

Giant Meckel's Diverticulum of Thoracic Localization in Combination with Intestinal Malrotation and Cervical Spine Anomaly in an Infant: Diagnostic Challenges and Tactical Errors During the Stages of Surgical Treatment

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Abstract The paper presents literature data and the results of our own observations on the diagnosis and surgical treatment of 47 children with Meckel's diverticulum, who were observed in the clinics of the Department of Hospital Surgery of the Tashkent Medical Pediatric Institute in the period from 2010 to 2022. The frequency, nature of complications, and features of pathological conditions were studied. DM without pathological changes was detected during surgery in 7 (14.9%) observations as an accidental finding, in 40 (85.1%) cases it was accompanied by various complications. A case of thoracic localization of Meckel's diverticulum in combination with intestinal malrotation and anomaly of the cervical spine in an infant is presented; various difficulties and tactical errors were observed at the stages of diagnosis and treatment.

Keywords Meckel's diverticulum, Combined anomalies, Complications, Diagnostics, Treatment

1. Introduction

Meckel's diverticulum (MD) is a developmental anomaly of the omphalomesenteric duct that occurs as a result of insufficient obliteration of the proximal portion of the vitelline duct. It is one of the most common congenital defects of the gastrointestinal tract and can be accompanied by various severe complications. The incidence of diverticulum is 2-3% in children of all age groups, with clinical manifestations and complications occurring in approximately 25% of cases [1,3,8,9]. This anomaly, both without visible changes and with various complications, is observed in children and adults, and is detected during laparotomy or laparoscopy, as well as during radioisotope studies and capsule video endoscopy of the gastrointestinal tract [14]. According to V.M. Timerbulatova et al. (2017), among 77 patients with complicated forms of DM, 47 patients were children aged 1 to 16 years, the remaining 30 were adult patients aged 21 to 80 years [22].

Typically, DM is a protrusion of a segment of the ileum located in the abdominal cavity, having a variable length and shape, which resembles a vermiform appendix. Usually, it is localized in the abdominal cavity, within the ileum at a distance of 40-70 cm from Baugin's valve [4,10,12,18,19]. The length of the diverticulum can vary from 1-2 cm to 15-20 cm, and its diameter often reaches the width of the

ileum and is considered a giant DM. The literature describes cases of giant Meckel's diverticula, up to 35 cm long and 15 cm in diameter [7,15]. Histologically, the diverticulum wall is similar to the wall of the small intestine, but the muscular layer is less pronounced. In the mucous membrane, areas of heterotopic gastric mucosa or pancreatic tissue may be found [17].

In the clinical picture of DM, most researchers distinguish a triad of symptoms: abdominal pain, gastrointestinal bleeding (GIB) and intestinal obstruction. The prevalence and severity of each of them depends on the anatomical features of DM, the presence of ectopic tissues in the DM wall with different morphofunctional characteristics, the patient's age and physiological characteristics [2,10].

Congenital anomalies of various organs and systems are characterized by a high degree of combination with other single or multiple developmental defects, which complicates diagnosis and the choice of optimal treatment tactics. This pathology can lead to various complications and / or serve as a cause for the formation of other pathological conditions. DM without complications is often detected during operations for pathologies of the abdominal organs. In DM, as in other developmental anomalies, there is a combination of various defects of the cardiovascular, genitourinary system, spine and spinal cord, or an association in the form of a triad of diaphragmatic hernia, intestinal malrotation, heterotopia of pancreatic tissue (HPT) in parts of the gastrointestinal tract

or in Meckel's diverticulum itself [6-20,23-24]. Parkash Mandhan et al. 2014 described a case of a combination of diaphragmatic hernia DG with Meckel's diverticulum and HPT in the wall of the jejunum in a newborn, designating this pathology as a triad: congenital diaphragmatic hernia (DG), DM and HPT. In 2016, L. Basani et al. reported a combination of congenital DG with DM and intestinal malrotation in a three-month-old child, which was the first description of such an association. Migration of organs through the diaphragm is more often observed on the left side with non-closure of the Larrey fissure, less often on the right side with a Morgagni defect. Usually, the DM moves together with other abdominal organs, either in a small volume or significantly, which can lead to pronounced respiratory-hemodynamic or gastroenterological disorders [5,21,23].

