

Aponeurotic Ring-Assisted Stoma Formation in Pediatric Anorectal Malformations: Surgical Stages and Early Outcomes

K. Ashirbay^{1,2*}, A.Dzhumabekov², A.Kussainov^{1,3}, G.Altynbayeva^{1,3}, E.Aitbayeva⁴, K.Khaironov¹, S.Saliyeva^{1,3}, N.Shilanbayev^{1,2}, G. Kaukenbayeva¹.

ABSTRACT

Aim

The aim of this study was to analyze the early results of creating a colostomy in children with anorectal malformations (ARM) by forming an aponeurotic ring. The frequency of postoperative complications after using a modified technique and a traditional approach was compared.

Methods

A retrospective study was carried out in Scientific Center of Pediatrics and Pediatric Surgery, Almaty, Kazakhstan from 2014 to 2023. The 63 patients under the age of 4 with ARM were included in the study and divided into two groups. The former group comprised patients who had stoma creation performed via the conventional technique. The second group consisted of patients who had stoma established by forming an aponeurotic ring.

Results

The predominant types of surgical intervention in both groups were end and divided colostomies. In the first group of 35 children, 10 had postoperative complications (28.5%). Early complications developed in 4 children, late complications in 6 children. In the second group of 28 children, complications developed in 2 patients (7.2%): in one case on the 6th day of life, and in another case after 2 months. Out of 63 children, one child died during treatment.

Conclusions

Compared to the conventional method, an aponeurotic ring-assisted stoma formation method allowed to reduce the risk of early and late stoma-associated complications by 4 times (RR=0.25, p=0.051).

Keywords

Anal atresia; anorectal malformation; colostomy; children.

INTRODUCTION

Anorectal malformations (ARM) are one of the most common indications for colostomy in pediatric practice⁽¹⁻³⁾. A protective colostomy in ARM serves as a temporary solution until final surgical correction is performed. The creation of a colostomy is essential for relieving bowel obstruction, preventing urine contamination by fecal matter when a rectourinary fistula is present, and safeguarding upcoming perineal reconstruction procedures^{^4,5}. Despite its importance, the optimal stoma type for children with anorectal malformations (ARMs) remains a subject of debate^{^6}. The two most frequently used techniques are the divided descending colostomy with a distal mucus fistula and the loop colostomy⁵. The divided descending colostomy is frequently chosen over the loop colostomy because of a lower incidence of prolapse and urinary tract infections⁽⁴⁻⁶⁾.

However, the frequency of complications associated with colostomy remains high, occurring in up to 70% of cases after formation and up to 29% after closure^(4,5). Post-reconstructive complications develop in

1. Scientific Center of Pediatrics and Pediatric Surgery», Almaty, Republic of Kazakhstan;
2. Kazakhstan Medical University «Higher School of Public Health», Almaty, Republic of Kazakhstan;
3. «Asfendiyarov Kazakh National Medical University», Almaty, Republic of Kazakhstan;
4. «Al-Farabi Kazakh National University», Almaty, Republic of Kazakhstan.

Correspondence

K. Ashirbay, Scientific Center of Pediatrics and Pediatric Surgery», Almaty, Republic of Kazakhstan, Kazakhstan Medical University «Higher School of Public Health», Almaty, Republic of Kazakhstan kana_089@mail.ru

5-40% of cases⁽⁷⁻¹⁵⁾. Given the significant frequency of complications associated with colostomy, researchers continue to search for new methods of creating a stoma aimed at reducing the frequency of complications and accelerating recovery in the early postoperative period.

Since 2020, our clinic has been creating colostomies by forming an aponeurotic ring. In this article, we decided to share the early results of colostomy creation in children with ARM by forming an aponeurotic ring. We conducted a comprehensive retrospective analysis of patients treated at our institution over a ten-year period and analyzed the frequency of complications associated with stomas created using the traditional method and the new method.

MATERIALS AND METHODS

Study population

A retrospective study was conducted involving all patients with ARM who underwent colostomy between January 1, 2014, and December 31, 2023, at our tertiary care hospital (Scientific Center for Pediatrics and Pediatric Surgery, Almaty, Kazakhstan).

