonate with imperforate anus (IA) depends on determining accurate location of sphincter muscle complex, pouch of rectum and rectourinary fistula. We aimed to investigate a novel minimally invasive technique of anorectoplasty assisted by intraoperative sonography pull-through for repair of anorectal malformation and rectourinary fistula.

Materials and Methods: Eight male patients with anorectal malformation and IA underwent formation of a diverting colostomy within 48 hours after birth. These patients had anorectoplasty about 6-8 weeks postoperatively. A urinary catheter was inserted per urethra into the bladder and sonography of perineum was carried out under general anesthesia. The neoanus was reconstructed by suturing the pulled-through anorectum to the anal sphincter muscle complex and the skin using absorbable 4/0 sutures.

Results: In all cases, the distance of pouch of rectum to the skin was 15-18 mm and entrance of the guide wire to fistula was 6-7 mm. Of eight patients 6 had removal of urinary catheter after two days, and 2 patients after 10 days because the fistula was not closed. The median range of hospital stay was 2.12 days and follow-up was 30 days. We calibrate all of patients with size 12F dilators. Patients were followed up at one week and one month postoperatively, and all had defecation frequency of 3-5 times a day. There were no complications.

Conclusion

Intra-operative sonography guided pulled-through anorectoplasty (ISPA) is a novel and safe technique for surgical treatment of IA and rectourethral fistula. ISPA is a minimally invasive approach, which preserves the external anal sphincter muscle complex with good functional outcome.

Key Words: Anorectal malformations, Children, Clinical Trial Study, Sonography.

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1- INTRODUCTION

Anorectal malformation (ARM) is a common congenital disease with an incidence of 1 per 3,000 to 5,000 live births (1). The disease is more prevalent in males with a male to female ratio of 1.2 to 2:1 in the worldwide. There are associated congenital abnormalities including renal, cardiac and esophageal anomalies (2, 3). Traditionally the lesions are divided to high, intermediate and low anomalies. Various surgical procedures are applied to treat the anorectal malformation ranging from the posterior sagittal anorectoplasty (PSARP) to more advanced methods like the laparoscopic-assisted anorectoplasty (LAARP). In PSARP dissection of perineum is done through a sagittal plane with minimal damage to local blood and nerve supply as these structures do not cross the midline (4).

However, this approach requires division of the external anal sphincter muscle complex, and extensive dissection of pelvic floor muscles, and mobilization of anorectum, which may cause long term sphincter damage, neuropathy and ischemia. On the other hand, the limitation of the LAARP is that the vertical muscle fibers between the pelvic floor and perineal parasagittal muscle fibers cannot be visualized and the rectal prolapse is common postoperative (5, 6.; furthermore the dissection of anal sphincter complex may lead to fibrosis and injury to the muscle fibers causing stenosis or fecal incontinence later in life (7).

The aim of this study was to investigate a novel minimally invasive technique of anorectoplasty assisted by intraoperative sonography pull-through for repair of anorectal malformation and rectourethral fistula. We consider that it's possible to perform pull-through of the rectum in the neonatal period without colostomy construction

2- MATERIALS AND METHODS

In this randomized non-blinded clinical trial (TCTR.20170813001), neonates born with anorectal malformation and imperforate anus were included in the study from October 2015 to August 2016. They underwent formation of divided sigmoid colostomy within 48 hours of life. Patients had ultrasound scan of kidneys, renal tracts and spine to exclude associated anomalies and had posterior sagittal anorectoplasty, 6-8 weeks later. None of the patients had undergone distal colostogram. The reason that we ignore distal colostogram in our approach is the harmful effects of radiation exposure in neonate population. We can identify the distal rectal pouch by placing and traction of the Foley catheter with sonography. The patient was placed in lithotomy position and a Foley catheter was inserted into the bladder. Another catheter was passed through the distal limb of colostomy into the rectum and the balloon was inflated. After skin preparation and draping, normal saline was injected into the rectum via this catheter. Perineal ultrasound examination was done to identify the rectal pouch, the rectourethral fistula, and its distance from the skin, and the anal sphincter muscle complex (**Figure.1**).

