

Outcome of Laparoscopic Treatment of Anorectal Malformations in Children

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Abstract

Background Operative correction of anorectal malformations (ARMs) remains a challenge in pediatric surgery. The study aimed to evaluate the outcomes of laparoscopic treatment of ARM in children.

Methods From 2007 to 2014, we performed 104 laparoscopic-assisted anorectal pull-through procedures in children with a mean age of 11.3 ± 0.4 months and ARMs. Clinical assessment, surgical durations, complications, and postoperative outcome were investigated.

Results The mean duration of the operation was 126.5 ± 17.2 min. Mean intraoperative bleeding was 20 ± 5.7 g. Three (2.9 %) children required conversion to laparotomy. One (0.9 %) child developed a pelvic abscess, requiring an additional intervention. Three to six months after laparoscopic-assisted anorectal pull-through, 72 patients were hospitalized for stoma closure. Good functional results were achieved in 39 (54.2 %) patients. On examination of the perineum, 11 (15.3 %) patients were found to have mucosal prolapse. The circular symmetric anal reflex to tactile stimulation was confirmed in 53 (73.6 %) patients and a tactile weakened anal reflex in 5 (6.9 %) children. At the 1-year follow-up, constipation was present in nine (12.5 %) patients and soiling in two (2.8 %) patients. A barium enema study performed after the operation showed good outcomes in 67 (93.1 %) patients and fair outcomes in 5 (6.9 %) patients. None had a poor outcome.

Conclusions Our experience confirms that laparoscopic-assisted anorectal pull-through enabled complete correction of ARM in the meanwhile avoiding damage to the rectum and anus. Also, the technique of double bipolar myostimulation of muscle complex in laparoscopic ARM might lead to these better results.

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Introduction

Anorectal malformations (ARMs) occur at an approximate frequency of one in 4000 live births, a rate that has changed little over time [1, 2]. Operative correction of this pathology remains a challenge in pediatric surgery [3–6]. Despite the development of innovative approaches, the rates of poor results and complications after radical intervention for these anomalies are high, ranging from 45 to 75 % [7, 8].

Several different strategies are used for the operative correction of ARMs. One of the first operations performed by pediatric surgeons was abdominoperineal proctoplasty. The aim of the procedure was to mobilize the left and rectosigmoid segments of the colon through an abdominal access; in the following sacroperineal stage, the mobilized bowel was pulled as close as possible to the sacrum, thus preserving the puborectalis sling [9, 10]. In 1980, posterior sagittal anorectoplasty, developed by Pena et al. [11], was introduced; it has since become the gold standard and therefore one of the most frequently performed surgeries in children with ARMs. This approach correlates positively with the external appearance of the perineum and operative findings. Nonetheless, both abdominoperineal proctoplasty and posterior sagittal anorectoplasty have their advantages and disadvantages [12–14].

A new development in the treatment of high ARMs was introduced in 1990, when Georgeson et al. [15] developed and implemented laparoscopic-assisted pull-through. This method, which has been adopted worldwide [16–18], avoids the division and weakening of the external sphincters. However, improved methods for the operative treatment of children with ARMs are still needed and the immediate and long-term results must be critically assessed [19–21]. In this study, we evaluated the outcomes of laparoscopic treatment of pediatric ARM.

Materials and methods

From 2007 to 2014, we performed 104 laparoscopic-assisted anorectal pull-through procedures in children with ARMs. The patients had a mean age of 11.3 ± 0.4 months, and all procedures were done at the Departments of Pediatric Surgery [Central Clinical Hospital of the Presidential Administration of the Russian Federation, Moscow; Stavropol State Medical University, Stavropol (Russia) and Dnepropetrovsk Regional Children's Hospital, Dnepropetrovsk (Ukraine)]. The studies were approved by the ethics committees of the participating institutions, and informed consent was obtained from the parents of the patients, as their legal representatives. There were 86 (82.7 %) boys and 18 (17.3 %) girls. ARMs in the boys included rectourethral fistula (prostatic and bulbar; $n = 72$, 69.2 %), rectovesical fistula ($n = 8$, 7.7 %), and ARM without fistula ($n = 6$, 5.8 %, Table 1). ARMs in the girls included persistent cloaca ($n = 13$, 12.5 %) and rectovaginal fistula (5, 4.8 %). Girls with persistent cloaca had a short cloacal channel of <3 cm. Two of these patients had a genitourinary sinus <3 cm and high inflow at the rectum into the vagina. All of the patients had different types of stomas: a double-barrel colostomy in 88 (84.6 %), an end