The 63 patients under the age of 4 with ARM were included in the study and divided into two groups. The first group included patients who underwent stoma formation using the traditional method. The second group consisted of patients whose stoma was formed by creating an aponeurotic ring. Informed consent was obtained from all parents of participants. The study was conducted in full compliance with the guidelines of the Helsinki Declaration on the protection of human research subjects.

The types of stoma and the frequency of stoma-related complications were analyzed. The type of colostomy was subdivided into loop stomas, defined as colostomies with at least one continuous layer of mucosa, and divided colostomies, characterized by complete separation of the mucosal layer between the distal and proximal stomas. An end colostomy attaches one end of the colon or ileum to an opening in the abdominal wall.

Complications were defined as unexpected events unrelated to the underlying disease that led to rehospitalization, reoperation, or an unplanned outpatient visit. Complications were divided into early (less than 1 month) and late (more than 1 month) from the time of surgery.

In addition, we assessed the frequency of associated malformations and epidemiological data.

ARM was classified according to the Krikenbeck classification¹⁶ into low-type malformations (anal stenosis, perineal fistula, and vestibular fistula) and high-type malformations (urethral fistula [bulbar or prostatic], rectovesical fistula, rectovaginal fistula, anal atresia without fistula, sacral colon, rectal atresia, and persistent cloaca).

Surgical technique

An incision is made on the skin at the proposed site for the stoma. Then, access to the abdominal cavity is gradually achieved. To create the ring, 6-8 peritoneal-aponeurotic interrupted sutures are applied to fix the intestinal wall, which also serve to open the surgical wound. Then, the location of the right section of the intestine is determined and it is completely severed. The distal part of the cut intestine is completely closed with sutures and inserted into the abdominal cavity. The transected proximal part of the intestine with the mesentery is passed through the wound and fixed to the previously placed knots of the peritoneal-aponeurotic ring, followed by the application of skin-mucosal sutures to the stoma, creating a “rose.”

The proposed method for creating an intestinal stoma is explained below with graphic material (Fig. 1).

Figure 2 illustrates the intraoperative images.

Statistical methods Clinical data were entered into a database (Microsoft Excel 2019) and statistical analysis was performed using SPSS. Qualitative variables are reported as absolute numbers or percentages. To determine whether the observed difference in complication rates between the two groups was statistically significant, Fischer’s exact test was used and the relative risk was calculated. The significance level was set at $p < 0.05$ to determine reliability.

Ethical clearance

This study was conducted in accordance with ethical standards. Ethical approval was obtained from the appropriate institutional review board, and informed consent was secured from all participants prior to data collection.

RESULTS

In the period under analysis, a total of 63 cases of ARMs were identified. The average birth weight of all patients was 3171 ± 580 g and the average gestational age was 37.6 ± 2.0 weeks.

The patients were divided into two groups. The first group included 35 children, which amounted to 55.5% of the total sample and included 9 boys and 26 girls with a median follow-up of 7 years and 1 month. The second group consisted of 28 children, which accounted for 44.5% of the total sample, including 7 boys and 21 girls. The median follow-up was 2 years and 1 month (Fig. 3).

Current global statistics indicate that ARMs are more common in boys, accounting for 58% of cases. Among these children, 72% have malformations of moderate or high severity, which are very difficult to treat⁽¹⁷⁻²⁰⁾. In our study the gender ratio was 1:2. The predominance of female children in our study is explained by the fact that our center is a tertiary facility, and patients with complex forms of ARM are referred mainly, so cloaca and vestibular fistula were diagnosed in 38 children, which was 60.1% of the total sample.

The total numbers of ARM types according to the classification and prevalence of associated malformations are shown in Table 1.

Anus atresia occurred with fistula in 40 patients (63.4%) and without fistula in 14 patients (22.2%). Persistent cloaca and cloaca extrophy was diagnosed in 8 patients (12.7%).

The frequency of associated congenital malformations in our cohort was 33.3%, including congenital heart defects in 16 children and urogenital abnormalities in 5 children. According to the ARM-Net registry, the association with anomalies of other organs is 58-78%, indicating the importance of comprehensive examinations in all patients with anorectal pathologies²¹.

One patient was diagnosed with Down syndrome, which amounted to 1.6% in the study cohort and is consistent with the literature, which reports that the incidence of Down syndrome in patients with anus atresia is about 2%, whereas in the normal population it is 0.15%^(22,23).

