Case Report

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Long-Term Complications of Neonatal Necrotizing Enterocolitis: Twisting of Stenosed Small Intestine

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ANNOTATION Neonatal necrotizing enterocolitis (NEC) is a nonspecific inflammatory disease of unknown etiology with multifactorial pathogenesis, which development is explained as a result of hypoperfusion of the immature intestinal mucosa of a newborn who has undergone perinatal hypoxia and, as a result, a change in blood flow in the mesenteral vascular system. This disease more often affects the intestinal wall of premature babies. In typical cases, mucosal necrosis develops in the terminal iliac and right parts of the colon, and when the process progresses, it can spread to the entire thickness of the intestinal wall, causing its perforation, so peritonitis is a frequent complication of NEC. But in addition, distant complications of NEC are distinguished, which include the development of intestinal obstruction in view of a decrease in the contractility of the intestinal wall section due to its fibrosis and, as a result, narrowing of the lumen.

We report the clinical case of surgical treatment of the twisting of stenosed portion of the ileum in a child who underwent NEC in the early neonatal period. A mother sought assistance with a child of 3 months at the Reginal Children's Hospital (born at 28 weeks of gestation). After birth, there was a violation of the absorption of enteral nutrition (periodic posseting, bloating), blood in the stool was determined. On the 14th day of life a pediatric surgeon examined the baby: necrotizing enterocolitis II A. After stabilizing the condition (2 months), the child was transferred from the perinatal center to a pediatric hospital with a diagnosis of bronchopulmonary dysplasia, a new form, a severe course, and a period of exacerbation. At the age of 2.5 months, he was discharged in a satisfactory condition to the outpatient stage. On the 15th day from the moment of discharge (3 months of life), the mother noted the expressed anxiety of the baby, bloating, stool retention. On the 16th day from the moment of discharge during feeding, the child began to suck sluggishly, did not absorb the age norm. He was examined by a pediatrician, the consultation of a pediatric surgeon was recommended. Due to the severity of the condition, the baby was hospitalized.

In the initial examination, no data for intestinal obstruction were detected. The patient received parenteral nutrition, antibacterial therapy. Ultrasound and radiography of abdominal organs were performed daily. On the third day of observation, deterioration is noted (abdomen bloated mainly in the upper parts, soft upon with palpation, the baby was anxious, peristalsis was reduced), with ultrasound: between the loops of the enlarged intestine, an echogenic band of up to 18 mm (adhesion?) was determined, there was the liquid component between the loops. A laparotomy was performed, during the revision there was a displacement of the large intestine into the left parts of the abdominal cavity. Ten cm from the ileocecal angle, a section of the ileum with a length of up to 15 cm was found, which was like a "double trunk" wrapped at the base around its axis around a cord-like adhesive stretching to the posterior abdominal wall. Visually, the loop was dark bard in color, its walls were swollen, infiltrated, and their cartilaginous density was determined upon palpation in the contact area of the walls of the intestine. The resection of this loop was performed, end-to-end anastomosis was formed according to the method of J. Louw. After the operation, the baby received treatment in the intensive care unit, enteral feeding on day 5, on day 9, after expanding the volume of feeding, he was transferred to the department of pediatric surgery. Discharged in satisfactory condition on the 12th day after surgery.

Keywords: necrotizing enterocolitis in newborns, intestinal stenosis, resection, intestinal obstruction, anastomosis

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BPD - bronchopulmonary dysplasia
CNS - central nervous system
GI-tract - gastrointestinal tract
IUI - intrauterine infection
NEC - necrotizing enterocolitis
RCCG - Regional Children's Clinical Hospital
RF - respiratory failure
RICU - resuscitation and intensive care unit

INTRODUCTION

Necrotizing enterocolitis (NEC) is a disease of the gastrointestinal (GI) tract of preterm neonates that results in inflammation and bacterial invasion of the intestinal wall. Despite advances in the care of preterm neonates, NEC remains one of the leading causes of morbidity and mortality in this cohort of patients, occurring in 1–5% of all neonates admitted to intensive care and in 5–10% of very low birth weight (<1.5 kg) [1, 2].