Purpose of the Study: was to determine the characteristic features of the clinical course of DM, the difficulties of diagnosis in case of atypical localization of Meckel's diverticulum in a child with intestinal malrotation and spinal anomaly in an infant.

2. Material and Methods of the Study

From 2010 to 2022, 47 children with DM, which caused complications and diseases that required surgical intervention, were observed in the clinical bases of the Department of Hospital Pediatric Surgery of TashPMI. In 7 (14.9%) patients, DM without pathological changes was detected during operations performed on the abdominal organs for appendicitis: in 4 patients; with malformations of the anterior abdominal wall - 2; inguinoscrotal hernia - in 1. Among the patients, there were 32 (68.1%) boys and 15 (31, 9%) girls. Pathology was detected in 6 (12.8%) children under 3 months, from 3 months. up to 1 year - in 4 (8.5%), from 1 year to 3 years - in 8 (17%), from 3 to 7 years - in 8 (17%), from 7 to 12 years - in 12 (25.5%) and from 12 to 18 years - in 9 (19.1%). This report analyzes diagnostic and operational-tactical errors, complications that arose in a patient with atypical localization of the apical and middle part of the DM in the chest cavity through a diaphragmatic defect on the right, and

its base originating from the ileum in the abdominal cavity.

At the diagnostic stages, laboratory, instrumental, ultrasound, radiation, as well as endoscopic and histomorphological studies were used.

3. Results and Discussion

No specific clinical manifestations were observed in uncomplicated DM. The remains of the rudimentary vitelline duct in the form of a fixed cord of Meckel's diverticulum with the umbilicus or mesentery caused intestinal volvulus and intestinal compression with the manifestation of symptoms of intestinal obstruction (Table 1).

Table 1. Forms of DM pathology identified during surgery intervention (n=47)

DM without changes or pathology	Number of patients	
	Abc.	%
Diverticulum without change	7	14,9%
Diverticulitis without peritonitis	13	27.7%
Diverticulitis with perforation	9	19,1%
Intussusception	6	12,8%
Strangulating intestinal obstruction	8	17,1%
Evagination of a patent vitelline duct	1	2,1%
Foreign body in the DM	1	2,1%
Ulcerations DM	1	2,1%
Thoracoabdominal form of DM	1	2,1%
Total:	47	100%

The remains of the rudimentary yolk duct in the form of a fixed cord of Meckel's diverticulum with the navel, mesentery can provoke intestinal volvulus, intestinal compression with manifestation of symptoms of intestinal obstruction. The main clinical signs and the nature of the complications that have arisen are presented in Table 2.

Of particular clinical interest is the case of a child with multiple malformations, including a diaphragmatic hernia on the right with displacement of the DM, intestinal malrotation and anomaly of the cervical spine.

Table 2. Clinical signs of Meckel's diverticulum with its complications in children (n=40)

Clinical signs	Intussusception (n= 6)	Strangulation obstruction (n= 8)	Intestinal bleeding (n= 1)	Diverticulitis (n= 13)	Diverticulum perforation (n= 9)	Total (n=39)
Pain syndrome	6	8	-	13	9	38
Vomiting	6	8	1	5	9	29
Increased body temperature	-	2	-	9	9	20
Melena	5	3	1	-	-	9
Signs of peritonitis	-	-	-	-	9	9

We present a clinical case.