After examination, the children underwent surgical treatment: in the absence of anus - within 15-20 hours, in the case of fistula, surgery was performed as planned.

A summary of stoma-related complications according to the location of stoma is presented in Table 2. The predominant types of surgical intervention in both groups were divided and end colostomies. In the case of atresia with a fistula an end colostomy and in cases of complete atresia, a divided or loop colostomy were performed.

In the first group of 35 children, 10 had postoperative complications. In 4 children, complications developed in the first week after surgery in the form of evagination, eventration, and intestinal obstruction, while in 6 children, complications developed 2-3 months after surgery and were presented as paracolostomy hernia, stenosis, eventration, and stoma insufficiency. In the second group of 28 children, complications developed in 2 patients: one child developed evisceration on the 6th day of life, and another child developed a paracolostomy hernia after 2 months. That is, patients operated with the modified method had a 4-fold lower risk of complications (RR=0.25, p=0.051) than with the traditional method.

The duration of surgery in group 1 lasted 40min on average, in group 2 not more than 30min. After surgery, all children were transferred to the intensive care unit and received appropriate treatment. Enteral nutrition in 80% of cases was started within 3-6 hours after open tube surgery in all groups.

Out of 63 children, one died during treatment. The cause of death was late diagnosis of the twistless form of anus atresia (more than 48 hours), VACTER association. The infant underwent surgery at the 52nd hour of life.

Anoplasty was performed at an average of 10 months after colostomy, and the stoma was closed at an average of 2.5 months after stage 2 surgery.

During surgery, it was observed that intraperitoneal adhesions formed in group 1 patients, which resulted in an increase in operative time to 40 minutes. In contrast, no adhesions were observed in group 2, resulting in a smoother procedure, shorter operation time (10 minutes), and shorter anesthesia duration.

DISCUSSION

More than 300 years have passed since the first colostomy was performed on a patient with rectal atresia by the French physician A. Littre⁽²⁴⁻²⁶⁾. To this day, pediatric surgeons have invented new methods for constructing intestinal stomas, but the results of various studies demonstrate that the frequency of complications associated with stomas ranges from 10 to 82%⁽²⁷⁻³²⁾. The authors attribute the causes of complications to both the continuation of the inflammatory process in the abdominal cavity and intestine, as well as technical difficulties. Early complications within the first 30 days include bleeding, hematoma formation, stoma

edema, skin irritation, sometimes with ulceration and stoma necrosis, retraction, evagination, and stoma eventration. Late complications, which appear more than 30 days post-surgery, most commonly include stoma stenosis, stoma prolapse, and parastomal hernia. The causes of most early complications are suboptimal stoma creation and insufficient care. The causes of late complications may be related to increased intra-abdominal pressure, which significantly increases the risk of stoma prolapse and parastomal hernia. An excessively large opening is a predisposing factor for a parastomal hernia, while excessive mobilization of the intestinal loop used to form the stoma increases the tendency for prolapsed⁽³³⁻³⁷⁾.

In our study, colonic stomas (colostomies) were performed in 96.8% of cases. End colostomy was performed in cases of fistulous atresia, while divided and loop colostomy were performed in cases of complete atresia. End colostomies are generally contraindicated irrespective of the type of ARM. This is primarily because they limit the ability to perform an augmented pressure distal colostogram at a later stage. We, such as G Brisighelli et al⁹ believe that specific types of ARMs, mainly rectoperineal and recto-vestibular fistulas, could benefit from an end colostomy as the presence of a mucus fistula to decompress the defunctionalised bowel is not needed and performing an augmented pressure distal colostogram would not be necessary to delineate the anatomy before the PSARP.

A total of 7 loop colostomies were performed, complications were observed in 3 (42.8%) patients. Divided colostomy was performed on 27 patients, complications were recorded in 5 (18.5%). End colostomy was performed on 27 patients, complications occurred in 2 (7.4%). Ileostomy was performed on 2 patients, and both experienced stoma eventration the early postoperative period.

When analyzing the frequency of complications in the first group, stoma-related complications occurred in 28.5% of patients, while in the second group, they occurred in 7.2%. Complications occurred both early and late in both groups.