In a sense, NEC is the result of advances in neonatology that have made nursing neonates with low gestational age possible. NEC is one of the most devastating diseases occurring in neonatal intensive care [3].

Historically, NEC has been thought to be the consequence of damage to the immature GI tract by various factors. However, the etiology of such a multifactorial process remains unclear so far. This causes a variety of clinical manifestations, including an increase in apnea episodes, bradycardia, lethargy, and temperature instability. Symptoms specific to the gastrointestinal tract may also occur, such as impaired absorption of enteral feeding, vomiting, bloody stools, bloating and tenderness of the abdomen, as well as discoloration of the abdominal wall (swelling in case of peritonitis) [4, 5].

Laboratory findings may indicate the presence of an infectious agent, a bleeding disorder, and fluid retention. And radiographic signs may reflect a picture of intestinal obstruction (dilated or fixed intestinal loops), hollow organ perforation syndrome (gas in the intestinal wall or free gas in the abdominal cavity) [6].

Medical treatment usually includes intestinal rest and decompression, antibiotic therapy, and syndromic treatment of other hematological or electrolyte imbalances. As a rule, such children require respiratory support [7]. However, about 20–40% of neonates with NEC will require surgical intervention [8].

Indications and timing of surgical treatment are still the subject of discussion [9]. The volume of surgical treatment in such patients is also widely discussed: peritoneal drainage in unstable patients (when a full-fledged surgical intervention may threaten the life of a child), laparotomy and drainage without removal of non-viable tissues, or resection of damaged intestinal sections with the formation of an intestinal stoma or primary anastomoses [10]. Despite high achievements in neonatology and neonatal surgery, there are reports of high mortality rates (about 50%) among patients with severe NEC who underwent surgery [11].

NEC has a significant number of severe complications, such as abdominal adhesions, cholestasis, short bowel syndrome, neurodevelopmental delay, etc.

Frequent long-term complications of NEC requiring surgical intervention include stenosis of the affected area of the intestinal tube, which is associated with fibrosis of the intestinal wall after ischemia and inflammation [12]. The severity of clinical manifestations in the separated period in children with a history of NEC depends on the severity and course of the primary disease. The manifestation of surgical complications, such as intestinal obstruction (due to the presence of a mechanical obstacle - narrowing of the intestinal lumen, the presence of adhesive adhesions), peritonitis (as a manifestation of decompensation of intestinal obstruction) is possible months and even years after the conditional recovery of such patients, which requires their careful outpatient monitoring [13, 14]. In addition, it is necessary to take into account the possibility of a combination of NEC and anomalies in the development of the gastrointestinal tract (the presence of a common mesentery of the small and large intestines, pronounced embryonic adhesions, high standing of the dome of the caecum, etc.), which causes difficulties in diagnosing and determining the tactics of treating such patients.

The aim of this study is to update the issue of the complexity of diagnosis and tactics of managing patients with this pathology by demonstrating the clinical observation of long-term complications of neonatal necrotizing enterocolitis (twisted stenosed portion of the small intestine).

Clinical observation

A 3-month-old baby (boy), from the first preterm birth, the second pregnancy at the 28th week of gestation, which occurred against the background of intrauterine infection (IUI), chorioamnionitis, chronic fetoplacental insufficiency; there was also chronic intrauterine fetal hypoxia. The amniotic fluid is light. Birth weight 1,300 g, height 38 cm, head circumference 25 cm, chest circumference 24 cm, APGAR score 5/6/6. Resuscitation in the delivery room: radiant heat, artificial lung ventilation, Curosurf (200 mg/kg).

Due to the severity of the condition after birth, the child received treatment at the Regional Perinatal Center. In the neonatal period, there was a violation of the absorption of enteral nutrition (periodic regurgitation, bloating), the presence of blood in the stool. On the 14th day of life he was consulted by a pediatric surgeon, the diagnosis was necrotizing enterocolitis, 2A st. IUI.