Patient T.M., 1 year 4 months. History of the disease. № 9894-386. Admitted to the Department of Thoracic Surgery 2 - City Children's Hospital on April 7, 2023 with a referral diagnosis: MCPD. Cystoadenomatous malformation of the upper lobe of the right lung. Condition after surgery for intestinal malrotation (Ledd's syndrome). Severe protein-energy malnutrition. From the anamnesis, it was found that the child is from the first pregnancy and first birth, which proceeded with toxicosis. He was born at a gestational age of 40 weeks with a body weight of 2400 g, length 46 cm. Antenatal examination was not performed. Assessment on the Apgar scale after birth at 1 and 5 minutes were 8 and 9. On the 2nd day of life, vomiting with an admixture of bile appeared. The child was hospitalized in the surgical department, where radiographic data revealed the following diagnosis: high partial intestinal obstruction. On the 5th day of life, the child was transferred to the neonatal surgery department of the Republican Perinatal Center (RPC). Upon admission, the child's condition was severe due to endotoxemia, the skin was subicteric. Respiratory rate was 58 per minute, pulse was 166 beats per minute. Heart sounds were muffled. Breathing was free, the chest was of regular shape, without visible deformations. Moist rales were heard during auscultation against the background of harsh breathing. The abdomen was soft, moderately distended, stagnant discharge was detected through a gastric tube. Blood tests: hemoglobin - 155.0 g/l, erythrocytes. 5.18×10^{12} , CI 0.92, thrombus. 274×10^9 /l, leukocytes - 9.1×10^9 , seg. nucleus - 46%, lymphocytes - 40%. Total protein - 58.0 g/l. Total bilirubin - 155.8 mmol/l, direct - 87.6 mmol/l, indirect - 68.2 mmol/l. Stool and urine tests are normal. Neurosonography shows signs of hypoxic CNS damage. Ultrasound of the gastrointestinal tract - dilation of the stomach and duodenum. Chest X-ray reveals an air cyst in the projection of the upper lobe of the right lung.



Figure 1. General chest radiography. Cystic formation in the projection of the right lung

Taking into account the phenomena of partial intestinal obstruction, a contrast study of the gastrointestinal tract was performed, the results of which were assessed as a moderate

retention of contrast in the stomach and duodenum. Echocardiography revealed an open oval window with a diameter of 4 mm and an open arterial duct with a diameter of 22.0 mm. Based on the research data and the results of corrective measures for the identified violations, on 01/10/2022 the child was operated on with a presumptive diagnosis: partial high intestinal obstruction. An upper supraumbilical transverse laparotomy was performed. During the revision, the pathology found was assessed as malrotation of the Ledd syndrome type, a Ledd operation and appendectomy were performed. The midgut was left in the original position of incomplete rotation. The postoperative course is uneventful, on 03/16/2022 the child was discharged from the hospital in a satisfactory condition, weighing 2500 g. Blood counts: heme 115 g/l, erythrocytes 4.17×10^{12} , CI 0.8, platelets - 274×10^9 /l, leukocytes 8.8×10^9 , segmental nucleus 46%, lym. 40%, total protein 44.4 g/l, total bilirubin 14.4 mmol/l, direct bilirubin absent. Regarding the "congenital lung cyst", a wait-and-see approach and dynamic observation are recommended. After discharge from the hospital, the child gained little weight and lagged sharply behind in physical development. On June 18, 2022, the child was hospitalized in the gastroenterology department of the scientific and practical center of pediatrics with a body weight of 4250 g for adjuvant nutritional therapy. and a height of 58 cm. with a diagnosis of severe protein-energy malnutrition. Condition after Ledda's operation. The child received complex treatment for 10 days aimed at correcting the identified disorders. In the hospital, he gained 200 grams in body weight. On June 27, 2022, he was discharged from the hospital with a body weight of 4450 grams in a moderate condition with recommendations regarding feeding and examination by a surgeon regarding lung pathology. Upon admission to the thoracic department at the age of 1 year 4 months, the condition was moderate, malnutrition, body weight was 7500 grams. (deficit - 25%). On the anterior abdominal wall, there is a scar from transverse supra-umbilical laparotomy. Upon examination, the liver protrudes 2-2.5 cm from under the edge of the costal arch, wheezing is heard in the lungs against the background of harsh breathing. Blood tests: hemoglobin - 99 g/l, erythrocyte sedimentation rate - 10 ... - 3.87×10^{12} , CP - 0.82, platelets - 234×10^9 /l, leukocytes - 9.2×10^9 , segmental nucleus - 56%, lymphocytes - 32%. Against the background of correction of protein-energy deficiency, hematological and electrolyte disturbances due to temperature reaction, antibiotic therapy with Ceftriaxone was started. On 11.04.2023, MSCT angiography of the lungs was performed. The walls of the pulmonary arteries are smooth, clear. The lumen to the subsegmental arteries is preserved. The walls of the thoracic aorta are moderately thickened due to small plaques of a mixed nature, its lumen is preserved. The mediastinal organs are not displaced. No fresh foci or infiltrative changes were found in the pulmonary fields on the right and left. The vascular pattern is normal. The bronchial tree is not deformed. The roots of the lungs are dilated, signs of infiltration of the root tissue and reaction of the root lymph nodes are not revealed. A defect of the right