The reduction of complications in the second group is attributed to the fact that the formation of the peritoneal-aponeurotic ring provides good visualization during stoma creation, allows for the assessment of the degree of intestinal compression attached to the ring during the operation, and evaluates the trophic function of the

vascular system. This technique also reduces the risk of intestinal wall trauma and preserves the integrity of the muscular-aponeurotic structures of the abdominal wall, as there is no need to use additional dilating instruments for better visualization. This helps prevent the development of necrosis in the diverted intestine.

The created aponeurotic ring ensures a stable position of the stoma, which reduces the risk of complications such as stoma prolapse and retraction, evagination, and paracolostomy hernia. The ring creates conditions for more effective drainage of intestinal contents, reducing the infection of wound edges by intestinal contents. Due to better tissue adhesion and less scarring, an improved cosmetic result is achieved, and the development of stenosis can be prevented.

When using this method, the invasion of the abdominal organs is minimal, the blood supply to the mesentery (arcade) is not disrupted, and consequently, the likelihood of developing adhesions in the abdominal cavity in the future is reduced.

Reducing the duration of the operation with the new method allowed us to shorten the duration of anesthesia.

The length of the incision for performing the surgery is also important; in our method, an incision of no more than 3 cm is sufficient, and accordingly, a better cosmetic result can be achieved.

The method technically simplifies the performance of surgery to restore the continuity of the gastrointestinal tract and the anatomical integrity of the anterior abdominal wall during repeated reconstructive surgery, and it improves the patient's quality of life after the operation.

CONCLUSION

Our study adds to the growing body of literature on ARM management. Our proposed method demonstrates a promising reduction in stoma-related complications compared to conventional techniques (RR = 0.25, $p = 0.051$). However, due to the retrospective design and limited sample size, further studies with larger cohorts and prospective methodology are necessary to validate these findings. Continued research and experience-sharing will further refine the approach to managing a creation of stoma in patients with ARM, with a focus on improving long-term outcomes and patient quality of life.

ACKNOWLEDGMENTS The authors are thankful to the physicians who permitted us to conduct our study. We

are also thankful to the patients for extending the full cooperation to the study.

Source of fund: (if any). No Funding

Conflict of Interest: The authors declare no conflict of interest. participants prior to data collection.

Authors's contribution: Data gathering and idea owner

of this study: A.Dzhumabekov, K.Khairrov

Study design: A.Kussainov, N.Shilanbayev

Data gathering: E.Aitbayeva, S.Saliyeva

Writing and submitting manuscript: G.Altynbayeva

Editing and approval of final draft: K. Ashirbay, G. Kaukenbayeva

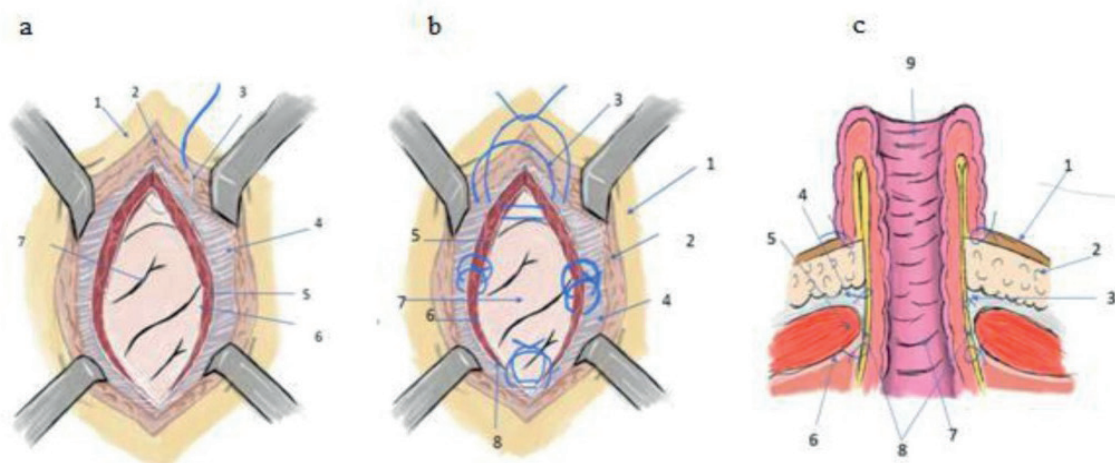


Figure 1: Schematic representation of the stages of creating a peritoneal-aponeurotic ring and proximal colostomy (a) stage of interrupted suture, (b) stage of peritoneal-aponeurotic ring creation, (c) stage of creating a proximal colostomy

1 - skin, 2 - subcutaneous adipose tissue, 3 - placement of interrupted suture, 4 - aponeurosis, 5 - muscles of the anterior abdominal wall, 6 - peritoneum, 7 - small intestine, 8 - construction of peritoneal-aponeurotic ring, 9 - the created proximal colostoma.

