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Early diagnosis and surgical intervention untie the Gordian knot in newborns with colonic atresia: report of two cases and review of the literature

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Abstract: Incidence of colonic atresia in living infants ranges from 1:5,000 to 1:60,000 (average 1:20,000). It constitutes 1.8 to 15% of all cases of atresia of the gastrointestinal tract. In 58.56–75% of all cases is right-sided.

We aim, through the presentation of two cases of colonic atresia which we encountered and after systematic research of the current literature, at addressing three major issues: diagnostic approach, operative strategy and management of the prognostic parameters of the colonic atresia.

The common parameter in these two cases was the early diagnosis, which played a significant role in the uncomplicated postoperative course. The first case was a type I sigmoid atresia. Contrast's escape during contrast enema examination due to accidental rupture of the distal part of the colon led to diagnosis. Side-to-side anastomosis, restoration of the rupture and a central loop sigmoidostomy were urgently performed. The second case was a type III atresia at the level of the ascending colon, which was early diagnosed via pregenital ultrasonography, in which colonic dilation was depicted. Restoration of the intestinal continuity early after birth was performed at a time.

In conclusion, we believe that early diagnosis, selection of the appropriate operative strategy and prompt recognition of potential post-operative complications, especially rupture of the anastomosis, contribute to the optimization of the prognosis in patients with colonic atresia.

Key words: colonic atresia, contrast enema, pregenital diagnosis, microcolon, newborn.

Introduction

Incidence of colonic atresia in living infants ranges from 1:5,000 to 1:60,000 (average 1:20,000). It constitutes 1.8 to 15% of all cases of atresia of the gastrointestinal tract (GI) [1–4]. It is more frequent in male infants (M/F: 1.33–2.2) [5, 6]. In 58.56–75% of all cases is right-sided [6, 7].

According to Louw and Barnard [5] and Harris *et al.* [8] colonic atresia is associated with the impact of a vascular event that occurs in the prenatal period; a consequence of either an embolic or thrombotic event in the placenta or herniation, torsion or confinement of an internal hernia. Guttmann *et al.* [9] describe comprehensively the possible channelization failure in the context of the transition of the GI tract from the solid to the tubular phase (revacuolization of the solid cord stage). Al Wafi *et al.* [10] refer to the atresia of the transverse colon due to the presence of a sizeable choledocheal cyst and the pressure it exerts. Schiller *et al.* [11], Hitchcock *et al.* [12], Sauve *et al.* [13], Rigsby *et al.* [14] and Alkalay *et al.* [15] attribute colonic atresia to intrauterine infection from Varicella Zoster virus (VZV, Fetal Varicella Syndrome). VZV causes damage to the intestinal nerve plexus and thus, due to defective development of the intestinal vascular branches, leads to ischemia. In a retrospective study by Etensel *et al.* [6] this hypothesis was confirmed in 3 of all 224 cases (6.72%). Fourcade *et al.* [16], through their experimental study, provide a general interpretative vision of the colonic atresia, concluding that is in general a disorder of morphogenesis of the GI tract.

Croaker *et al.* [17], Kim *et al.* [18], Williams *et al.* [19], Powell *et al.* [20], Kamak *et al.* [21], Cox *et al.* [22] and Anveden-Hertzberg *et al.* [23] estimate that in one third of all cases of colonic atresia other congenital anomalies co-exist, such as complex abnormalities of the urinary tract, skeletal and ocular abnormalities, malformation of the extra-hepatic bile ducts, abdominal schises and multiple types of atresia. Especially in patients with gastroschisis, rectal atresia co-exists in 2.5% of all cases [24]. In their retrospective clinical study, Etensel *et al.* [6] estimated that in 47.3% (106/224) of all patients suffering from colonic atresia there was at least one concomitant congenital anomaly. Gobran *et al.* [25] encountered 13 patients with colonic atresia. In eight of them (8/13, 61.53%) other congenital abnormalities co-existed.

Based on the revised classification of Martin *et al.* [26] and Grosfeld *et al.* [27], colonic atresia is divided into 4 types (Table 1).