dome of the diaphragm with a diameter of 2.7 cm at the level of the posterior triangle with an outlet of the hernial sac into the chest cavity with dimensions of 3.4 x 5.1 x 7.6 cm is

determined; the contents of the sac are fatty tissue and intestinal loops compressing the medial sections of the right lung.

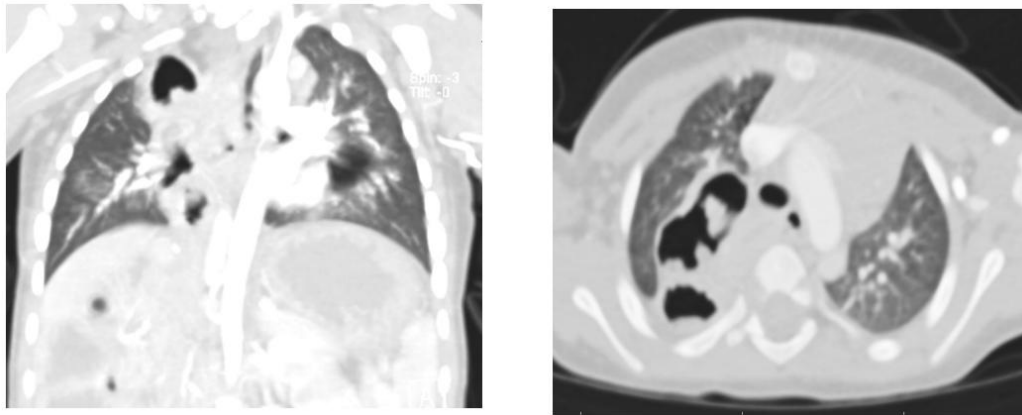


Figure 2. MSCT (description in text)

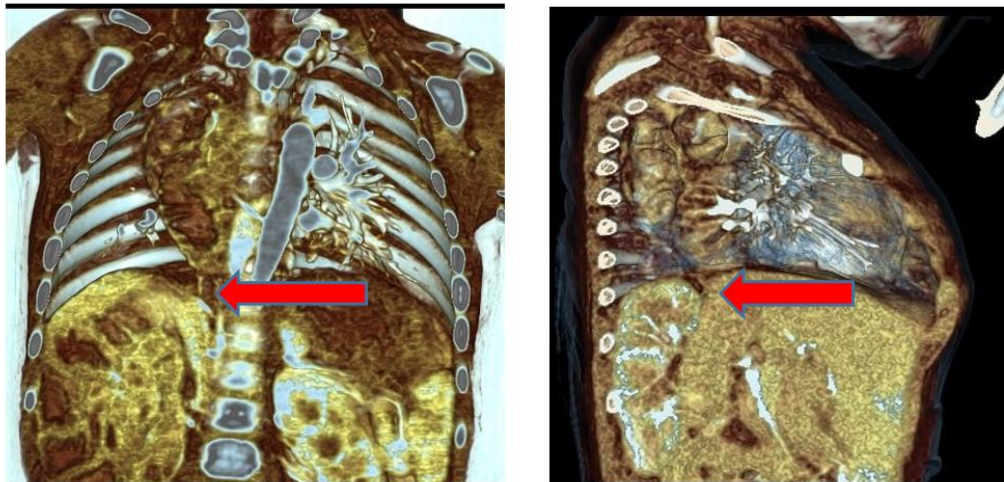


Figure 3. MSCT angiography. A defect is determined in the projection of the posterior triangle of the right dome of the diaphragm and the displacement of intestinal loops into the chest cavity

