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Research Article

Experience in Timely Diagnosis and Surgical Treatment in Case of Threat of Spontaneous Perforation of the Ileum in Children with Extremely Low Birth Weight

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Abstract

The aim of the study was to study the results of treatment of immature patients with extremely low birth weight in cases of development of the pseudo-obstruction of the ileum before spontaneous perforation based on the developed algorithm of radiation control and the choice of rational surgical tactics.

During the 4-year chronological period of the study (2014-2017), our own practical data on the observation and treatment of a cohort of deeply premature neonates (n = 12), similar in clinical manifestations and radiologic features, operative and histological findings, were blindly retrospectively analyzed. Morphological examination indicates muscular-neural dysplasia of the ileum in the places of the highest risk of perforation. Timely diagnosis of the condition preceding the spontaneous perforation of the ileum and rational surgery made it possible to avoid a catastrophe in the abdominal cavity and create favorable conditions for enteral feeding.

Keywords: *A Pseudo-Obstruction of the Ileum; Spontaneous Intestinal Perforation; Muscular-Neural Dysplasia; Opportunities for Enteral Nutrition; Extremely Immature Infants*

Introduction

Unlike full-term babies in the adaptation period, in extremely immature babies, due to the structural and functional immaturity of the basic systems of the body, adaptive mechanisms cannot be activated after birth. Morphofunctional immaturity, which prevents full adaptation in babies with extremely low birth weight (ELBW), can be a trigger in the development of life-threatening conditions.

With the beginning of enteral feeding, stretching of smooth muscles and irritation of baroreceptors in the walls of the hollow organs of the digestive tract lead to reflex impaired motor activity. With a forced expansion of the volume of feedings, along with the inability to contract during overstretching, the intestinal wall experiences vascular "low density", leading to local ischemia.

On the contrary, the rhythmic introduction of minimal volumes of milk can naturally (physiologically) stimulate peristalsis and, according to researchers, reduces the risk of sepsis in infants in the absence of food tolerance [1]. Gastrointestinal insufficiency in infants ELBW requires special attention and careful correction, which is especially important for sepsis and intraventricular hemorrhage.

The clinical manifestations of digestive disorders are similar to the initial symptoms of urgent surgical conditions. The key to the algorithm for diagnosing a surgical disease is a decrease in food tolerance: the contents are evacuated from the stomach, bloating and stretching ("glossiness") of the abdomen later appear, there may be a lack of stool and gas.

To overcome violations of the gastrointestinal tract, conservative measures that are effective in most cases (from 36 - 39 to 94%) are used in the world: decompression of the stomach and the enteral administration of acetylcysteine, cleansing, gastrographin and glycerin instillations of the colon [2]. Therapy is carried out only in the absence of "precursors" of disaster in the abdominal cavity.

The most common cause of an acute process in the abdominal cavity is perforation of the hollow organ, which can occur unobtrusively, but the prognosis for an extremely immature patient is significantly worsened.

According to the results of studies, in patients with ELBW, intestinal perforations are most often caused by necrotizing enterocolitis (NEC) in 53.4% of cases, less often - congenital or acquired intestinal obstruction - in 27.2%, and spontaneous intestinal perforation (SIP) in 19.4%. Lesions of the small intestine are found in 68.9%, of which the ileum suffers in 47.6%, jejunum perforations are more than 2 times less frequent (21.4%). Perforation of other parts of the intestinal tube is much less common [3].

Verification of the diagnosis of intestinal perforation is carried out by radiation research methods, but radiologically during perforations, "explicit" pneumoperitoneum is detected only in 62% of cases [4].

Spontaneous intestinal perforation (SIP) often simulates necrotizing enterocolitis (NEC); therefore, differential diagnosis between these conditions is mandatory [5].

The characteristic radiation signs of NEC ("static loop", intestinal pneumatosis, infiltrates in the abdominal cavity and the appearance of gas in the portal vein system) with the X-ray picture of pneumoperitoneum can be "veiled".

At the same time, laboratory tests may indicate an infectious and inflammatory process in case of SIP, if this happened against the background of bacterial sepsis.

In numerous studies, starting in 1969, it is indicated that spontaneous perforation of the intestine occurs due to congenital or acquired deficiency of the muscle layers of the intestinal wall [6].