We aim, through the presentation of two cases of colonic atresia which we encountered and after systematic research of the current literature, at addressing three major issues: proper diagnostic approach, operative strategy and management of the prognostic parameters of the colonic atresia.

Table 1. Colonic atresia classification by Martin *et al.* [26] and Grosfeld *et al.* [27].

Type	Characteristics
I	Bowel lumen is interrupted by a membrane, while mesentery is intact
II	Bowel is discontinuous; the two atretic segments are connected by a fibrous cord
III (A-B)	A: there is distance between the two atretic segments and V-like deficit of the mesentery B: apple peel atresia
IV	Multiple atresias

Case 1

A 2-days old newborn (38 weeks of gestation, birth weight 2800 grams, free prenatal history, and vaginal delivery) was referred to our Department from a Neonatology Department due to failure to pass meconium, feeding refusal and bilious vomiting. Neonatologists had already attempted contrast enema examination with gastrografin, via a catheter which was launched 7 cm centrally to the anal ring.

During the initial clinical assessment of the patient, the main findings were: metallic bowel sounds, abdominal distension and visible intestinal loops in the anterior abdominal wall. Digital rectal examination revealed the absence of stool. Alongside with the diagnostic approach, the patient was supported with intravenous fluids (R/L). Performance of nasogastric decompression through an 8 Fr nasogastric tube led to the aspiration of 25 ml of gastric content with bilious impurity. We also administered intravenous cefuroxime 80 mg/kg in 3 daily doses and metronidazole 7.5 mg/kg in 3 daily doses. After conduction of erect abdominal radiograph, we evaluated the contrast's escape in the lesser pelvis, without the presence of free subdiaphragmatic air (Fig. 1).

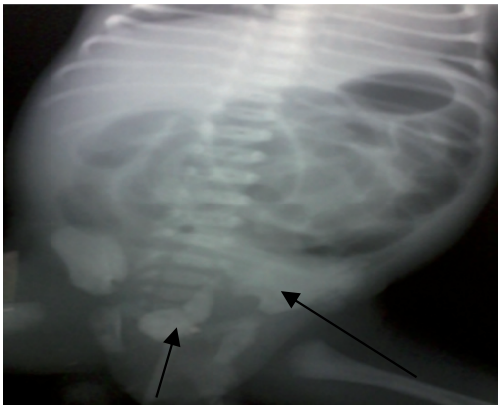

Fig. 1. Presence of the contrast in the rectum and in the lesser pelvis (arrows).

After stabilization of the newborn's vital signs, urgent exploratory laparotomy followed with transverse subumbilical incision, under general endotracheal anesthesia. The presence of dilated sigmoid was revealed, with a diameter of 4.5 cm. It extended 8 cm above the recto-vesical pouch. The peripheral segment of the colon — without disruption of its continuity (type I of atresia) — had macroscopic features of microcolon, with a diameter of 1.2 cm (ratio of the diameter of the two colonic segments = 3.75).

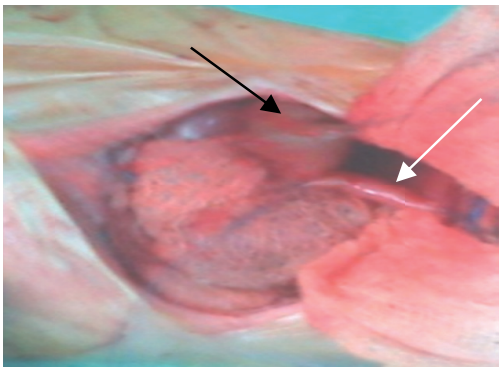

Fig. 2. The proximal segment (black arrow) with a diameter of 4.5 cm and the distal segment (white arrow) with a diameter of 1.2 cm. There is no disruption in the continuity of the serous membrane (atresia type I). The two segments are mutually perpendicular.

