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## Clinic, diagnosis and treatment of dunbar syndrome in children

**Andrey V. Andreev**

Assistant of the Department of "Children's Diseases" of the I.N. Ulyanov Chuvash State University, Head of the X-ray Department, radiologist of the Republican Children's Clinical Hospital of the Ministry of Health of Chuvashia, Cheboksary  
Corresponding author email: [Yurina\\_iriha@mail.ru](mailto:Yurina_iriha@mail.ru)

**Mikhail V. Krasnov**

MD, Professor, Head of the Department of "Children's Diseases" of the I.N. Ulyanov Chuvash State University, Cheboksary

**Larisa P. Nikitina**

Radiologist of the Republican Children's Clinical Hospital of the Ministry of Health of Chuvashia, Cheboksary

**Abstract**---Aim: To determine the clinical, diagnostic and therapeutic features of Dunbar syndrome in children. Methods: Scientific work was carried out based on the "Republican Children's Clinical Hospital" and the "City Children's Clinical Hospital", Cheboksary. The basis of this study was a survey of 200 children with celiac artery compression syndrome. The results obtained in the course of the work were analyzed on the basis of the Department of Children's Diseases of the I.N. Ulianov Chuvash State University. In addition to routine diagnostic methods, Instrumental methods have been adopted: ultrasound Doppler scanning (Hitachi Vision Avius, Japan), CT angiography (SOMATOM Emotion, SIEMENS (China), Revolution EVO, GE (Russia)) and Magnetic resonance angiography (Optima MR450w 1.5, USA). The duration of the study is short (after surgery) and long-term (up to 2 years). Results: Dunbar syndrome is more often observed in girls, which in 75% of cases is accompanied by signs of chronic abdominal ischemia. Significant and prolonged stenosis of the celiac artery can cause the development of a number of complications (pancreatic-duodenal artery aneurysms, etc.). Pathophysiological changes against the background of compression syndrome of the celiac trunk were characterized by regional hemodynamic disorders - the flow of blood flow from the superior mesenteric artery into the basin of the CT, the degree of which depends on the degree of diameter of the stenosed segment. Changes in the rheological features of blood confirm their pathogenetic significance in the formation of morphological disorders of the upper gastrointestinal tract. The

intensity of these changes is directly associated with the degree of compression and duration of pathology. The clinical symptoms of Dunbar syndrome manifest themselves in three forms - painful, dyspeptic, and neurovegetative. Ultrasound Doppler scanning of the abdominal aorta, the celiac trunk and the superior mesenteric artery seems to be a fairly reliable and accurate way to diagnose the compression syndrome of the celiac trunk and its treatment, with an efficiency of 98.5%. Early diagnosis and timely surgical intervention of Dunbar syndrome contribute to achieving complete recovery in 87.5% ( $p < 0.01$ ) of patients, significant improvement – in 7.4% ( $p < 0.01$ ) of patients, and unsatisfactory results – in 5.1% ( $p < 0.01$ ) of patients.

**Keywords**---celiac trunk compression syndrome, children, Dunbar syndrome, doppler ultrasound, surgery.

## **Introduction**

Different classifications of vascular diseases are presented in the literature, but a more detailed one, taking into account the factors of origin, anatomical structures and intensity of microcirculatory disorders, is the classification by V.P. Kulikov in 2007. Celiac artery compression syndrome (CFCS, ICD – 10:177.4) is a pathology caused by simultaneous extravasal mechanical compression of the celiac trunk (CT) of the abdominal aorta, the median arched ligament of the diaphragm and/or its legs, or neurofibrous tissue of the CT (Zainulabidov et al., 2021; Kohaut et al., 2017).

The relevance of this work is stated from the following: high frequency of occurrence (4.49-20.2%); complex clinical course; absence of a standard clinical strand in the diagnosis of CFCS and its subsequent therapy; progressive mortality rate (Zainulabidov, Razumovsky, Khavkin, 2020; Meister et al., 2020). One of the main clinical manifestations of CFCS is abdominal pain syndrome, manifested as a result of its compression. This syndrome can clinically occur in a recurrent form. According to modern literature, the frequency of chronic pain in the compression syndrome of the celiac trunk is 11.2-15.2% of children. More than half of patients with CFCS are girls (61.8%), and the rest are boys (Razumovsky et al., 2020; Leelakanok et al., 2017). According to research by Dawn M Coleman et al. (2017) it was revealed that the annual occurrence of detected patients with chronic abdominal ischemia increases by 14.3-18.1 % (Andreev, Krasnov, 2018; Pandey et al, 2015).