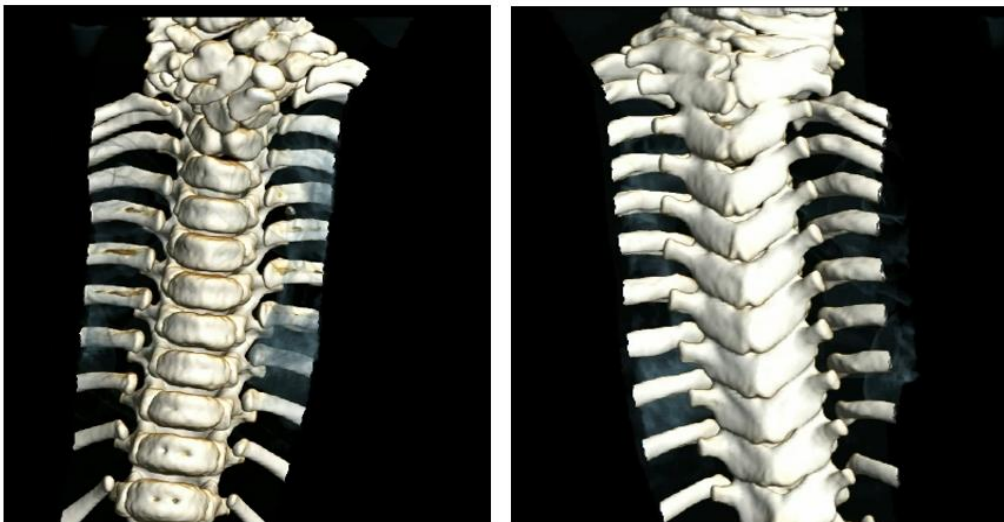


Figure 4. MSCT of the cervicothoracic spine. Description in text

In the lower cervical region, local lordosis is determined. The spinal canal at the level of the lower cervical region is expanded to 1.8 cm. The spinal column in the cervicothoracic region is C-shaped curved. The cervical vertebrae (C5-6-7) are formed, consist of many individual semi-vertebrae. The TH1 vertebra is presented as a semi-vertebra, the right half of the body and 1 rib on the right are not determined, the arches of the TH1 and TH2 vertebrae are fused.

Due to the atypical location of the diaphragm defect with the formation of a hernia on the right, contrast irrigography and passage of contrast through the gastrointestinal tract were performed to clarify the diagnosis. On the irrigogram, most of the contrasted colon is located in the left half of the abdominal cavity, the transition of the contrast in the form of reflux into the lumen of the small intestine localized in the abdominal cavity is clearly visible. Movement into the chest is not determined.

During the passage through the gastrointestinal tract, the contrast passes freely through the esophagus, there is no evidence of hernia of the esophageal opening and paraesophageal hernia. Evacuation from the stomach is timely. In subsequent images, a violation of the normal configuration of the duodenum is visible without its typical branches and fixation in the area of the Treitz ligament. The initial sections of

the jejunum are slightly expanded. In the images, at 1 hour 30 minutes and 3 hours, the passage of the contrast into the chest cavity, further movement through the small intestine, a gradually increasing volume of contrast and noticeable peristalsis in the thoracic section of the intestine are determined.



Figure 5. Contrast irrigography, left-sided location of the colon

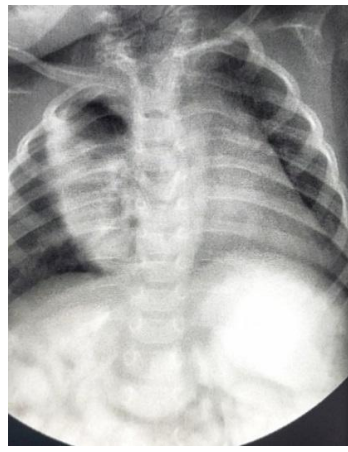
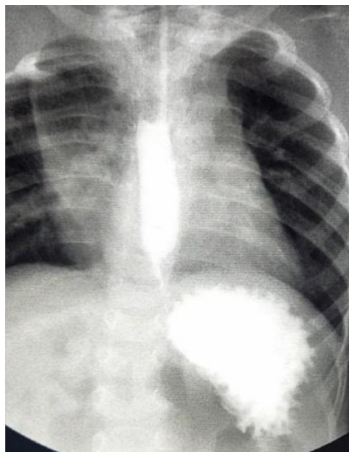