A linear rupture 1 cm in length was found in the anterior surface of the microcolon and 2 cm above the recto-vesical pouch. After thoroughly washing the peritoneal cavity, we conducted a side-to-side anastomosis 2 cm in length, after wall section, laterally in the central (dilated) and longitudinally in the peripheral (microcolon) segment of the sigmoid (Fig. 3 and 4). The operation was completed with the restoration of the rupture located in the anterior surface of the microcolon in 2 layers with PDS 5/0, after taking full thickness biopsy from the margins of the rupture, and the conduction of a loop sigmoidostomy centrally.

The newborn was hospitalized in a neonatal intensive care unit for 2 days postoperatively. On the 3rd post-operative day, after documenting that the sigmoidostomy is functional, we removed the nasogastric tube and we gradually initiated oral feeding.

Post-operative course was uneventful. The newborn was discharged home on the 8th post-operative day, with instructions to the parents to perform colonic washout through the peripheral orifice of the loop sigmoidostomy with 20 ml N/S twice a day.

The diagnostic approach was completed three weeks later, after conduction of examination by a pediatric cardiologist and ultrasound examination of the spine and the urinary tract. No concomitant congenital anomalies were found.

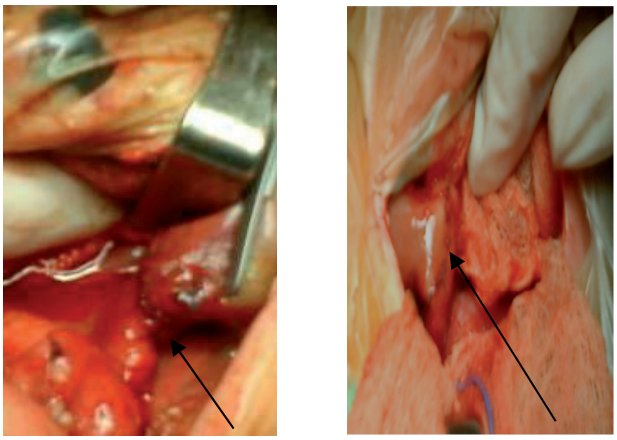

Fig. 3, 4. Conduction of the side-to-side anastomosis (arrows).

During the 6th post-operative week, after administrating contrast through the peripheral orifice of the loop sigmoidostomy and successful imaging both of the anastomosis and of the diameter of the peripheral colonic segment, we decided the occlusion of the sigmoidostomy (Fig. 5).

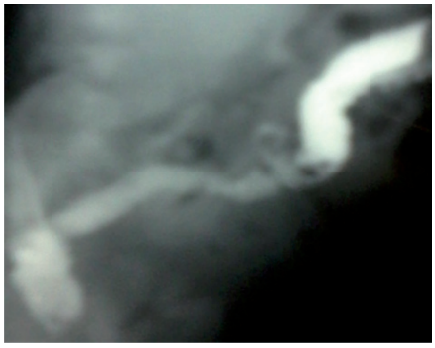

Fig. 5. Administration of contrast through the peripheral orifice of the loop-like sigmoidostomy and successful imaging both of the anastomosis and of the diameter of the peripheral colonic segment.

Patient was initiated in a follow-up on a 6-month basis. Two years later, neither complications nor concomitant congenital abnormalities have been detected.

Case 2

A one-day old female newborn (40 weeks of gestation, birth weight 3220 grams and vaginal delivery) was referred to our Department 16 hours after birth, due to two episodes of bilious vomiting and gradually deteriorating flatulence.

In the context of the prenatal ultrasound examination conducted in the 27th week of gestation, the presence of a dilated intestinal convolution, 12.3 mm in diameter, in the right lower quadrant of the peritoneal cavity was documented (Fig. 6).

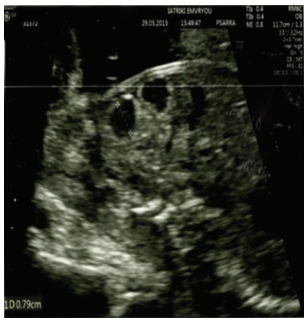

Fig. 6. Prenatal ultrasound examination conducted in the 27th week of gestation with the presence of a dilated intestinal convolution, 12.3 mm in diameter, in the right lower quadrant of the peritoneal cavity.