Despite the obvious development in the technological transformation of healthcare and the introduction of modern technologies into its system, at present, the issue of modernization of diagnostic metodes of CT stenosis and improvement of its therapeutic tactics in children pathogenetically remains poorly understood. Moreover, this pathology seems promising for a number of authors, not only pediatricians, but pediatric surgeons (Razumovsky et al., 2017; García-Hernández et al., 2020; Arakelyan et al., 2014).

**The purpose of the study**

To determine the clinical, diagnostic and therapeutic features of Dunbar syndrome in children.

**Material and methods**

Scientific work was carried out based on the "Republican Children's Clinical Hospital" and the "City Children's Clinical Hospital", Cheboksary. The basis of this study was a survey of 200 children with celiac artery compression syndrome. The results obtained in the course of the work were analyzed on the basis of the Department of Children's Diseases of the I.N. Ulianov Chuvash State University.

Criteria for the participation of patients in the study. Age - 5-17 years; gender male and female; own consent (parents) to participate in the work; confirmation of the clinical diagnosis of "compression syndrome of the celiac trunk" with the help of clinical, biochemical and instrumental diagnostics; compliance with medical instructions; benign cause; treatment – surgical. Criteria for canceling patients. Age < 5 and > 17 years; personal refusal (of parents) to participate in the study; severe concomitant (malignant, somatic, psychological, etc.) diseases; conservative treatment; pregnancy; lactation.

In the course of studying the sex and age composition of the study groups, it was found that the frequency of girls was 120 (60.0 %) of the patient, and boys - 80 (40.0 %). The average age of the patients was  $9.7 \pm 0.34$  years. At the same time, preschool age was registered in 10 (5.0 %), primary school age – in 75 (37.5 %), and adolescent age - in 115 (57.5 %). The diagnosis was established in a planned form in 110 (55.0 %) patients, and in an emergency – in 90 (45.0 %). A high proportion of diagnosis detection in urgent conditions may cause doubts about the diagnosis, or a decrease in the quality of therapeutic therapy, which leads to a fatal outcome.

The duration of the disease from the manifestation of primary signs to the establishment of a clinical diagnosis took a long time – from one month to 50 years ( $13.7 \pm 0.69$  years). This confirms the untimely verification of the diagnosis, which can serve as a factor in the progression of pathology and a high risk of mortality. In the course of studying the family history, it was found that genetic predisposition was registered in the families of 65 patients (32.5 %). This examination indicates the role of a hereditary factor in the formation of Dunbar syndrome.

To compare the studied parameters with reference values, conditionally normal children were examined,  $n=30$ , age - 5-17 years, gender - boys (11 (36.7 %)) and girls (19 (63.3 %)). The following methods are used in the study. In addition to routine methods for the diagnosis of celiac artery compression syndrome, the following methods were used: Laboratory: assessment of the aggregation coefficient of blood capacity and fluidity, blood viscosity, adhesion and platelet aggregation.

In addition to routine diagnostic methods, instrumental methods have been adopted: ultrasound Doppler scanning (Hitachi Vision Avius, Japan), CT angiography (SOMATOM Emotion, SIEMENS (China), Revolution EVO, GE (Russia)) and Magnetic resonance angiography (Optima MR450w 1.5, USA). The duration of the study is immediate (after surgery) and long-term (up to 2 years). Static analysis of the obtained results was performed on a personal computer using the Statistica 6.0 digital software package. Methods of descriptive parametric and nonparametric, and comparative statistics were adopted.

## **Result and discussion**

When studying the period of development of the disease, it was found that a gradual onset was recorded in 150 (75.0 %) patients, and acute in -50 (25.0 %). In this group, during several months and before the clarification of the clinical diagnosis, the progression of the condition of their patients in the form of intense abdominal pain and other clinical signs was established.

When analyzing the clinical manifestations of Dunbar syndrome (according to both sick children and their parents), it was shown that 189 (94.5 %) patients had abdominal pain. The rest of the children (11 (5.5 %)) neurovegetative signs and a feeling of heaviness in the epigastric region were revealed. Pain syndrome was registered in 3 groups. The first group of patients had constant abdominal pain, observed in 25 (12.5 %). In the second group, sustained abdominal pain was noted (in 145 (72.5 %) patients). The third group of patients was characterized by paroxysmal pain – in 30 (15.0 %).