Figure 6. Passage through the gastrointestinal tract. Description in the text

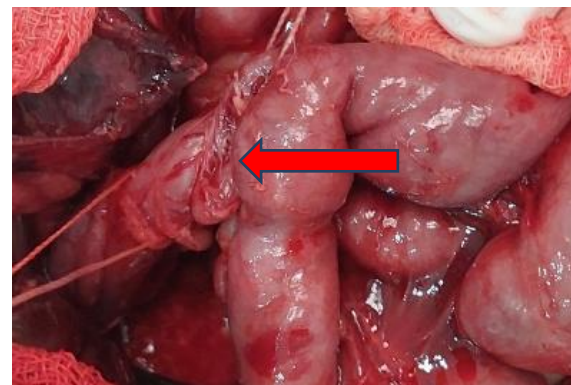
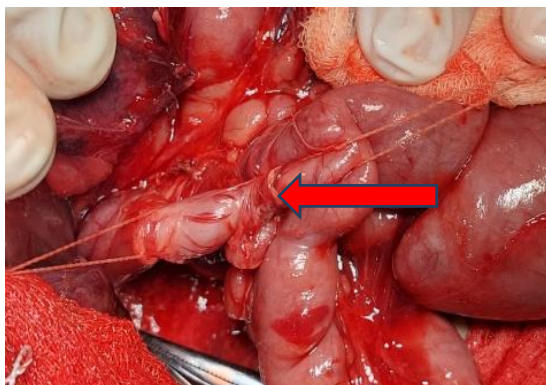


Figure 7. Meckel's diverticulum originating from the wall of the ileum

The data of contrast irrigography and gastrointestinal tract passage correspond to the condition after surgery for intestinal malrotation. Characteristic data for a diaphragmatic hernia on the right are determined. Based on the results of complex studies, the diagnosis was established: MPR. Diaphragmatic hernia on the right, PDA, anomaly of the spine and spinal cord. Severe protein-energy malnutrition. The condition after surgery is intestinal malrotation. It was decided to perform surgery to eliminate the diaphragmatic hernia using laparotomic access. On 04/26/2023, the operation began with excision of the old supra-umbilical postoperative transverse scar (operated by Professor Ergashev N.Sh.). During revision, massive interintestinal adhesions and adhesion of intestinal loops to the peritoneum and on the surface of the liver were revealed. With technical difficulties, the intestine was mobilized from adhesions. During the mobilization and revision it was established that the duodenum does not have a typical anatomical structure in the form of a horseshoe and syntopy - its fixation and duodenojejunal flexure are absent. The moderately dilated duodenum passes onto the dilated jejunum. The indicated sections are fixed with strands forming non-sharp bends. The large intestine is mostly located in the left flank of the abdominal cavity, the ileocecal angle is located along the midline at the level of the navel, the vermiform appendix was removed during the first operation. During the release of the small intestinal loops from interintestinal adhesions, the gallbladder and surrounding formations in the right hypochondrium, immersion of the small intestine into the chest cavity through a diaphragmatic defect in the middle part of the costophrenic angle was established, freely passing the index finger. When separating the adjacent sections of the intestine, it was found that at a distance of 40 cm from the ileocecal angle, the intestinal outgrowth plunges into the chest cavity through the specified defect. The proximal and distal sections of the intestine have a normal lumen with preserved patency. The pathology is assessed as Meckel's diverticulum with dislocation into the chest cavity in the form of a diaphragmatic hernia.



Figure 8. Resected section of small intestine carrying DM

After moderate expansion of the diaphragmatic defect, this formation was gradually mobilized by pulling it into the wound and step-by-step coagulation of the bleeding vessels to a depth of 10-12 cm. In this case, the mobilized DM up to 25 cm long was brought out into the abdominal cavity. Signs

of damage to the lung parenchyma or blood vessels were not noted. A drainage tube with lateral openings was left in the bed, the diaphragmatic defect was sutured with interrupted sutures. Having made sure that the formation was a dislocated Meckel's diverticulum with a wide base, segmental resection of the intestinal section carrying the diverticulum was performed with the imposition of an end-to-end anastomosis.