During the initial assessment of the patient, the main findings were: sparse metallic bowel sounds and flatulence. After digital rectal examination we documented the absence of an obvious anorectal anomaly, while there was mucoid material without any bilious admixture in the ampulla.

Alongside with the diagnostic approach, we started supporting the patient with intravenous crystalloid solutions (R/L), while we also administered intravenously cefuroxime 80 mg/kg in 3 daily doses and metronidazole 7.5 mg/kg in 3 daily doses. Performance of nasogastric decompression through an 8 Fr nasogastric tube led to the aspiration of 12 ml of gastric content with bilious impurity.

Erect abdominal radiograph revealed accumulation of gas resulting in dilation of the central part of the gastrointestinal tract, while there was absence of gas in the peripheral part (Fig. 7).

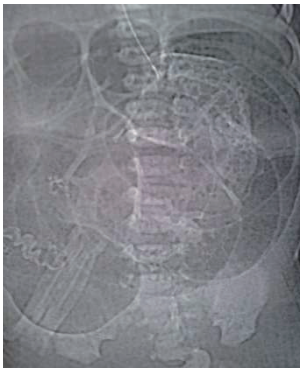

Fig. 7. Gas accumulation in the central part of GI tract, absence of gas in the lesser pelvis.

Performance of contrast enema examination by using ultravist followed. It documented the presence of microcolon, by imaging only the left colon, which was not found in the left abdomen, but shifted to the mean line (Fig. 8).

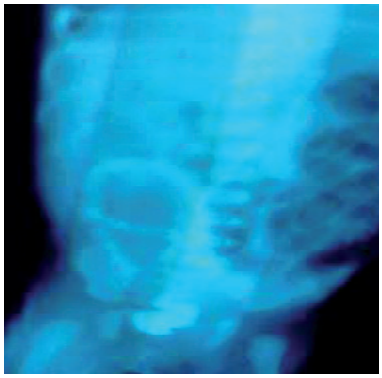

Fig. 8. Contrast enema examination with ultravist documented the presence of microcolon.

The above findings were indicative of complete intestinal obstruction. Thus, we conducted urgent exploratory laparotomy, through a broad Mc Burney's incision. The presence of a colonic atresia type III in the ascending colon was revealed (Fig. 9).

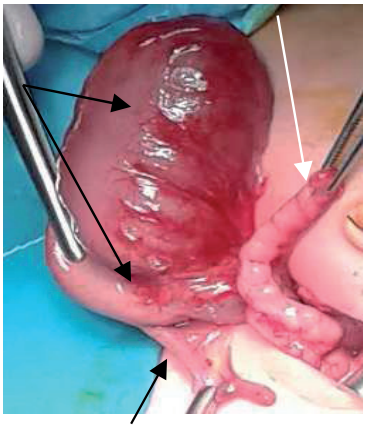

Fig. 9. Colonic atresia type III. Note: the dilated cecum (black arrow), the ascending colon (black arrow), the appendix (black arrow) and the microcolon (white arrow).

Diameter of the distal segment of the colon (microcolon) was 0.8 cm, while the diameter of the proximal segment was 4 cm (diameter ratio = 5). We decided the restoration of the intestinal continuity conducting a broad side-to-side anastomosis in 2 layers by using PDS 5/0, without protective stoma (Fig. 10, 11).

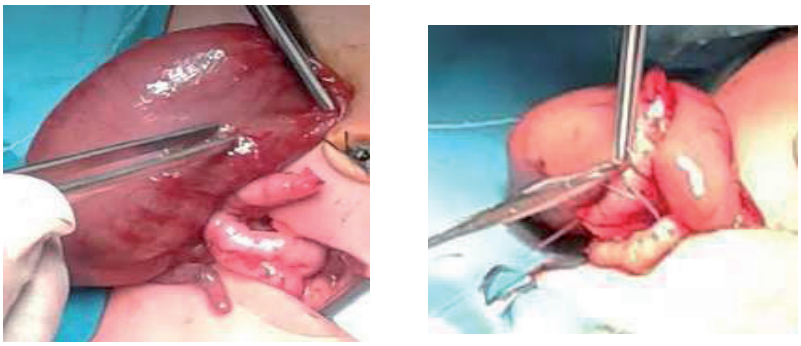

Fig. 10, 11. Restoration of intestinal continuity after conduction of a broad side-to-side anastomosis in 2 layers.