According to the localization of the pain syndrome, it was recorded that the pain was mainly in the epigastric region and hypochondrium (175, 87.5 %), also in the lower abdomen (18, 9 %), and throughout the abdomen (7, 3.5 %). Along with this, the pain was localized in the lumbar and interscapular areas - in 185 (92.5 %) patients. At the same time, among 135 (67.5%), the pain syndrome was moderate and the rest (65, 32.5 %) – pronounced, who had a decrease in the quality of life and ability to work. The pains were also characterized by aching appearance (in 142, 71.0 %) and cutting (58, 29.0 %).

During the anamnesis of the disease, it was shown that the provoking factor of the occurrence and intensification of pain syndrome was food intake – in 120 (60.0 %) patients, physical activity – in 60 (30.0 %), emotional stress – in 20 (20.0 %). 64 (32.0 %) children were forced to reduce the amount of food intake due to fear of increased abdominal pain. The feeling of abdominal overflow of food intake was registered in 110 (55.0 %) patients.

Dyspeptic manifestations were observed in the studied children in the form of a decrease in the desire for food - in 75 (37.5 %) patients, nausea – in 62 (31.0 %), vomiting – in 52 (26.0 %), belching – in 46 (23.0 %), heartburn – in 39 (19.5 %), constipation or diarrhea – in 84 (42.0 %), a feeling of fullness of the abdomen – in 44 (22.0 %). Neurovegetative disorders were also observed in children with Dunbar syndrome, their intensity was varied. Asthenia was detected, observed in 108 (54.0 %), a feeling of pulsation in the abdomen – in 71 (35.5 %), emotionally unstable - in 96 (48.0 %), palpable palpitations - in 57 (28.5%), hyperhidrosis – in

43 (21.5 %), pain in the head – in 65 (32.0 %), episodes of difficulty breathing – in 37 (18.5 %) and loss of consciousness – in 15 (7.5 %).

Objective manifestations of celiac trunk compression syndrome revealed during abdominal palpation were pain in the epigastric region - in 175 (89.0 %) patients, pronounced body weight deficiency (when calculating body mass index) – 83 (41.5 %), yellow pigmentation of the eye mucosa – in 5 (2.5 %). Blood pressure corresponded to the reference index in the majority of patients (169, 84.5%), at an elevated level – in 11 (5.5 %), and at a reduced level – in 20 (10.0 %). The body temperature index was recorded in the normal value in 191 (95.5 %) and in the subfebrile form in 9 (4.5 %).

In 87 (43.5 %) patients, concomitant pathologies of the digestive (gastritis, gastroesophageal reflux, peptic ulcer, etc.), respiratory (ARVI, etc.), cardiovascular (aneurysm of the lower pancreatic-duodenal (PDA) arteries (in 4 (2.0 %)), upper posttraumatic arteriovenous fistula of the upper mesenteric artery (in 3 (1.5 %)), stenosis of the left renal artery were noted (in 1 (0.5 %))), urinary (urolithiasis, chronic nephritis, etc.), musculoskeletal (osteochondrosis, scoliosis of the spine, etc.) system. So, the main clinical signs of celiac trunk compression syndrome appeared in 3 symptom complexes: painful - in 189 (94.5 %), neurovegetative – in 136 (68.0 %), and dyspeptic – in 115 (57.5 %). Depending on the degree of intensity of clinical signs of Dunbar syndrome, two stages were observed in the study patients (decompensatory - in 193 (96.5 %), and subcompensatory - in 7 (3.5 %)), and three forms (progressive - in 171 (85.5 %), persistent - in 19 (9.5 %), and recurrent - in 10 (5.0 %)).