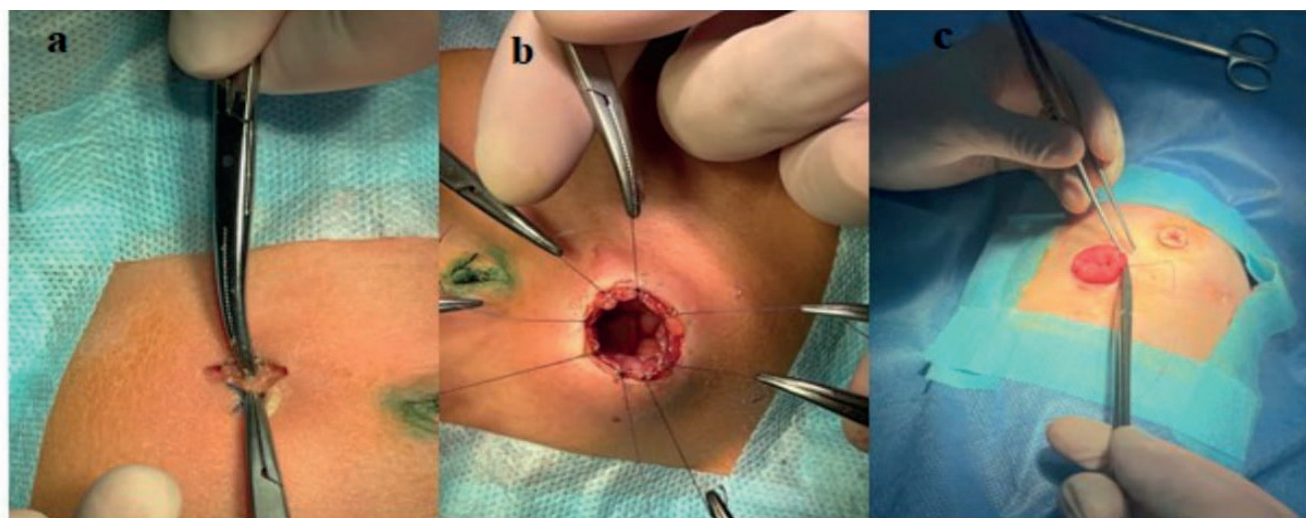


Figure 2: Creation of the peritoneal-aponeurotic ring and colostomy (intraoperative data)

- a) skin incision not more than 3 cm, b) creation of the peritoneal-aponeurotic ring with interrupted sutures, c) the created colostomy.

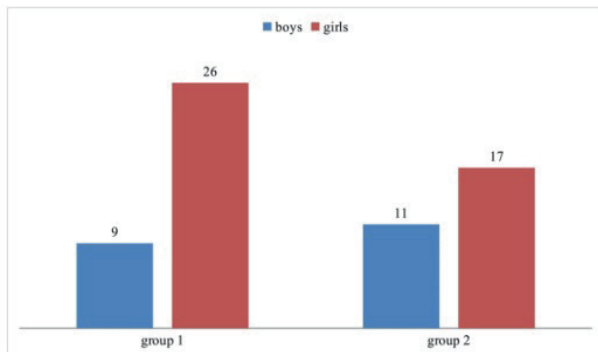


Figure 3: Gender distribution of patients

Table 1: General data of the patients included in the study

Indicators	Categories	Group 1	Group 2
Type of ARM	Anus atresia without fistula	10	4
	Anus atresia with fistula	rectourethral	1
		rectovesical	1
		vestibular	17
		perineal	1
	Persistent cloaca	3	1
	Cloaca extrophy	1	3
	Anal stenosis	1	-
Associated malformations	Congenital heart defects (CHD)	9	7
	Congenital malformations of the urinary system	3	2
	Down syndrome	1	-

Table 2: Types of surgical interventions for stoma creation and postoperative complications