Table 1 Clinical forms of ARM in children

Clinical forms	Boys	Girls
Rectourethral fistula (prostatic and bulbar)	72 (69.2 %)	
Rectovesical fistula	8 (7.7 %)	
ARMs with no fistula	6 (5.8 %)	
Cloaca		13 (12.5 %)
Rectovaginal fistula		5 (4.8 %)
Total	86 (82.7 %)	18 (17.3 %)

colostomy in 3 (2.9 %), a double-barrel ileostomy in 4 (3.9 %), and an end ileostomy in 9 (8.6 %).

The following measures were implemented to prevent complications. First, both a distal loopogram and an urethrogram were performed. Second, colonoscopy was performed through the stoma. Third, five girls with persistent cloaca underwent intravenous contrast-enhanced computed tomography, performed with additional contrast enhancement of the distal colon. Finally, colonoscopy was performed in five children and urethroscopy in seven children. A comprehensive survey helped in planning the operations.

Before starting the operation, the peridural space was catheterized for ropivacaine anesthesia, administered preoperatively and postoperatively to children without sacral anomalies. A sterile 6- to 10-Ch Foley catheter was positioned, depending on the size of the meatus and the age of the child. A 5-mm port was then fitted through a puncture at the top of the navel, after which two 5-mm ports were introduced in the right half of the abdomen. For improved visualization in the deep pelvis, we used a U-stitch on the bladder wall, placed through the abdominal wall. Mobilization was performed only at the distal rectum, avoiding the fistulous opening into the bladder, or into the urethra in boys or the vagina in girls, taking care to prevent future prolapse. The distal part of the colon was transfixed using an extracorporeal or intracorporeal suture, thus cutting off the fistula (Fig. 1). The fistula at the confluence of the pelvic organs (urethra, bladder, and vagina) was ligated with a transfixing suture. Double bipolar myostimulation was then applied using a modified Peña electrostimulator, first in the abdomen and then at the perineum. Laparoscopy included bipolar stimulation of the puborectal muscle via a trocar, followed by bipolar perineal electrostimulation to determine the topographic projection of the anus. The levator ani muscles and the pull-through site were clearly visualized from both the abdomen and the perineum. At the site of the projected perineal anus, the skin was dissected to 1 cm to create a tunnel, which was then progressively dilated using increasingly larger Hegar's dilators. The

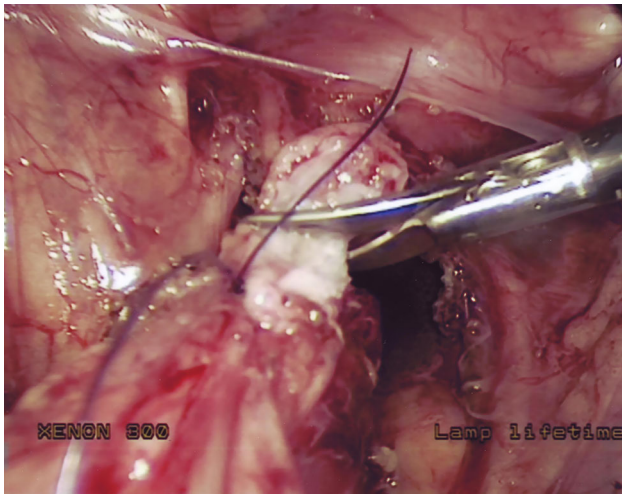


Fig. 1 Laparoscopic separation of the rectum and urethra. After circumferential dissection of the rectum, the fistula is divided and the rectal end is ligated. The fistula is cutoff

distal end of the dissected rectum was brought through the tunnel in the perineum under direct laparoscopic vision. The bowel was fixed to the skin with absorbable sutures (Fig. 2). A partial genitourological mobilization was conducted in five girls with persistent cloaca, using a perineal approach. The abdominal intestine was fixed to the parietal peritoneum of the seromuscular coat. Particular attention was paid to the correct formation of the anal canal in the musculofascial complex of the pelvic diaphragm and an adequate bowel through the anal sphincter.