When studying the angiographic examination data, the main reference parameters of ultrasound Doppler scanning (USDS) of the celiac trunk and upper mesenteric artery were established in the norm group of three modes - calm breathing, maximum inhalation and maximum exhalation. At the same time, the diameter of the celiac trunk was  $6.31 \pm 0.15$  mm,  $6.41 \pm 0.18$  mm, and  $6.11 \pm 0.12$  mm, respectively. The linear peak systolic blood flow velocity (LPSBFV) of the celiac trunk was  $1.02 \pm 0.05$  m/s,  $0.93 \pm 0.03$  m/s and  $1.11 \pm 0.07$  m/s. The gradient of arterial pressure (GAP) of CT was  $4.31 \pm 0.29$  mm Hg,  $4.01 \pm 0.25$  mm Hg, and  $5.12 \pm 0.32$  mm Hg. On the other hand, the diameter of the mouth of the superior mesenteric artery (SMA) was  $6.21 \pm 0.15$  mm,  $6.34 \pm 0.18$  mm, and  $6.28 \pm 0.13$  mm, and its LPSCC was  $1.03 \pm 0.08$  m/s,  $1.0 \pm 0.06$  m/s, and  $1.10 \pm 0.12$  m/s, and GAP –  $4.61 \pm 0.26$  mm Hg,  $4.32 \pm 0.21$  mm Hg, and  $5.47 \pm 0.34$  mm Hg. Thus, there is no statistically significant difference between these indicators ( $p > 0.05$ ).

When examining patients with celiac trunk compression syndrome with the help of USDS, it was noted that the length of compression of the celiac trunk from its mouth was  $10.52 \pm 0.36$  mm. The diameter of the stenosed segment of CT with calm breathing was  $2.72 \pm 0.04$  mm, with maximum exhalation –  $2.61 \pm 0.06$  mm, and with maximum inhalation  $3.42 \pm 0.07$  mm. So, the diameter of CT in the study patients was lower than the normal group with calm breathing - by 56.9 % ( $p > 0.01$ ), with maximum inhalation and exhalation - by 59.2 and 44.0 % ( $p > 0.01$ ).

The LPSBFV in the studied patients was  $2.11 \pm 0.07$  m/s with calm breathing,  $1.48 \pm 0.04$  m/s with maximum inhalation, and  $2.51 \pm 0.09$  m/s with maximum exhalation. When compared with the reference parameter, the linear peak systolic blood flow velocity was higher by 106.8, 59.1, and 126.1 % ( $p < 0.01$ ), respectively.

The gradient of blood pressure in children with Dunbarapri syndrome on calm breathing was  $17.21 \pm 0.81$  mm Hg ( $p > 0.01$ ), with maximum inhalation and exhalation -  $10.16 \pm 0.47$  mm Hg and  $27.51 \pm 1.73$  mm Hg ( $p > 0.01$ ). At the same time, the value of GAD in patients was higher when compared with the norm group by 299.3, 153.3, and 437.0 % ( $p < 0.01$ ). A detailed analysis of the ultrasound Doppler scan data revealed that the diameter of the distal segment of the celiac trunk stenosis was  $8.62 \pm 0.43$  mm. The value of this indicator was greater than the proximal department of CT stenosis by 5.89 mm (216.5%,  $p > 0.01$ ).

In general, according to the results of ultrasound and angiography, the degree of compression syndrome of the celiac trunk in 178 (87.5 %) patients was more than 50% ( $p > 0.01$ ) in diameter. In this group, the linear peak systolic blood flow velocity of the celiac trunk was 2.31-2.74 m/s, and the blood pressure gradient was 22 - 81 mm Hg. In the remaining patients of the study (22, 11.0%), the degree of compression of CT was less than 50% ( $p > 0.01$ ), in whom the LPSBFV was  $1.61 \pm 0.13$  m/s, and the GAP was 6 - 45 mm Hg.

According to the USDS of the superior mesenteric artery, it was found that in 145 (72.5 %) patients, the degree of stenosis was 13-31 % ( $18.1 \pm 0.17$  %) ( $p > 0.01$ ), the LPSBFV at the maximum output was accelerated to 1.45 m/s (76.7 % lower than the reference index). When comparing LPSC in CT situations and VBA using ultrasound Doppler scanning, it was observed that the difference between their maximum exhalation and inhalation was 0.96 and 0.38 m/s ( $p < 0.01$ ). This confirms the fact that LPSBFV in the SMA has a direct relationship with the degree of compression syndrome of the CT, in which there is an increase in blood flow from the SMA to the branches of the celiac trunk. When studying the rheological properties of blood serum in the study patients, their marked deterioration was recorded (Fig. 1).