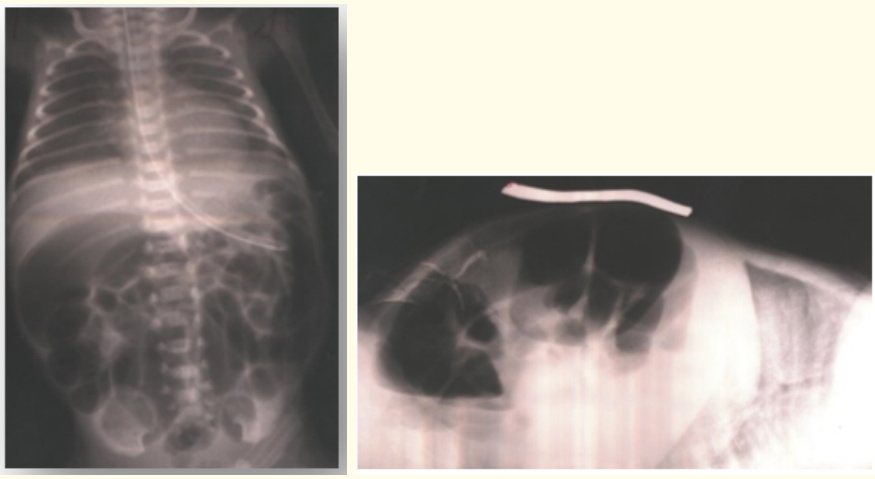
According to the timing of occurrence, two forms of spontaneous perforation of the intestine are distinguished: "early" - up to 72 hours of life, and "late" - later than 72 hours, but most often between 7 and 10 days of life [7]. The origin of the "early" form is explained by congenital muscle deficiency due to chronic fetoplacental insufficiency (fetal hypoxia) and infection by the mother [6].

While the "late" form may be the result of the implementation of postnatal risk factors, including the use of glucocorticoid and indomethacin therapy [8,9].

A correlation was established between SIP with extreme immaturity, with fungal infection and sepsis caused by coagulase-negative staphylococcus; SIP often occurs in children with intraventricular hemorrhage, but the causes and pathogenesis of SIP have not yet been fully elucidated [9].

For timely diagnosis of a predisposition to SIP in patients with ELBW, it is advisable to consider all children of this group as threatened by the development of a surgical disease.

Using the simple radiological method of research makes it possible to diagnose a condition preceding spontaneous ileal perforation. The identification of pathognomonic X-ray signs in patients at risk with known predisposing factors with the correct interpretation of the "earliest" clinical signs helps to make a decision about the operation (Figure 1a and 1b).



(a) *(b)*

Figure 1a and 1b: *On the chest and abdominal radiographs, in the direct and lateral positions: the distal ileum sharply widened in comparison with the proximal loops of the jejunum (compiled by the author).*

Photographs of patient M, 26 weeks + 6 days are presented, in which on the 2nd day of life the presented x-ray picture is revealed, in a short time prepared for surgery; a surgical aid before the development of spontaneous ileum perforation was performed for the first time and was the beginning in this series of cases.

Materials and Methods

Over the 4-year period of 2014 - 2017, we observed 12 children, 8 girls and 4 boys born prematurely at gestational periods of 23 to 28 weeks + 5 days, with a body weight of 550 g to 1150 g and severe concomitant pathologies, of which the most common: respiratory distress syndrome (75%; n = 9), sepsis (50%; n = 6), bronchopulmonary dysplasia (41.6%; n = 5), intrauterine pneumonia (25%; n = 5), open ductus arteriosus (16.6%; n = 2). In severe condition, from the moment of birth, tolerance to enteral nutrition was reduced in all patients. Given the scarcity of symptoms that we noted earlier, it was this clinical manifestation that we regarded as the key to the algorithm for diagnosing surgical processes in the abdominal cavity [10].

On 2 - 3 days of life in 75% (n = 9) of patients, on 6 - 7 days of life in 25% (n = 3) of children, X-ray signs of the condition preceding ileal perforation were revealed: striking 2.5 - 3- multiple gas expansion of the diameters of the distal ileal loop in comparison with the diameter of the loops of the jejunum (Figure 2).

All patients were operated on before intestinal perforation: on a laparotomy, they found a cyanotic gas-filled (without chyme or with a meconium meager), sharply enlarged ileal loop with a thinned wall and dotted areas of the most pronounced thinning - preperforation (Figure 3).

Immediately after enterotomy, the intestinal loop “deflated” like a balloon and assumed its usual size and pink color while preserving the thinning of the intestinal wall; no mechanical obstructions to the passage were revealed. Within the visually defined “healthy” tissues, a resection (from 6 to 9 cm long) of the ileum was performed, the borders of which are difficult to determine precisely. The operations were completed by the creation of a single-row T-shaped anastomosis with proximal Santulli ileostomy.