Postoperatively, all patients were administered antibacterial therapy for 7 days. Feeding began 1–2 days after the operation, when peristaltic sounds were heard. Epidural spinal anesthesia was continued for 2–3 days. The urethral catheter was removed on postoperative day 8. The first anal dilation was started 2 weeks after the operation.

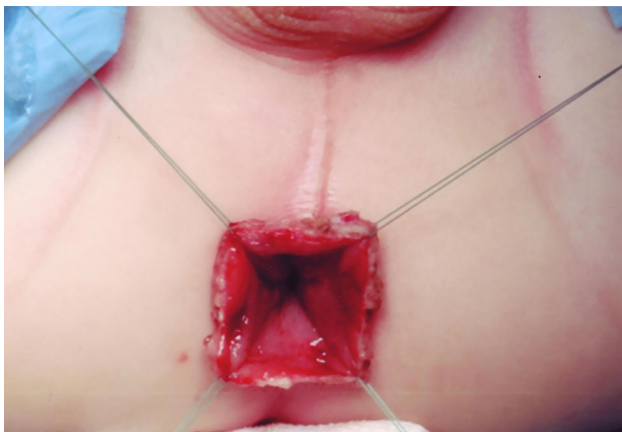


Fig. 2 Reduction and fixation of the neorectum to the perineum

Laparoscopic-assisted anorectal pull-through by ileostomy or closure by colostomy was conducted 3–6 months postoperatively.

The operative techniques and follow-up were similar in all hospitals participating in the study. Two pediatric surgeons from each hospital took part in the surgical and clinical assessments. Follow-up of the patients was conducted within 3–6 months and after more than 1 year after the laparoscopic-assisted anorectal pull-through. The data were recorded in the medical charts. The Krickbeck diagnostic criteria were used in the diagnosis of ARMs and in the postoperative evaluation [22]. Clinical assessment of the circular symmetry of the anal reflex was estimated in all of the patients. Voluntary bowel movements were evaluated in patients older than 5 years of age. All of the patients underwent a barium enema study. The findings were classified according to the three categories defined by Kimura et al. [17]. *Good*: The anorectal angulation was acute, and an empty segment corresponding to the anal canal was recognized. *Fair*: The anorectal angulation was less prominent, and the mucosa of the anal canal was slightly outlined by its barium coating. *Poor*: The anorectal angulation could not be identified. The mucosa of the anal canal was well outlined by its barium coating, and there was no identifiable empty segment. Numerical data are expressed as the mean \pm standard deviation. Student's *t* test was used in the statistical analysis. A *p* value <0.05 was considered to indicate statistical significance. This study was conducted with the approval of the respective institutional review boards.

Results

The mean duration of the operation was 126.5 ± 17.2 min. The amount of intraoperative bleeding was 20 ± 5.7 g. Three (2.9 %) children had technical complications during the early development of this technique and required conversion to laparotomy. Despite careful preoperative preparation, fecal contamination involving the distal colon and the fistula occurred in patients with double-barrel colostomies. One (0.9 %) child developed a pelvic abscess, and thus required an additional intervention. All subsequent patients underwent preoperative colonoscopy with visual assessment of the distal colon. Fecal masses identified on endoscopy were removed.

Three to 6 months after laparoscopic-assisted anorectal pull-through, 72 patients were hospitalized for closure of the stoma. In 16 children, the colostomy was closed at other hospitals (they excluded from our investigation). Good functional results were achieved in 39 (54.2 %) children (Fig. 3). Examination of the perineum revealed mucosal prolapse in 11 (15.3 %) patients and was corrected



Fig. 3 Appearance of the perineum in a male patient 3 months after surgery

with closure of the stoma. Circular symmetry of the anal reflex in response to tactile stimulation was confirmed in 53 (73.6 %) patients, circular asymmetry in 14 (19.5 %) patients, and a weakened anal reflex in 5 (6.9 %) patients. These five patients showed signs of an underdeveloped perineum (absence of the coccyx, sacral hypoplasia, and sagging crotch).