Upon stabilization of the condition (at the age of 2 months), the baby was transferred to the pulmonology department of the Regional Children's Clinical Hospital (RCCH), where he stayed for two weeks with a diagnosis of bronchopulmonary dysplasia, a new form, severe course, a period of exacerbation. Respiratory failure (RF) I–II. Consequences of perinatal hypoxic-ischemic lesions of the central nervous system, recovery period: mild movement disorder syndrome, hyperexcitability. Functional disorders of the gastrointestinal tract: infantile colic. Right-sided inguinal-scrotal hernia. Early anemia of prematurity. In a satisfactory condition, he was discharged to the outpatient stage, under the supervision of a district pediatrician at the place of residence.

After 2 weeks from the date of discharge, the mother notes that he has severe anxiety, bloating, and stool retention during the day. The baby spent the night restlessly. The next day he became lethargic, did not ate the age norm. During the day the boy began to refuse food. There were no vomiting or fever. He was examined by the district pediatrician, a consultation of the pediatric surgeon of the CSTO was recommended, where the baby was hospitalized to the RICU.

Upon admission, the general condition of the baby was severe, due to a violation of the motor-evacuation function of the GI tract, intestinal paresis against the background of bronchopulmonary dysplasia (BPD). The baby was conscious, and reacted to examination with increased motor activity. The scream was loud. The muscle tone was unevenly reduced. There were no convulsions. The skin was clean, pale pink. Breathing was independent, without respiratory support, auscultatory breathing was weakened, heard evenly on both sides, no wheezing. Heart sounds were muffled, the rhythm was correct. The abdomen was evenly swollen, soft, the baby worried during palpation. The peristalsis was reduced. There was no edema, hyperemia of the anterior abdominal wall. The liver was at the edge of the costal arch, the spleen was not enlarged. In the inguinal regions, protrusions were determined, of a soft elastic consistency, reducible into the abdominal cavity, without signs of infringement at the time of examination. The genital organs are formed correctly according to the male type, both testicles are in the scrotum. There was mucus in the stool after enema.

Upon admission, the following instrumental studies were performed:

- neurosonography: signs of hypoxia against the background of immaturity, initial dilatation of the external fluid spaces;
- echocardiography: open oval window — 2.5 mm;
- according to the ultrasound examination of the abdominal organs (ultrasound of the OBP): echo signs of paresis of the gastrointestinal tract, flatulence is significant;
- fibroesophagogastroduodenoscopy: proximal erosive hemorrhagic gastritis, polyp of the middle third of the esophagus.
- frontal X-ray of the abdominal organs in a vertical position: increased pneumatization of intestinal loops, no levels of liquid or free gas were revealed (Fig. 1A).