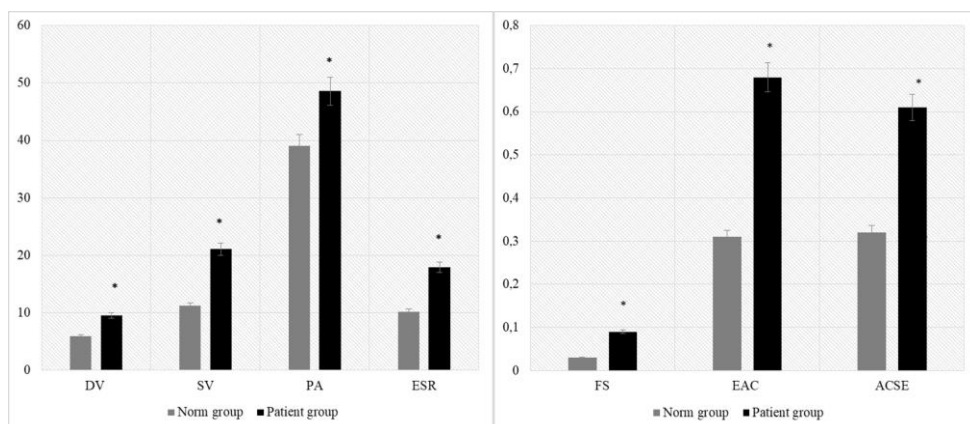


Figure 1. State of rheological properties of blood serum (DV – dynamic viscosity, FS – flow stress, SV – structural viscosity, EAC – erythrocyte aggregation

coefficient, PA – platelet adhesion, ESR – erythrocyte sedimentation rate, ACSE – aggregation coefficient of shaped elements)

Dynamic blood viscosity exaggerated the initial parameter in the preoperative period by 64.8% ( $p < 0.01$ ), serum yield strength - by 198.2% ( $p < 0.01$ ), structural viscosity - by 88.3% ( $p < 0.01$ ), platelet adhesion - by 24.2% ( $p < 0.01$ ), erythrocyte aggregation coefficient - by 90.6% ( $p < 0.01$ ), erythrocyte sedimentation rate - by 75.4% ( $p < 0.01$ ).

It should be noted that changes in the rheological properties of blood were recorded to the greatest extent in patients with compression of the celiac trunk of more than 50%. The main indications for surgical therapy were the following: the degree of stenosis of the CT according to the USDS is more than 50%, the LPSBFV of the emergency is more than 2 m / s, the GAP is 15 mmHg, the aneurysm of the PDA.

2 methods of surgical intervention were used – the first, isolated decompression of the celiac trunk (performed in 179 (89.5 %,  $p < 0.01$ ) patients), the second, decompression of CT with abdominal organs surgery (aorto-femoral bypass surgery, Geller cardiomyotomy, embolization and resection of the aneurysm of the PDA, gastric resection, elimination of adhesive intestinal obstruction, etc.) (in 21 (10.5 %,  $p < 0.01$ ) patients).

Intraoperative complications were bleeding from the damaged right diaphragmatic artery (in 1 (0.5 %),  $p < 0.01$ ), purulent-inflammatory process of the surgical wound (in 2 (1.0 %),  $p < 0.01$ ), and hemoperitoneum of unknown origin (in 1 (0.5 %),  $p < 0.01$ ).

In the postoperative period, hemorheological and clinical (abdominal pain, dyspeptic disorders, neurovegetative symptoms, etc.) disorders were restored within 10 days in 72 (36.0 %,  $p < 0.01$ ) patients, and 38-50 days – in the rest (128, 64.0 %,  $p < 0.01$ ). This indicates that the degree of hemorheological disorders of the blood is directly related to the degree of emergency stenosis and the duration of pathology.

The disappearance of clinical manifestations and improvements in performance were recorded after 2-3 months of surgical treatment were observed in 169 (84.5 %,  $p < 0.01$ ) patients, and in the remaining 31 (15.5 %,  $p < 0.01$ ) - these disorders persisted. When studying the results of wandering (after 4 months) postoperative treatment, recovery was found in 88.1% ( $p < 0.01$ , improvement - in 8.0 % ( $p < 0.01$ ), and without change - in 3.9 % ( $p < 0.01$ ) (Fig. 2).