One year after the operation, an evaluation of 72 patients revealed constipation in nine (12.5 %): five (6.9 %) of these patients required laxatives and dietary modifications (grade 2 in the Krickenbeck classification), while the remaining four (5.6 %) required the use of regular cleansing enemas (grade 3 in the Krickenbeck classification). Continence was present in two (2.8 %) children: one (1.4 %) with grade 1 and one (1.4 %) with grade 2, according to the Krickenbeck classification. Barium enema studies performed >1 year after the operation showed that the outcome was good in 67 (93.1 %) patients and fair in 5 (6.9 %) patients; there were no patients with a poor outcome.

Discussion

A laparoscopic procedure is an excellent alternative to laparotomy in children with ARMs, as it significantly reduces the rate of complications such as prolapse, posterior urethral diverticulum, and urethral injuries [16]. Previous publications have confirmed that the outcomes achieved with laparoscopic anorectoplasty are comparable to those of open surgery, based on short- and medium-term follow-up [23–25].

In a study that compared laparoscopic abdominoperineal rectoplasty (ARP) with open ARP [17], the authors investigated the benefits of the laparoscopic approach in infants with high ARMs. The main advantage of laparoscopic versus open ARP was a significant reduction in intraoperative bleeding (12 ± 11 and 65 ± 44 g,

respectively). The anorectal reflex was positive in three (23.1 %) children after laparoscopic ARP and in one (6.7 %) after open ARP. Mucosal prolapse occurred in none of the patients after laparoscopic ARP and in 10 (66.7 %) after open ARP.

A previous study of 15 pediatric patients described an alternative method using the posterior sagittal approach combined with laparoscopy for the repair of recto-bladder-neck fistula or high rectoprostatic fistula [2]. Laparoscopic fistula ligation was performed in all 15 patients. A laparotomy was needed in two (13.3 %) boys to mobilize a very high rectum with selective vessel ligation. Follow-up of the patients 3 months to 10 years after surgery showed that five (33.3 %) boys were fecally incontinent and one (6.7 %) boy was fecally continent. Four (26.7 %) boys underwent surgical repair of a rectal mucosal prolapse.

In our study, data on 104 patients with ARM treated at three pediatric surgical centers during eight consecutive years (2007–2014) were evaluated. Only 84.6 % patients had a standard double-barrel sigmoid colostomy. The other children had atypical stomas (double-barrel ileostomy, end colostomy, and end ileostomy in 3.9, 2.9, and 8.6 %, respectively). Two-thirds of the patients came to us from other hospitals, after stoma creation. One-third of the children had been operated on during the neonatal period, after being diagnosed with a congenital disease (ileal atresia) or necrotizing enterocolitis.

Laparoscopic-assisted anorectal pull-through in children allows direct visualization of the pelvic structures and thus minimizes accidental injury. In addition, it enables identification of the muscular layer, complete fistula excision, and exact distal rectum placement within the muscle complex. Double bipolar myostimulation combines the advantages of laparoscopic and open techniques. The bipolar laparoscopic dissector provided a clear visualization of the levator ani muscles and the pull-through site, with bipolar myostimulation performed through a trocar, as described by Lima et al. [26]. But while those authors only used laparoscopic electrostimulation, we additionally applied a modified Peña bipolar myostimulator to the perineum.

In conclusion, our experience has shown that laparoscopic-assisted anorectal pull-through enables the correction of ARMs and avoids damage to the rectum and anus. The intraoperative technique of double bipolar myostimulation of the muscle complex in laparoscopic ARM probably accounts for the better outcomes. However, our results remain to be confirmed in integrated studies of the long-term results in children with ARMs.

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Compliance with ethical standards

Conflict of interest Drs. Sergey Minaev, Igor Kirgizov, Aleksandr Gladkiy, Ilya Shishkin, and Igor Gerasimenko have no conflicts of interest or financial ties to disclose.

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