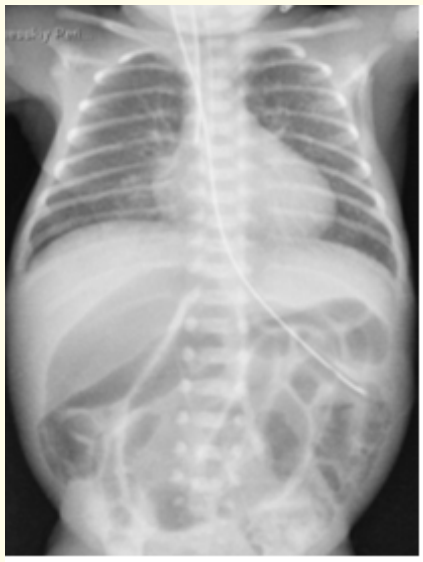


Figure 2: *On the chest and abdominal radiographs, in the direct and lateral positions: the distal ileum are dramatically expanded in comparison with the proximal loops of the jejunum (compiled by the author).*

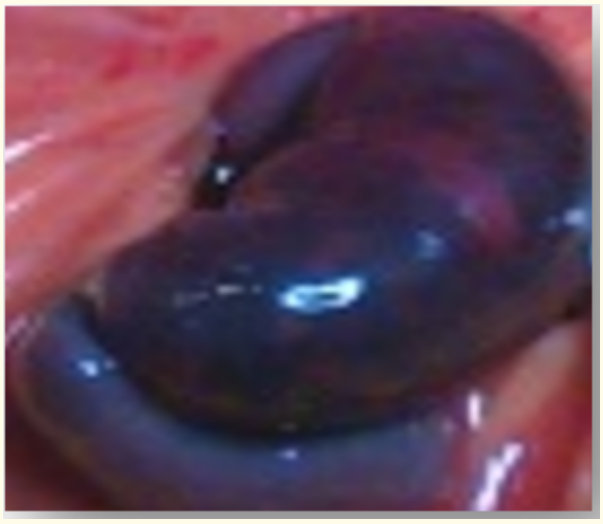


Figure 3: *Intraoperative photos: the ileum loop with a thin wall and sharply widened sections of the most pronounced thinning - the preperforation - is sharply widened (compiled by the author).*

A histopathological examination (Figure 4 and 5) revealed violations of the structure of the muscle layers (longitudinal and circular) in the intestinal wall in the form of hypoplasia and dysplasia with chaotic branching of the dilated thin-walled sinusoidal vessels.

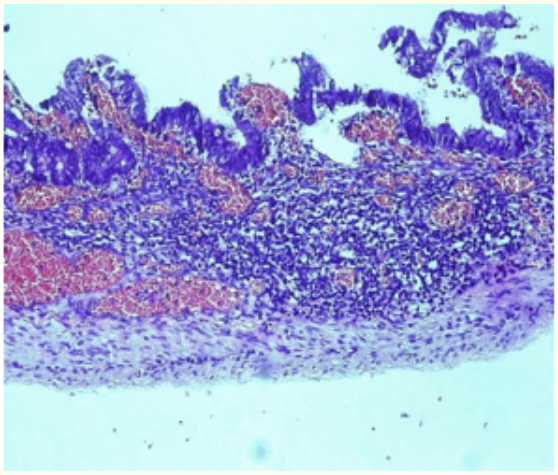


Figure 4: *Microphoto (compiled by the author). The central part of the resected portion of the ileum: the "deficiency" of the muscle layers and hypogangliasis of the ileum (X10; color: hematoxylline-eosin).*

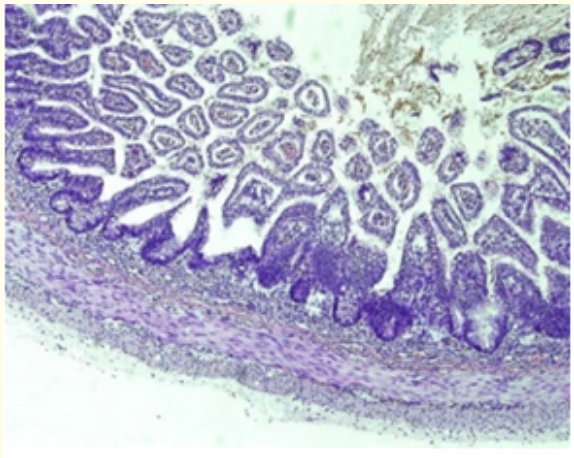


Figure 5: *Microphoto (compiled by the author). The proximal part of the resected portion of the ileum: the "deficiency" of the muscle layers and the zone of hypoglionia of the ileum (X10; color: hematoxylline-eosin).*

