Acta Scientific Clinical Case Reports

Volume 3 Issue 5 May 2022

Case Report

Colon Reduction in a Child with Anorectal Malformation with Rectourethral Fistula After Abdominal-perineal Proctoplasty and Coloesophagoplasty of the Left Half of the Colon (Clinical Observation)

Evgeniy A Okulov[1], Aleksey V Dotsenko[1], Igor N Khvorostov[1], Elena Yu Dyakonova[1,2], Aleksey A Gusev[1,2]* and Sergey P Yatsyk[1]

[1]National Medical Research Center for Children's Health, Russia, Moscow, Russia

[2]RUDN University (Peoples' Friendship University of Russia), Moscow, Russia

***Corresponding Author:** Aleksey A Gusev, National Medical Research Center for Children's Health, Russia, Moscow, Russia.

Received: March 11, 2022

Published: April 08, 2022

Abstract

A clinical case of a 10-year-old boy with esophageal atresia with lower tracheoesophageal fistula (Type A) and anorectal malformations (rectourethral fistula) operated on in the neonatal period is presented.

Bougienage was not performed in the postoperative period, which led to anal stenosis and megarectum with a significant expansion of the left colon

The child had an ileostomy. extirpation of the dilated intestine and abdominoperineal anorectoplasty were performed after 6 months.

Long-term results of surgical treatment of a rare complication are presented in the article.

Keywords: Anorectal Malformations (ARMs); Rectourethral Fistula; Child

Introduction

Anorectal malformations (ARMs) occur with the incidence of 1:5000 live births, and have various clinical manifestations. Considerable progress have been made in the management of children with ARMs over the past 20-30 years, both in terms of surgical technologies and in achieving good functional outcomes. However, despite all the technical advances in surgical management of ARMs, still there are complications that require re-operation. Constipation and incontinence are major problems of the postoperative period that require examination. Indications for re-operation are based on the MRI results, visual examination with electro-myographic signals detection, X-ray examination; depend on the type of specific pathology and require individual management approach.

Clinical Case

The child was born at the 38th week of gestation by caesarean section. Vater syndrome was diagnosed immediately after birth. The child underwent surgery on the first day on account of esophageal atresia (type A) with establishment of gastrostomy tube, cervical esophagostomy, and divided colostomy.

Abdominoperineal proctoplasty with disconnection of recto-prostatic fistula was performed at the age of 8 months. Retrosternal coloesophagoplasty with the graft from left colon with left colon artery was performed at the age of 16 months.

Constipation and neoanus stenosis not cured via bougieurage were revealed in the postoperative period after abdominoperineal proctoplasty. Plus, the child had history of urinary tract infections that required inpatient treatment.

The child was examined for the first time in our Center 9 years after previous surgeries with the main complaint on uncontrolled constipation.

Anal stenosis 4 cm long was revealed, it was not passable for bougie №8. Significant enlargement of left half with maximum diameter of up to 14 cm was revealed on irrigography.

Figure 1: Irrigography of 10 years old child with significantly enlarged left colon.

Ileostomy was performed on 03.11.2020.

The size of left colon was decreased by about three times 6 months later.

Figure 2: Irrigography of 10 years old child 6 months after protective ileostomy (diameter has decreased by 3 times).

Next surgery was performed on 02.03.2021: midline laparotomy, total adhesiolysis, resection of lower colon, abdominoperineal proctoplasty, and abdominal drainage.

Surgery description

"Enlarged colon was mobilized along its entire length. Extrarectally precesionic extraction of intestine up to sharply stenosed anal canal was performed. The extirpation was finished on the perineum side. The mobilized intestine was lowered to the perineum without any tension. Electric stimulation has shown circular contraction of external sphincter fibers and associated movements of levators. Muscle contractions were weak but anal canal is in the center of existing perineal muscles. The surgery was completed with anoplasty, bougie №18 has passed freely".

Figure 3: Enlarged left colon was mobilized and extirpated.