We promoted a thin catheter through the distal colonic segment and performed wash-out with normal saline. Full thickness biopsy was taken from the margins of the distal segment, in order to exclude the presence of colonic aganglionosis. Finally, we performed appendicectomy.

Patient remained in an incubator for two days, while nutritional support began the 2nd post-operative day by administration of total parenteral nutrition. On the 4th post-operative day the patient started to pass stool and on the 5th we decided to stop parenteral nutrition and to initiate oral feeding. Patient was discharged home on the 7th post-operative day in excellent general condition.

The diagnostic approach was completed two weeks later, after conduction of examination by a pediatric cardiologist and ultrasound examination of the spine and the urinary tract. No concomitant congenital anomalies or other pathological signs were found. Patient was initiated in a follow-up on a 6-month basis. Eighteen months later, neither complications nor concomitant congenital abnormalities have been detected.

Discussion

Prenatal imaging of the dilated colon either by ultrasonography or magnetic resonance can contribute to the early diagnosis of atresia, thus substantially improving the final prognosis. Colon can be distinguished from the small intestine noticing the haustral markings and its localization peripherally within the peritoneal cavity. Imaging of the colon is not reliable before the 18th week of gestation. However, its imaging is feasible via prenatal ultrasonography after the 25th week of gestation [28]. Anderson *et al.* [29] were the first that described a case of colonic atresia, which was prenatally diagnosed via ultrasonography, conducted in the 34th week of gestation. They included in differential diagnosis megacolon, rectal atresia and meconium disease. Parulekar [28]

also estimates that documentation of dilated colon during the prenatal period is essential for the early diagnosis, at an asymptomatic stage. Correlation between colon's diameter and gestational age is shown in Table 2, below.

Table 2. Correlation between gestational age and colon's diameter.

Gestational age (weeks)	Colon's diameter (mm)
15–20	3.6
20–25	4.4
25–34	8
30–35	11.4
35–40	16.8
>40	18.7

In our second case, colonic dilation was verified on 27th week of gestation. Colon's diameter was 12.3 mm. Based on data exposed in table 2, it is clear that it was an abnormal finding, which we utilized in order to make the diagnosis of colonic atresia within the first 24 hours after birth.

The initial imaging study conducted in a newborn with worsening flatulence is erect or supine abdominal radiograph. It should be noted that colonic dilation can be mistaken as ground glass appearance or pneumoperitoneum [30]. Both the abundant intestinal gas and the lack of gas peripherally are essential for the diagnosis. In our first case, erect abdominal radiograph documented the presence of contrast not only in the rectum but also in the lesser pelvis, due to the accidental sigmoid's rupture. Thus, it contributed substantially to the prompt diagnosis of sigmoid atresia complicated with rupture. There are only two similar published case reports in the literature, in which accidental rupture distally of the atretic colonic segment is described: Etensel *et al.* (6] describe one case in the context of contrast enema conduction, leading to pneumoperitoneum and Boles Jr *et al.* [31] describe another one, after contrast enema conduction, as in our first case.

Key points for the diagnosis of colonic atresia are: highlighting of the microcolon during the contrast enema conduction and the abrupt interruption of colonic imaging (abrupt halt or hook sign) [31, 32]. Microcolon distally of the atretic, thus occluded colonic segment is attributed to the fact, that meconium does not pass through the colon during the intra-uterine life. It is obvious that meconium passage is substantial for colonic development. Pasto *et al.* [33] propose the conduction of abdominal ultrasonography after completion of contrast enema examination. Through this approach, they try to estimate if the level of obstruction is the same in the two imaging methods. If the obstruction documented in the ultrasonography corresponds to that highlighted in the contrast enema examination, then the possibility of multiple atresia existence is limited.