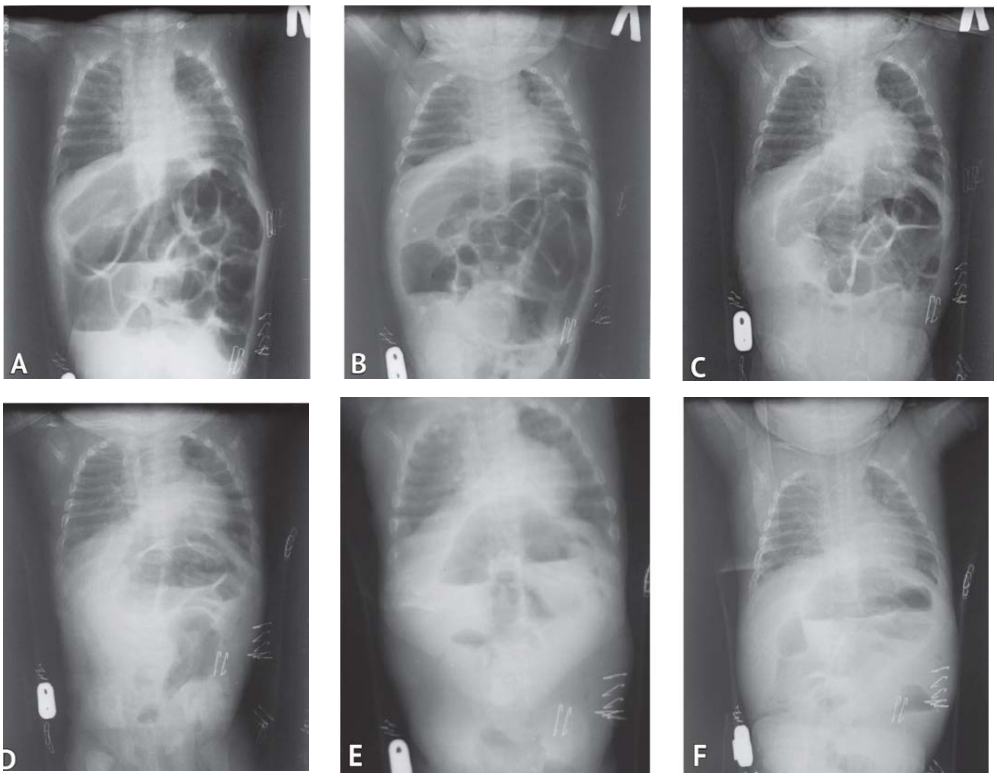

Fig. 1. Plain X-ray scan of the abdominal cavity and chest over time: A — upon admission; B - 14 hours after hospitalization; C - 20 hours after hospitalization; D - 36 hours after hospitalization; E - 48 hours after hospitalization; F-64 hours after hospitalization

Preliminary diagnosis: "Partial intestinal obstruction? Bronchopulmonary dysplasia, new form, severe course, period of exacerbation. RF II–I. Consequences of perinatal hypoxic-ischemic lesions of the central nervous system (CNS), recovery period: mild movement disorder syndrome, hyperexcitability. Bilateral inguinal-scrotal hernia. Early anemia of prematurity.

Given the child's condition and a history of NEC, enteral feeding was canceled, he was switched to parenteral nutrition, taking into account physiological needs, a nasogastric tube was installed. Treatment: Maxipime, Metrogyl; stimulation of intestinal motility: Drotoverin, Metoclopromide. According to the data of instrumental methods of examination, it was decided to refrain from active surgical tactics. The child was discussed daily at medical conferences; in dynamics, frontal X-ray examination of abdominal organs was performed in a vertical position (Fig. 1):

– 14 and 20 hours after admission (Fig. *1B, C*): loops of the small intestine are filled with a large amount of gas, displaced to the left sections of the abdominal cavity;

– 36 hours after admission (Fig. 1D): positive *R*-changes are noted, such as a decrease in gas filling of the small intestine, single swollen loops are determined in the left half of the abdominal cavity (upper compartment and left iliac region); nasogastric tube in the projection of the stomach; free gas and liquid levels are not determined.

– 48 hours after admission (Fig. 1E): no free gas was detected; in the upper compartment of the abdominal cavity, intestinal loops inflated with air are determined with the formation of arches with horizontal fluid levels and single horizontal fluid levels; pneumatization of the lower sections in dynamics decreased.

Within two days of observation, the clinical dynamics of the condition was not observed, according to the X-ray data there were slight positive changes. Scanty discharge via the probe. However, on the 3rd day of observation, the child's condition worsened, manifested by restless behavior, a painful cry during examination and palpation of the abdomen, and an increase in the amount of gastric discharge through a nasogastric tube. Frontal abdominal X-ray in a vertical position (64 hours after admission, Fig. 1F): taking into account X-ray data the day before, there were radiographic signs of partial intestinal obstruction.

Transabdominal ultrasound of abdominal organs: the liver is not significantly enlarged, its parenchyma is homogeneous, echogenicity is slightly increased; gallbladder with an inflection, common bile duct is not dilated. The spleen is homogeneous, not enlarged. In the stomach there is scant liquid content. The area of the pylorus and duodenum is shaded by gases. In the right half of the abdomen, the loops are expanded to 30 mm, the contents are full of gas bubbles, echogenic. The walls of the intestinal loops are thickened up to 2.5-3.5 mm, moderately edematous. The motility is episodic, extremely low.

On the left, intestinal loops are predominantly collapsed. The mesentery is moderately infiltrated, the typical vascular ring is not defined. Vessels of the mesentery are moderately plethoric. In the paraumbilical region between the loops of the dilated intestine, an echogenic cord up to 18 mm is determined (embryonic adhesion?). There is a liquid component between the loops (homogeneous effusion?) (Fig. 2).