Categories	Surgical intervention	Amount of intervention	Amount of complications	Types of complications	
				Early complications	Late complications
Group 1	Loop colostomy	6	3	-	eventration after 2 months paracolostomy hernia after 3 months, stoma stenosis after 2 months
	End colostomy	10	2	intestinal obstruction on day 6, evagination on day 5	-
	End ileostomy	1	1	eventration on day 8	-
	Divided colostomy	18	4	eventration on day 5	paracolostomy hernia in 2 patients after 3 months insolvency after 3 months
Group 2	Loop colostomy	1	-	-	-
	End colostomy	17	-	-	-
	End ileostomy	1	1	eventration on day 6	-
	Divided colostomy	9	1	-	paracolostomy hernia after 3 months

REFERENCES

1. Millar AJ, Lakhoo K, Rode H, Ferreira MW, Brown RA, Cywes S. Bowel stomas in infants and children. A 5-year audit of 203 patients. *S Afr J Surg* 1993;**31**:110–113.
2. Nour S, Beck J, Stringer MD. Colostomy complications in infants and children. *Ann R Coll Surg Engl* 1996;**78**:526–530.
3. Cigdem MK, Onen A, Duran H, Ozturk H, Otcu S. The mechanical complications of colostomy in infants and children: analysis of 473 cases of a single center. *Pediatr Surg Int* 2006;**22**:671–676.
4. Hartford L, Brisighelli G, Gabler T, Westgarth-Taylor C. Single-stage procedures for anorectal malformations: A systematic review and meta-analysis. *J Pediatr Surg* 2022;**57**:75–84.
5. Youssef F, Arbash G, Puligandla PS, Baird RJ. Loop versus divided colostomy for the management of anorectal malformations: A systematic review and meta-analysis. *J Pediatr Surg* 2017;**52**:783–790.
6. Bischoff A, Levitt MA, Peña A. Update on the management of anorectal malformations. *Pediatr Surg Int* 2013;**29**:899–904.
7. van den Hondel D, Sloots C, Meeussen C, Wijnen R. To split or not to split: colostomy complications for anorectal malformations or Hirschsprung disease: a single center experience and a systematic review of the literature. *Eur J Pediatr Surg* 2014;**24**(1):61–69.