Diagnosis of colonic atresia is indicative for urgent surgical intervention, which we performed in our two cases. Delay in the surgical intervention dramatically aggravates the final prognosis. This is due to the fact that between the ileocecal valve and the occluded colon progressively develops a blind loop, leading to the rupture of the dilated colon [30]. If therapeutic intervention is made timely, then mortality in cases of pure colonic atresia does not exceed 10% [19]. If intervention is delayed for 72 hours or even worse for 96 hours or more, then the mortality rate dramatically exceeds up to 60 and 100% respectively [34, 35]. In a retrospective study conducted by Etensel *et al.* [6] in newborns with colonic atresia survival rate was 74.3%. In that cases in which therapeutic intervention was made within the first 72 hours 80% of the suffering newborns survived ($p < 0.004$), while from the 24 newborns treated after the first 72 hours, only 13 survived (54.17%).

Based on the classical therapeutic approach it is recommended that, if atresia is located in the ascending colon, then the restoration of intestinal continuity in a time is the optimal choice, while, if atresia is located distally to the splenic flexure, then the suggested treatment option is the performance of colostomy centrally to the occluded colonic segment and restoration of intestinal continuity at second time [20, 27, 36, 37]. In their retrospective study Etensel *et al.* [6] refer to 190 cases of rectal atresia that they encountered, essentially describing the three operative tactics, that pediatric surgeon should follow, when facing a case of colonic atresia. In 132/190 cases they firstly performed colostomy centrally to the occluded segment, in 3/190 cases they performed a central colostomy, but they also restored intestinal continuity at the same time and in 45/190 cases they conducted anastomosis of the central and distal segment, after excision of their ends, but without tapering of the central segment. In our first case, we primarily restored the atresia, as described by Etensel *et al.* [6] in 3/190 of their cases, with performance of a central colostomy, because, on the one hand it was a case of peripheral atresia and on the other hand, at the time of diagnosis we also encountered development of peritonitis after accidental colonic rupture [25].

The concept of restoration of intestinal continuity in patients with colonic atresia in a time began to develop over the last 30 years [38, 39]. In the context of this strategy, it seems difficult to create a secure anastomosis, when the two segments differ substantially in dimensions. This constitutes an essential disadvantage, since the plication of the dilated lumen is not enough, thus enterectomy is needed, leading to anatomic loss of intestine and especially of the ileocecal valve [25]. Cox *et al.* [34] consider as safe the conduction of an anastomosis, in those cases when diameter ratio of the two segments, central and distal, does not exceed 3/1. In our second case, in which we restored intestinal continuity in a time, diameter ratio of the two segments was 5 (4/0.8). Besides, Hsu *et al.* [1] successfully attempted restoration of intestinal continuity at a second time, with prior performance of a colostomy, with the diameter

ratio of the two segments to be 5. However, it should be noted, that before performing an anastomosis, it is substantial to check the lumen's patency of the distal part of the colon, as we did in our second case [38, 40].

Pohlson *et al.* [38] and Sturim *et al.* [41] suggest the excision of the ends of the two segments, in order to create a secure anastomosis, as they do not feature normal innervation and vasculature. Although we did not apply this proposal, we did not encounter any complication, mainly shunt from the anastomosis or its dysfunction, especially in our second case.

In our opinion, the selection of the indicated operative strategy should be made upon the general condition of the newborn and the safety of the selected approach, in order to minimize the possible development of post-operative complications.

It is undeniably important to exclude megacolon from differential diagnosis, a conventional practice that we also applied. There are less than 25 published reports of colonic atresia with concomitant megacolon [19, 24]. Marjorie J Arca *et al.* [42] estimate that this falls into 2% of all cases of colonic atresia. They consider this co-existence as rare (incidence 1/106 live born neonates).

In conclusion, we believe that the major objectives of a pediatric surgeon when encountering a case of colonic atresia should be: early diagnosis, patient's stabilization, selection of the appropriate operative strategy and early recognition and treatment of potential post-operative complications.

Conflict of interest

None of the contributing authors have any conflict of interest, including specific financial interests or relationships and affiliations relevant to the subject matter or materials discussed in the manuscript.

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