It is possible to determine anterior and posterior of the urethra with sonography in this approach. A guide-wire was passed through the center of muscle complex, identified by electric stimulator, into the rectal pouch under sonography guidance (**Figure.2**). Then dilators size 3 to 11 F was passed over the guide-wire and dissection of perineum was carried on this plane to the rectal pouch. The rectum was grasped and stay sutures applied to retract it to the skin surface (**Figure.3**). Perirectal dissection was performed to identify the rectourethral fistula aided by pre-operative sonography findings. The fistula was cut and if necessary ligated with 4/0 absorbable sutures and the rectum pulled through to complete reconstruction of

neoanus within the external sphincter muscle complex. All patients had examination of anus under local anesthesia using 2% Lidocaine gel, and drinking Saccharose 26%, one week and one-month after the anorectoplasty. They also had sonography of perineum one month postoperatively. All patients were followed after the procedure. All the procedures

were conducted by the resident of pediatric surgery, a sonographer and a pediatric surgeon. Data presented as median and range. Data were analyzed using SPSS for windows (version 21.0, SPSS Inc., Chicago, IL. USA). Frequencies, percentage, mean and standard deviation (SD) were determined.



Fig.1: Insertion of a needle and guidewire through the center of external anal sphincter muscle complex under sonography guidance. Normal saline is injected into the rectal pouch via a Foley catheter inserted in distal limb of colostomy.

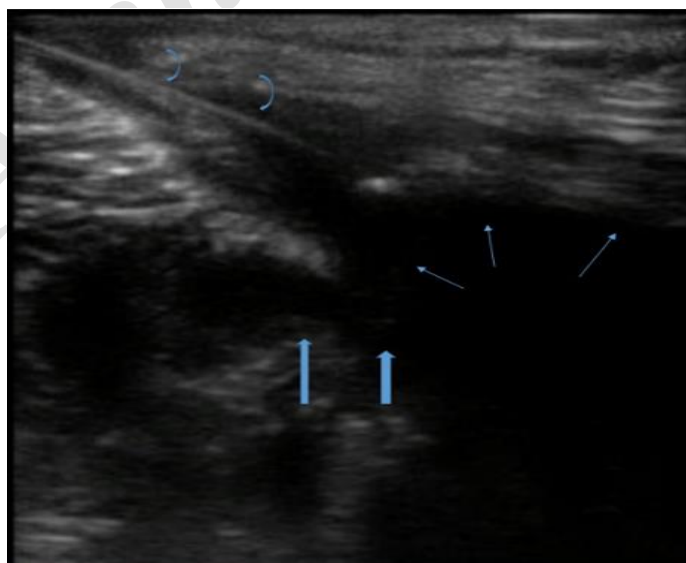


Fig.2: Sagittal view of perineal area. Thin arrows demonstrate rectal pouch and thick arrows shows rectourethral fistula tract and curved arrows delineate needle in center of hypoechoic anal sphincter muscle complex.