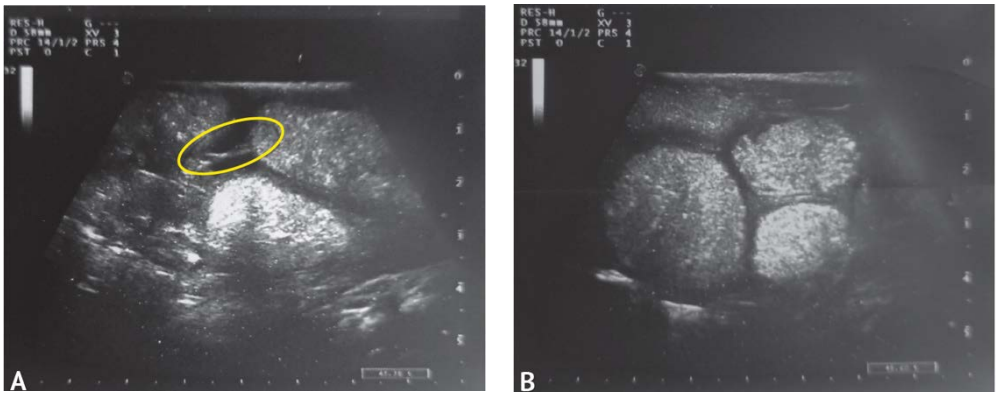

Fig. 2. Transabdominal ultrasound examination: A - an echogenic band was identified between the loops of the small intestine (cord-like adhesion); B - dilated loops of the small intestine (transverse scan)

The medical council made a decision on the need for laparotomy, revision of the abdominal organs with the determination of further treatment tactics, taking into account the intraoperative picture.

Under endotracheal anesthesia right transverse laparotomy was performed. After opening the abdominal cavity, about 5 ml of a transparent effusion was released. During the revision of the abdominal cavity, the displacement of the colon and small intestine to the left was noted, their common mesentery was found. During the revision of the upper compartment of the abdominal cavity, a significant number of dense adhesive adhesions in the area of the duodenum, deforming its wall, were noted. Loops of the large intestine and small intestine, collapsed almost along its entire length. At a distance of 10 cm from the ileocecal angle, a section of the ileum up to 15 cm long was found, which is a "double-barreled" wrapped at the base around a cord-like adhesion extending to the posterior abdominal wall.

The walls of the ileum in this area are tightly fixed to each other by adhesions. Visually, the loop is maroon in color, its walls are edematous, infiltrated, their cartilaginous density is determined by palpation in the zone of contact of the intestinal walls (Fig. 3). When checking for the patency of this area by squeezing air and contents from the proximal sections, partial patency is seen, the contents slowly seep into the cupula of the caecum, the distal sections are slightly straightened by air.

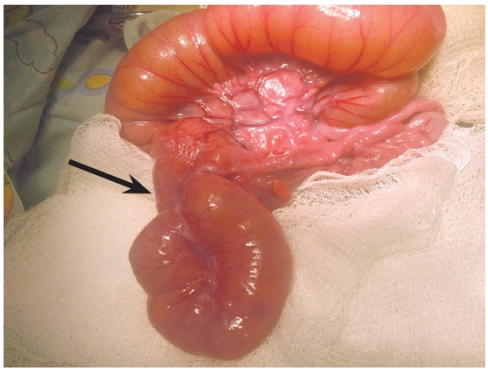

Fig. 3. Intraoperative picture: twisted stenosed portion of the small intestine, the arrow indicates the torsion area around the cord-like adhesion

The lumen is not determined in the distal section upon palpation, the walls of cartilaginous density. Proximal to the specified area, the diameter of the loops of the afferent intestine is 5 cm, distally - 2 cm. The sections of the intestinal tube proximal and distal to the "double-barrel" are viable: low peristalsis is observed, the vessels pulsate, the color of the wall is not changed.

Further revision revealed a vermiform appendix up to 5 cm long, up to 8 mm in diameter, located down to the small pelvis, injected with blood vessels, edematous, adjacent to the "double-barreled" portion. An antegrade appendectomy was performed. Taking into account the nature and severity of the adhesive process, changes in the intestinal wall in the area of the "double-barrel" (when trying to separate the intestinal walls, the risk of perforation is high), a decision was made to perform resection of this area. The distal and proximal sections of the ileum were adapted in diameter, a two-row small-intestinal end-to-end anastomosis according to *J. Louw* was formed.