Figure 4: The intestine is lowered to the perineum in the center of muscle complex.

Figure 5: Irrigography 6 months after the surgery: no signs of enlarged left colon.

Figure 6: General appearance of patient perineum 6 months after the abdominoperineal proctoplasty: there is moderate mucosal prolapse of lowered intestine.

Ileostomy reversal was performed without any technical difficulties 6 months after radical pull-through.

Physiotherapy treatment for bladder stimulation and endorectal stimulation courses have been started after hospitalization.

Currently, urination is normal, there is no dysuria. There is no residual urine according to control ultrasound examination, and urine is sterile.

Regarding bowel evacuation: The child feels urge to defecate at formed stool, whereas, there is episodes of fecal incontinence in case of loose stool [1-7].

Discussion

- The incidence of anorectal malformations is 1:5000 live births.
- Fistula forms predominate in the structure of pathology in boys. Association of combined malformations in the structure of anorectal malformations does not exceed 7%.
- Considering the rarity of defects combinations in the structure of VATER associations, the majority of such cases require individual surgical treatment.
- Well-recognised treatment strategy for esophageal atresia (Type A) is the application of cervical esophagostomy and gastrostomy followed by delayed coloesophagoplasty at the age between 6 months and 1 year.
- In case of fistula forms of ARMs divided colostomy is required in boys. Radical surgeries on esophagus repair and proctoplasty are performed between the age of 4 and 12 months.
- Later, after radical surgery children require long-term postoperative rehabilitation that include: neoanus bougieurage, individual intestinal control program depending on the type of prevailing disorders - constipation or fecal incontinence.
- Non-compliance to fulfill the principles of postoperative rehabilitation can lead to severe complications up to the need for re-operations.
- Such complications requiring re-operations include: decompensated megarectum, extrasfincter intestine lowering, and cicatricial deformity of neoanus. The incidence of such complications varies from 10 to 60%.
- Combination of anal stenosis and giant megarectum in most cases requires re-operation.
- In our case, the cause of stenosis was hypovascularization of distal parts of lowered intestine combined with inadequate postoperative bougieurage.
- Ileostomy was performed on the first stage.
- The second stage included abdominoperineal lowering of left colon with preliminary assessment of the possibility of lowering itself without blood supply disturbance.

- Otherwise, the right colon should be lowered, but it significantly worsen the prognosis of bowel continence.

Conclusion

- The purpose of this article was to describe the rare clinical case in a patient with combined pathology.
- The correct sequence of staging surgery has allowed to preserve the existing colon as much as possible, to solve the problem of scar anal stenosis, and, thereby, to preserve child's successful socialization.

Bibliography

1. Fernandez E., *et al*. "Esophageal atresia in patients with anorectal malformations". *Pediatric Surgery International* 30.8 (2014): 767-771.
2. Ludwig K., *et al*. "Congenital anomalies of the tubular gastrointestinal tract". *Pathologica* 114 (2022): 40-54.
3. Hamza AF. "Colonic replacement in cases of esophageal atresia". *Seminars in Pediatric Surgery* 18.1 (2009): 40-43.
4. Arnold M., *et al*. "Oesophageal atresia without tracheo-oesophageal fistula and an anorectal malformation: advantages of a primary laparoscopically assisted anorectal pull-through". *South African Journal of Surgery* 49.1 (2011): 30-32.
5. Panda SS., *et al*. "Esophageal atresia, duodenal atresia, and imperforate anus: Triple atresia". *Journal of Clinical Neonatology* 4 (2015): 188-192.
6. Byun SY., *et al*. "Anorectal malformations associated with esophageal atresia in neonates". *Pediatric Gastroenterology, Hepatology and Nutrition* 16.1 (2013): 28-33.
7. Iozsa Dan Alexandru., *et al*. "Esophageal atresia associating gastrointestinal malformations: a study of clinical approach". *Journal of Mind and Medical Sciences* 8.2 (2021).