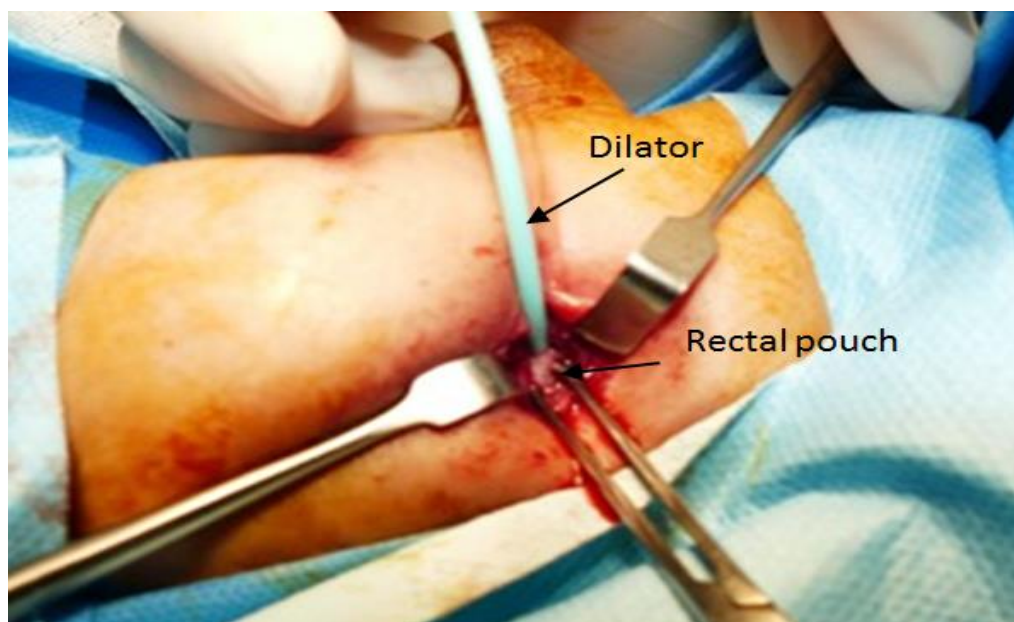


Fig.3: Minimal dissection of perineum along the dilator path preserving the external anal sphincter muscle complex.

3- RESULTS

Eight patients were enrolled in this study. The median age of patients at the time of anorectoplasty was 6.75 ± 0.127 weeks (6-8 weeks). All patients were male and their median weight was 5.75 kg (5-6 kg). Four of eight patients had other associated anomalies shown in **Table.1**.

In all cases, the distance from the rectal pouch to the skin was between 11 and 22 mm and the distance of entrance of wire into the fistula was 6-7 mm except in one patient (n= 8) who had Down's syndrome and no urinary fistula.

All patients started feeding at a median of 2.5 days (2-3 days). Demography and intraoperative sonography findings of patients in this series are summarized in **Table.1**.

First patient had removal of the urinary catheter 2 weeks after posterior sagittal anorectoplasty. He had closure of colostomy one month post-operatively, and was discharged home after three days. The second and third patients had removal of their urinary catheters 10 days following formation of neoanus and closure of colostomy one week later.

All remaining patients (sixth, seventh and eighth) had ligation of rectourethral fistula, removal of urinary catheter was removed on day two postoperatively. They were discharged home after 2.125 days (2-3). At one month following anoplasty, all patients had stool frequency of 3-5 times daily and their anus was calibrated to size 12 F Hegar. There were no complications in this study.

Table-1: Demography and transperineal sonography findings of neonates with anorectal malformation and imperforate anus

Patients	Gestational age (week)	Age of anoplasty (week)	Weight (kg)	Associated anomalies	Distance of rectal pouch to skin (mm)	Distance of wire entrance from rectal pouch to fistula	Age of colostomy closure (week)	Hospital stay
1	37	6	6	Urinary reflux	11	6	10	3
2	38	7	6	Single-kidney, ASD	22	6	11	2
3	38	8	6	No	20	7	12	2
4	37	6	5.5	No	21	6	10	2
5	39	7	6	Hydronephrosis, ASD	22	6	11	2
6	37	6	5.5	No	19	6	10	2
7	37	6	6	No	21	6	10	2
8	36	8	5	Down Syndrome, Cardiac and renal problems.	20	6	12	2

ASD: Atrial septal defect.

4- DISCUSSION

The accuracy of ultrasound scan (US) to determine the location of the rectal pouch in patients with anorectal malformation and imperforate anus has been reported in the literatures. The distance between perineal skin and rectal pouch can be determined in coronal and sagittal views by sonography (8, 9). However, application of this method has not been widely adopted. In this study we applied intra-operative sonographic guidance to determine precise location of the external anal sphincter muscle complex, pouch of rectum and the rectourethral fistula. Some noticeable advantages of the US include ability to image the end of rectal pouch and visualize the rectourethral fistula to measure their distances from the perineal skin. In addition, transperineal US is non-invasive and cheaper compared to MRI and CT scan, which often require general anesthesia (6, 7). Our study is limited by a small number of patients and short term follow up. Schuster and Teele evaluated function of US as a guide to determine the type of anorectal malformation and they

reported no associated pain or discomfort with this technique (9). Alehossein et al. reported transperineal US as a useful tool in deciding surgical approach in infants with imperforate anus. Initial evaluation of the level of rectal pouch reduced operating time and complications related to extensive perineal dissection during surgery (8). These include avoiding accidental damage to the urethra, bladder neck, ectopic ureter and misplacement of neoanus outside the sphincter muscle complex (10, 11). George et al. reported MRI guide laparoscopic-assisted anorectoplasty (LAARP), may be helpful in anatomy correct placement of the rectum within the vertical muscle complex (12). MRI isn't valuable technique in operation room in added that is more expensive than sonography. On the other hand, our presented approach may be more compatible with Iranian culture; people in Iran are sensitive to perineal region as like as many countries. This manner means that they don't prefer any scar in this region. The presented procedure seems more acceptable on this manner for parents. We have no claim about damages or

disadvantages of other anorectoplasty approaches that presented.