Results and Discussion

Given the absence of mechanical obstruction to the passage of food, such clinical and intraoperative findings are interpreted as ileum pseudoobstruction, although the surgeon doubts whether the bowel looks like this before spontaneous perforation. There were no surgical complications. Enteral load was resumed from 7 days after the operation, which was due to the severity of the condition of the patients with the underlying disease. As the T-shaped anastomosis began to work, the distal part of the intestine (colon) was included in

the digestion, as evidenced by the appearance of an independent stool from the anus. Food passage was fully restored in 3 weeks; while the ileostomy function "decreased".

Mortality due to sepsis was 16.6% (n = 2); children died on the 13th and 22nd days after operations - without surgical reasons. Enterostomy closed for surviving patients from 42 to 89 days after the first operation.

At risk for the development of SIP are all patients with ELBW who, taking into account the known predisposing factors (extreme immaturity, previous hypoxia, fungal or staphylococcal lesions of the placenta and the child, the presence of a patient with high fatty acids, antenatal hormone prophylaxis courses, treatment courses), a cautious interpretation is required "Early" clinical manifestations (decreased tolerance to enteral nutrition).

Although our study has limited ability to determine the significance of risk factors for ileal perforation, it should be borne in mind that early onset and rapid expansion in the volume of enteric load and nCPAP can be resolving factors that increase intrathecal pressure, contributing to perforation at the sites of thinning of the muscle layers.

According to the classification developed by Attridge., *et al*. The majority of our patients 75% (n = 9) with conditions predisposing to ileum perforation were detected in the "early" period (2 - 3 days of life), which indicates a probable correlation of this condition with infection transmitted by the mother and chronic placental insufficiency (fetal hypoxia) [7].

It turned out to be possible to diagnose a condition preceding spontaneous ileum perforation using the usual x-ray method of investigation, which facilitates the decision-making on surgery before a catastrophe in the abdominal cavity.

In all our patients, the location of the lesion was the ileum, which is confirmed by radiation, intraoperative and pathomorphological data.

In a situation where perforation has not yet occurred, and the wall of the ileum is clearly thinned, and it is macroscopically difficult to clearly determine the boundaries of the resection, the method of choice was the T-shaped anastomosis with proximal Santulli ileostomy. This solution option relieves the "leading" part of the intestine, which is the prevention of problems after surgery and creates favorable conditions for enteral nutrition and nursing [11].

In all cases, histologically revealed focal deficit (thinning) of the longitudinal and circular muscle layers of the intestinal wall and hypogangliosis in areas of high risk of perforation compared with neighboring normal areas of the intestine.

Thinning of the muscle layers of the intestinal wall is morphologically similar to the histological "picture" described in spontaneous perforation of the intestine. Given the local deficiency of the intramuscular nerve ganglia, it can be assumed that the condition preceding intestinal perforation (pre-SIP) is due to muscular-neural dysplasia and is not associated with necrotizing enterocolitis (NEC) or myopathy.

A certain role in the maturation of the muscular structures of the intestinal wall is played by the process of migration and maturation of the ganglia and plexuses of the enteric nervous system. It is reasonable to assume the secondary maturation of muscle structures in relation to the development of the enteric nervous system.

With the used type of inter-intestinal anastomosis (T-, Y-shaped), the delayed "inclusion" of the distal intestine can be a confirmation of the subsequent "ripening" of the morphological structures of the intestine (including the migration and maturation of the ganglia and plexuses of the enteric nervous system), but this version requires confirmation in multicenter studies.

Findings

1. Immature children with ELBW are threatened by the development of SIP, which implies control of tolerance to enteral nutrition.
2. The use of conventional types of x-ray examination allows you to timely diagnose a condition preceding spontaneous perforation of the ileum in patients at risk, making it easier to decide on surgery before the development of a catastrophe in the abdominal cavity.
3. The morphological characteristic of the ileum at the highest risk of perforation indicates muscular-neuronal dysplasia.
4. Creation of favorable conditions for enteral nutrition in extremely immature patients with ELBW is one of the most important tasks of surgical treatment that outstrips ileal perforation. Performing an intestinal anastomosis (T-, Y-shaped) with Santulli proximal enterostomy prior to perforation is safer, most preferably for preserving the life and nursing of children with ELBW.

Disclosure

The authors of this article confirm the lack of financial or any other support and conflict of interest that would need to be reported.

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