The operation was completed with layer-by-layer suturing of the surgical wound and leaving a drainage tube in the abdominal cavity. After the operation, with adequate hemodynamic and hematological parameters, the child was transferred to the intensive care unit and connected to the monitoring system. Auxiliary mechanical ventilation was continued, a course of supportive and antibacterial therapy was prescribed, and planned analgesia with fentanyl. During dynamic observation, the child's multiple organ failure progressed. Despite corrective therapy, cardiac arrest occurred 12 hours after the operation. Resuscitation measures were ineffective, biological death was confirmed. Autopsy revealed no complications associated with the operation. Pathological diagnosis: multiple malformations: intestinal malrotation, common mesentery, dislocation of the DM into the chest in the form of a diaphragmatic hernia, polysplenia. Multiple interorgan adhesions in the abdominal cavity, polysegmental pneumonia, obstructive endobronchitis, pulmonary edema. Toxic hepatosplenomegaly, intestinal paresis, toxic nephrosonephritis, myocarditis, adrenal hemorrhage. Hypotrophy. In the presented observation, a number of anatomical features of the pathology, the nature of the combination of concomitant anomalies and diagnostic and tactical errors, causes of death in the early stages after surgery can be noted. Our case belongs to the category of rare concomitant multiple anomalies involving DM, they consisted of intestinal malrotation with characteristic data for Ladd's syndrome, right-sided diaphragmatic hernia through a defect in the sternal and costal beginning of the diaphragm - the foramen of Morgagni, with the movement of only DM without other parts of the gastrointestinal tract. Although the length of the DM was about 12 cm, its narrowness limited the increase in intrathoracic tension, and moderate respiratory disorders were observed. However, the formation detected in the right hemithorax during radiographic examination led to an erroneous interpretation of the pathology as a cystadenomatous lesion of the right lung. Subsequent computed tomography studies using contrast agents clarified the diagnosis. During the primary operation for intestinal obstruction, a right-sided diaphragmatic hernia with DM displacement remained unrecognized due to incomplete revision. During the operation, polysplenia was detected in the form of separate additional lobes of the spleen, confirmed during autopsy, which confirms the presence of polydefects. The patient also had an open oval window and non-closure of the arterial duct without pronounced hemodynamic disorders. Retrospective analysis of the computed tomography data allowed us to identify abnormalities in the cervical spine without precise detailing of the nature of osteoneural pathology. The final surgical method in this case was segmental resection

of the intestine carrying the DM with the imposition of an end-to-end anastomosis and suturing of the diaphragmatic defect using translaparotomic access. According to most surgeons, in case of intraoperative establishment of DM with pathological changes and in cases without changes, active surgical tactics should be followed - segmental resection of the intestine carrying the DM, or diverticulectomy with immersion of the stump, depending on the nature of the change and the diameter of its base. According to literary data, the immediate and remote results of treatment are quite favorable. The causes of death in the early stages of the postoperative period in our case remain completely unclear. It can be assumed that the combination of identified anomalies and secondary changes developed in other organs against the background of hypotrophy and protein-energy deficiency had a negative effect on the tonatogenesis of death.

4. Conclusions

In conclusion, it is important to note that Meckel's diverticulum (MD) as a congenital anomaly of the gastrointestinal tract is often detected during surgical interventions. Its frequency is 2-3%, while clinical manifestations and complications are observed in 25% of cases. In the clinical picture of the pathology, a triad of symptoms can be distinguished: abdominal pain, gastrointestinal bleeding (GIB) and intestinal obstruction of varying severity. The absence of pathognomonic signs of DM creates significant difficulties in diagnosis. DM is often combined with malformations of individual organs or is an association of various anomalies of other organs and systems, which aggravates their course. In recent publications, there is an increase in the descriptions of such combinations. Mandatory resection of both pathologically altered and intact DM is the main surgical tactic, since the risk of subsequent complications is incommensurate with simple diverticulectomy. The main method of treating DM is diverticulectomy. The options for its implementation and the types of simultaneous interventions depend on the severity and prevalence of the inflammatory process in the wall of the ileum and the diverticulum itself, as well as on the nature of the complications and secondary pathology from the peritoneum and abdominal organs."

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