5- CONCLUSION

We have shown in this study that intraoperative sonographic guided pull-through anorectoplasty (ISPA) is a novel and minimally invasive technique. ISPA obviates the need for extensive perineal dissection and preserves the external anal sphincter muscle complex and blood supply and innervation of the anorectum. This technique is feasible and safe and there is no need for invasive distal colostogram and micturating cystourethrogram (MCUG) to identify location of the rectourethral fistula. Patients have minimal surgical scar in the perineum and the short term functional outcome is satisfactory. Also we think in this procedure, after fistulae cutting, anterior dissection of the rectum will be minimum and the rectum and urethra don't separate completely therefore the possibility of the rectal prolapsed will decrease. On the other hand, we think in this procedure, after fistulae cutting, anterior dissection of the rectum will be minimum and the rectum and urethra don't separate completely therefore the possibility of the rectal prolapsed will decrease. We recommend further studies with larger number of patients and long term follow in the future.

6- CONFLICT OF INTEREST: None.

7- REFERENCES

1. Brenner EC Congenital defects of the anus and rectum. *Surg Gynecol Obstet.*1915; 20:579-88.
2. Zhu Y-P, Li E-H, Sun W-L, Xu D-L, Liu Z-H, Zhao W, et al. Maternal exposure to di-n-butyl phthalate (DBP) induces combined anorectal and urogenital malformations in male rat offspring. *Reproductive Toxicology.*2016; 61:169-76.
3. Nah SA, Ong CCP, Lakshmi NK, Yap T-L, Jacobsen AS, Low Y. Anomalies associated with anorectal malformations according to the Krickbeck anatomic classification. *J Pediatr Surg.*2012; 47(12):2273-78.
4. De Vos C, Arnold M, Sidler D, Moore S. A comparison of laparoscopic-assisted (LAARP) and posterior sagittal (PSARP) anorectoplasty in the outcome of intermediate and high anorectal malformations. *AJOL.*2011; 49(1):39-43.
5. Arhan P, Faverdin C, Devroede G, Dubois F, Coupris L, Pellerin D. Manometric assessment of continence after surgery for imperforate anus. *J Pediatr Surg.*1976;11(2):157-66.
6. Gołębiewski A, Murawski M, Łosin M, Królak M, Czauderna P. Laparoscopic surgical technique to facilitate management of high anorectal malformations—report of seven cases. *Videosurgery Miniinv.*2011; 6(3):150-54.
7. Raschbaum GR, Bleacher JC, Grattan-Smith JD, Jones RA. Magnetic resonance imaging-guided laparoscopic-assisted anorectoplasty for imperforate anus. *J Pediatr Surg.*2010; 45(1):220-23.
8. Thomas T, Teitelbaum DH, Vellody R, Spremo D, Elkins S, Ladouceur R, et al. Development of an operative suspension system for the performance of MRI-OR-guided laparoscopic anoplasty. *Pediatr surg int.*2016; 32(3):301-6.
9. Schuster SR, Teele RL. An analysis of ultrasound scanning as a guide in determination of "high" or "low" imperforate anus. *J Pediatr Surg.*1979;14(6):798-800.
10. Ale HM, Molaeian M, Salamati P, Pourang H, Sedighi A. Validity of Transperineal Ultrasound in Infants with Imperforate Anus.2004.
11. Khaleghnejad-Tabari A, Saeeda M. The results of posterior sagittal anorectoplasty in anorectal malformations. *Arch Iran Med.*2005; 8(4):272-76.
12. George R, Raschbaum, John C. Bleacher, J. Damien Grattan-Smith, Richard A. Jones. Magnetic resonance imaging-guided laparoscopic-assisted anorectoplasty for imperforate anus. *Journal of Pediatric Surgery.* 2010; 45(1): 220-23. DOI: 10.1016/j.jpedsurg.2009.10.033