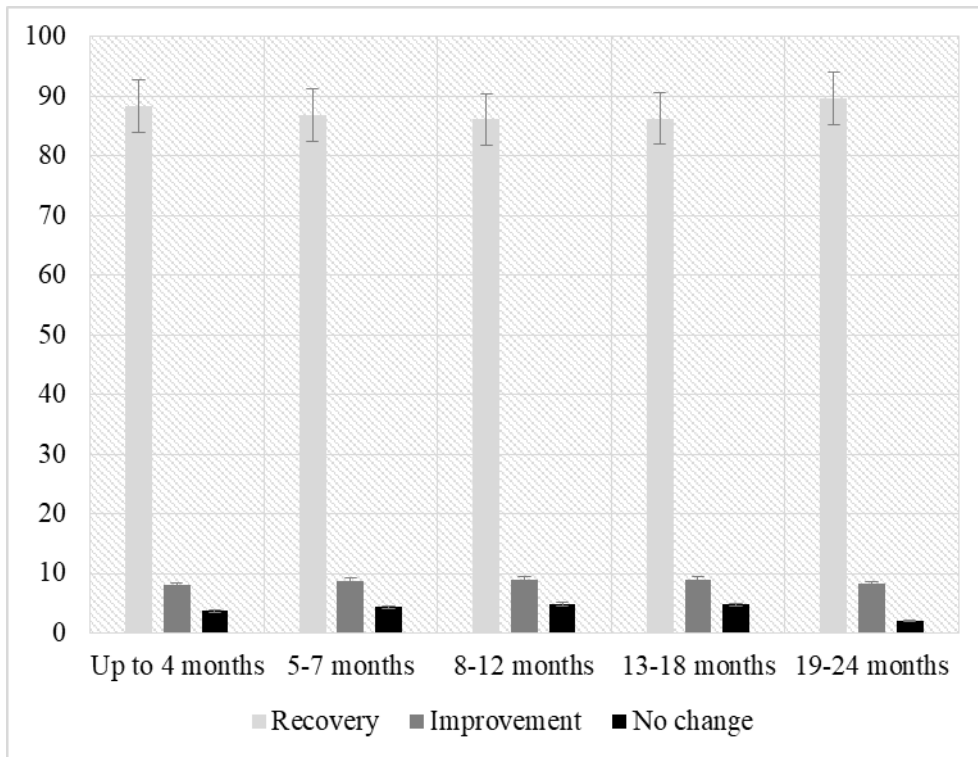


Figure 2. Patient treatment outcomes

With a duration of 5-7 months of surgical therapy, the results confirm recovery in 88.8 % ( $p < 0.01$ ) of patients, improvement in 5.0 % ( $p < 0.01$ ), and without significant changes in 6.2 % ( $p < 0.01$ ). After 8-12 months, these parameters were 85.9, 9.1 and 5.0 % ( $p < 0.01$ ), respectively. As a result of complex treatment after 13-18 and 19-24 months, recovery was registered in 86.5 and 89.6 % ( $p < 0.01$ ), improvement - 9.3 and 8.5 % ( $p < 0.01$ ), and unchanged - in 4.2 and 1.9 % ( $p < 0.01$ ), respectively.

So, the long-term results of observation of 200 patients with compression syndrome of the celiac trunk, subjected to surgical therapy, during the period from one year to 2 years showed recovery - in 87.5 % ( $p < 0.01$ ), improvement - in 7.4 % ( $p < 0.01$ ), and without significant changes - in 5.1 % ( $p < 0.01$ ).

## Conclusions

1. Dunbar syndrome is more common in girls, which in 75 % of cases is accompanied by signs of chronic abdominal ischemia. Significant and prolonged stenosis of the celiac trunk can cause the development of a number of complications (pancreatic-duodenal artery aneurysms, etc.).
2. Pathophysiological changes against the background of celiac trunk compression syndrome were characterized by regional hemodynamic disorders – the flow of blood flow from the superior mesenteric artery into the CT basin, the degree of which depends on the degree of diameter of the stenosed segment.

3. Changes in the rheological features of blood confirm their pathogenetic significance in the formation of morphological disorders of the upper gastrointestinal tract. The intensity of these changes is directly associated with the degree of compression and duration of pathology.
4. The clinical symptoms of Dunbar syndrome manifest themselves in three forms - painful, dyspeptic, and neurovegetative.
5. Ultrasound Doppler scanning of the abdominal aorta, the celiac trunk and the superior mesenteric artery seems to be a fairly reliable and accurate way to diagnose the compression syndrome of the CT and its treatment, with an efficiency of 98.5 %.
6. Early diagnosis and timely surgical intervention of Dunbar syndrome contribute to achieving complete recovery in 87.5% ( $p < 0.01$ ) of pediatric patients, significant improvement – in 7.4% ( $p < 0.01$ ) of patients, and unsatisfactory results – in 5.1% ( $p < 0.01$ ).

### **Conflict of interest**

The authors declare no conflict of interest and the presence of a financial interest in the submitted material.

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