8. Bischoff A, Peña A, Levitt MA. Laparoscopic-assisted PSARP - the advantages of combining both techniques for the treatment of anorectal malformations with recto-bladderneck or high prostatic fistulas. *J Pediatr Surg* 2013;**48**(2):367–371.
9. Brisighelli G, Di Cesare A, Morandi A, Paraboschi I, Canazza L, Consonni D, et al. Classification and management of rectal prolapse after anorectoplasty for anorectal malformations. *Pediatr Surg Int* 2014;**30**(8):783–789.
10. Divarci E, Ergun O. General complications after surgery for anorectal malformations. *Pediatr Surg Int* 2020;**36**(4):431–445.
11. Holbrook C, Misra D, Zaparackaite I, Cleeve S. Post-operative strictures in anorectal malformation: trends over 15 years. *Pediatr Surg Int* 2017;**33**(8):869–873.
12. Jenetzky E, Reckin S, Schmiedeke E, Schmidt D, Schwarzer N, Grasshoff-Derr S, et al. Practice of dilatation after surgical correction in anorectal malformations. *Pediatr Surg Int* 2012;**28**(11):1095–1099.
13. Kumar B, Kandpal DK, Sharma SB, Agrawal LD, Jhamariya VN. Single-stage repair of vestibular and perineal fistulae without colostomy. *J Pediatr Surg* 2008;**43**(10):1848–1852.
14. Short SS, Bucher BT, Barnhart DC, Van Der Watt N, Zobell S, Allen A, et al. Single-stage repair of rectoperineal and rectovestibular fistulae can be safely delayed beyond the neonatal period. *J Pediatr Surg* 2018;**53**(11):2174–2177.
15. Tofft L, Salö M, Arnbjörnsson E, Stenström P. Wound dehiscence after posterior sagittal anorectoplasty in children with anorectal malformations. *BioMed Res Int* 2018;2018:2930783.
16. Holschneider A, Hutson J, Peña A, Beket E, Chatterjee S, Coran A, et al. Preliminary report on the International Conference for the Development of Standards for the Treatment of Anorectal Malformations. *J Pediatr Surg* 2005;**40**(10):1521–1526.
17. Babaei H, Ahmadipour S, Mohamadimoghdam J, Mohsenzadeh A. The Study of Newborns with Congenital Gastrointestinal Tract Obstruction. *J Krishna Inst Med Sci Univ* 2014;**3**(2):101–106.
18. Smith CA, Avansino J. Anorectal Malformations. 2023 Aug 8. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2024. PMID: 31194415.
19. Singh M, Mehra K. Imperforate Anus. 2023 Aug 28. In: StatPearls [Internet]. Treasure Island (FL): StatPearls Publishing; 2024. PMID: 31747191.
20. Levitt MA, Peña A. Anorectal malformations. *Orphanet J Rare Dis* 2007;**2**:33.
21. Hageman IC, Midrio P, van der Steeg HJJ, Jenetzky E, Iacobelli BD, Morandi A, et al; ARM-Net Consortium. The European Anorectal Malformation Network (ARM-Net) patient registry: 10-year review of clinical and surgical characteristics. *Br J Surg* 2024;**111**(2):znae019.
22. Matias A, Montenegro N, Blickstein I. Down syndrome screening in multiple pregnancies. *Obstet Gynecol Clin North Am* 2005;**32**(1):81–96.
23. de Buys Roessingh AS, Mueller C, Wiesenauer C, Bensoussan AL, Beaunoyer M. Anorectal malformation and Down's syndrome in monozygotic twins. *J Pediatr Surg* 2009;**44**(2):e13–6.
24. Schärli WF. The history of colostomy in childhood. *Prog Pediatr Surg* 1986;**20**:188–98.
25. Yesildag E, Muñiz RM, Buyukunal SN. How did the surgeons treat neonates with imperforate anus in the eighteenth century? *Pediatr Surg Int* 2010;**26**(12):1149–1158.
26. Schärli AF. Malformations of the anus and rectum and their treatment in medical history. *Prog Pediatr Surg* 1978;**11**:141–172.
27. Harris DA, Egbeare D, Jones S, Benjamin H, Woodward A, Foster ME. Complications and mortality following stoma formation. *Ann R Coll Surg Engl* 2005;**87**(6):427–431.
28. Formijne Jonkers HA, Draaisma WA, Roskott AM, van Overbeeke AJ, Broeders IA, Consten EC. Early complications after stoma formation: a prospective cohort study in 100 patients with 1-year follow-up. *Int J Colorectal Dis* 2012;**27**(8):1095–1099.
29. Nastro P, Knowles CH, McGrath A, Heyman B, Porrett TR, Lunniss PJ. Complications of intestinal stomas. *Br J Surg* 2010;**97**(12):1885–1889.
30. Caricato M, Ausania F, Coppola R. Temporary stoma after elective anterior resection of the rectum: an unsolved debate. *Colorectal Dis* 2005;**7**(2):196.
31. Robertson I, Leung E, Hughes D, Spiers M, Donnelly L, Mackenzie I, et al. Prospective analysis of stoma-related complications. *Colorectal Dis* 2005;**7**(3):279–285.
32. Vogel I, Eeftink Schattenkerk LD, Venema E, Pandey K, de Jong JR, Tanis PJ, et al. Major stoma related morbidity in young children following stoma formation and closure: A retrospective cohort study. *J Pediatr Surg* 2022;**57**(10):402–406.
33. Ambe PC, Kurz NR, Nitschke C, Odeh SF, Möslin G, Zirngibl H. Intestinal Ostomy. *Dtsch Arztebl Int* 2018;**115**(11):182–187.
34. Shabbir J, Britton DC. Stoma complications: a literature overview. *Colorectal Dis* 2010;**12**(10):958–964.
35. Chong C, van Druten J, Briars G, Eaton S, Clarke P, Tsang T, et al. Neonates living with enterostomy following necrotising enterocolitis are at high risk of becoming severely underweight. *Eur J Pediatr* 2019;**178**(12):1875–1881.
36. Wolf L, Gfroerer S, Fiegel H, Rolle U. Complications of newborn enterostomies. *World J Clin Cases* 2018;**6**(16):1101–1110.
37. Bethell G, Kenny S, Corbett H. Enterostomy-related complications and growth following reversal in infants. *Arch Dis Child Fetal Neonatal Ed* 2017;**102**(3):F230–F234.