Clinical diagnosis after surgery: "Condition after NEC 2A. Stenosis of the ileum. Low intestinal obstruction. Incomplete bowel rotation. Common mesentery of the small and large intestine. Adhesions of the abdominal cavity. Secondary appendicitis. Serous peritonitis. Bronchopulmonary dysplasia, new form, severe course, period of exacerbation. RF II–I. Consequences of perinatal hypoxic-ischemic damage to the central nervous system, recovery period: mild movement disorder syndrome, hyperexcitability. Bilateral inguinal-scrotal hernia. Early anemia of prematurity. Acute proximal erosive hemorrhagic gastritis. Polyp of the middle third of the esophagus. The diagnosis was confirmed histologically — the wall of the resected ileum showed signs of chronic ileitis with moderate subepithelial fibrosis, as well as phlegmonous appendicitis.

Due to the severity of the underlying disease and the surgical intervention to continue intensive care, the baby was transferred to the RICU. The enteral feeding was initiated on the 4th day after the operation, the feeding age norm was gained on the 9th day, then the patient was transferred to the department of pediatric surgery. The baby was discharged in a satisfactory condition on the 12th day after the operation under the supervision of a pediatrician and pediatric surgeon at the place of residence. At the repeated examination in a year, there were no complaints from the mother of the child. The boy is gaining weight, neuropsychic and physical development corresponds to his age.

DISCUSSION

NEC is a nonspecific inflammatory disease of unknown etiology with multifactorial pathogenesis, which development is explained as a result of hypoperfusion of the immature intestinal mucosa of a newborn who has undergone perinatal hypoxia and, as a result, a change in blood flow in the mesenteric vascular system [15].

This disease often affects the intestinal wall of premature neonates. Until 1964, this pathology was not identified as a separate nosological form and existed under a wide variety of diagnoses: functional intestinal obstruction, intra-abdominal abscess, spontaneous perforation of the ileum, necrotizing colitis of newborns with perforation, ischemic enterocolitis, neonatal intestinal infarction, appendicitis [10]. The current term "necrotizing enterocolitis" was suggested by *H. Rossier* and *S. Schmid*, in 1959.

In typical cases, mucosal necrosis develops in the terminal ileum and right colon, and as the process progresses, it can spread to the entire thickness of the intestinal wall, causing its perforation; therefore, peritonitis is a common complication of NEC [3]. But, in addition, there are long-term complications of NEC, which include the development of intestinal obstruction due to a decrease in the contractility of the intestinal wall due to its fibrosis and, as a result, narrowing of the lumen [6].

The diagnosis of this condition is a very time-consuming process, which is often associated with an unclear clinical picture, characterized by violation of the absorption of enteral feeding (bloating, profuse regurgitation). This can mislead the child's parents in the presence of a severe comorbidity (in this case, BPD), and cause a late visit to a pediatric surgeon. In addition, this malnutrition can characterize the presence of congenital malformations of the gastrointestinal tract in a child (semimembranes, enterocystoma, doubling of the intestinal tube, etc.), manifesting a clinic of partial intestinal obstruction. Therefore, an important stage in the examination of such clinically "unclear" patients is a thorough history taking (the presence of NEC in the neonatal period), dynamic observation, stage radiographs and transabdominal ultrasound. This will allow you to determine as accurately as possible the further tactics of managing the patient and avoid such severe complications of this condition (stenosis), as perforation of the intestinal wall and peritonitis.

CONCLUSIONS

Thus, the clinical observation presented by us clearly characterizes the complexity of diagnosing such a formidable complication of necrotizing enterocolitis as stenosis of a section of the small intestine, which led to the development of a more serious twisting of the stenosed portion. This results in intestinal obstruction. Regurgitation, lethargy, bloating are manifestations of a wide range of pathological conditions that manifest themselves in a similar way (both somatic and surgical) in children of a younger age group. This work emphasizes the need for an in-depth study of long-term complications of necrotizing enterocolitis, namely clinical manifestations and development of management tactics for such patients.

The continuity between institutions providing care to patients with necrotizing enterocolitis at various stages of development is an important step in the observation of such patients as well.

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