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Особенности физического и моторного развития детей с врождёнными пороками сердца

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АННОТАЦИЯ

Введение. Для детей с врождёнными пороками сердца (ВПС) характерны особенности развития, в основе которых лежит патология сердечно-сосудистой системы. Тип, тяжесть ВПС, возраст коррекции порока и ряд других факторов оказывают непосредственное влияние на выраженность его не кардиологических осложнений, таких как нарушения физического и моторного развития, обусловленных особыми условиями формирования нервной системы в условиях ВПС.

Цель. Систематизировать имеющиеся в настоящее время данные об особенностях физического и моторного развития детей с ВПС, о лежащих в их основе факторах, обосновать важность более углублённого обследования детей на всех этапах их развития и необходимость коррекции выявленных нарушений с момента их диагностики.

В обзоре представлены данные о распространённости и особенностях отклонений в моторном развитии у детей с ВПС в разные возрастные периоды; описаны факторы, предрасполагающие к их развитию, а также возможности диагностики и коррекции таких нарушений. Кроме того, приведены сведения о развитии функции равновесия, которая также страдает у данной когорты пациентов. Так как физическое и моторное развитие детей тесно связаны между собой, с развитием нервной и состоянием мышечной систем, а сила мышц и равновесие являются неотъемлемыми компонентами некоторых моторных навыков, каждому из этих параметров посвящен отдельный раздел статьи. При ацианотических пороках чаще выявляются нарушения в прибавке массы тела, а при цианотических — отставание детей в росте и массе тела. Ещё одним проявлением задержки физического развития детей с ВПС является дефицит развития мышечной массы, что отражается в подтверждённом исследованиями снижении силы мышц у данной категории пациентов. Нарушения в мелкой и/или крупной моторике отмечается у ⅓–⅔ детей с ВПС с раннего возраста до совершеннолетия. Нарушения в двигательной сфере в детском возрасте влекут за собой гиподинамию, психологические и когнитивные проблемы, трудности в социализации ребёнка, значительно снижая качество жизни.

Заключение. Для детей с ВПС характерны особенности физического и моторного развития, которые оказывают существенное влияние на жизнь ребёнка. Поэтому в настоящее время крайне остро стоит необходимость обследования не только сердечно-сосудистой системы, но и физического развития, двигательной сферы у детей с ВПС, своевременного выявления отклонений в физическом и моторном развитии и их целенаправленной реабилитации.

Ключевые слова: *врождённые пороки сердца; дети; моторное развитие; физическое развитие; крупная моторика; мелкая моторика; равновесие; мышечная сила; реабилитация*

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Features of Physical and Motor Development of Children with Congenital Heart Diseases

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ABSTRACT

INTRODUCTION: Children with congenital heart diseases (CHD) have certain peculiarities of development determined by the pathology of the cardiovascular system. The type, severity of CHD, the age of correction of the disorder and a number of other factors have a direct impact on the severity of its non-cardiologic complications, such as disorders in physical and motor development attributable to specific conditions of formation of the nervous system in children with CHD.

AIM: To systematize the currently available data on the peculiarities of physical and motor development of children with CHD and their underlying factors, to justify the importance of a more in-depth examination of children at all stages of their development and of the need to correct the identified disorders starting from the moment of their diagnosis.

The review presents the data on the incidence and peculiarities of deviations in motor development of children with CHD at different age periods; describes factors predisposing to their development, as well as the possibilities of diagnosis and correction of such disorders. Besides, information is provided on the development of the balance function that is also impaired in this cohort of patients. Since the physical and motor development of children are closely interrelated and related to the development of the nervous and muscular systems, and the muscle strength and balance are integral components of some motor skills, each of these parameters is considered in a separate section of the article. Acyanotic heart diseases are more commonly associated with disorders in the weight gain, and cyanotic ones — with retardation of children in growth and body mass. Another manifestation of retardation of physical development of children with CHD is deficit of muscle mass development reflected in the research-confirmed reduction of muscle strength in this category of patients. Disorders in fine and/or large motor skills are noted in one to two thirds of children with CHD from an early age to adulthood. Disorders in the motor sphere in childhood entail physical inactivity, psychological and cognitive problems, difficulties in socialization significantly reducing the quality of life.

CONCLUSION: Children with CHD are characterized by peculiarities of the physical and motor development, which have a significant impact on the child's life. Therefore, currently of paramount importance is not only examination of the cardiovascular system of children with CHD, but also their physical and motor development, as well as timely identification of deviations in the physical and motor development and their targeted rehabilitation.

Keywords: *congenital heart diseases; children; motor development; physical development; large motor skills; fine motor skills; balance; muscle strength; rehabilitation*

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ECO•VECTOR

LIST OF ABBREVIATIONS

AC — artificial circulation
CHD — congenital heart disease
HLHS — hypoplastic left heart syndrome
SD — standard deviation
SV — single ventricle
TGA — transposition of the great arteries

INTRODUCTION

Congenital heart diseases (CHDs) are identified in approximately 9 newborns per 1 thousand, which makes 1.35 million annually worldwide [1], and about a quarter of patients require surgical treatment in the first months of life [2]. Taking into account the increased survival rate of children with CHDs compared to the past century, with the majority of them reaching adulthood, correction of non-cardiologic complications of CHDs including disorders in motor development, is becoming increasingly urgent.

Children have a natural need for physical activity. The child's motor activity contributes to his general development at an early age and his becoming an adequate member of society at an older age. Children with CHDs are characterized by developmental retardation, the severity of which depends on the initial severity of the defect and the presence of genetic diseases, which are detected in 30% of children with CHDs [3]. In addition, patients with CHDs are referred to the category of increased risk of brain damage due to the peculiarities of intrauterine development, postnatal effects of hypoxia and intraoperative factors (for example, use of artificial circulation (AC)), which entails deviations in neuromotor development. Features of hemodynamics and metabolism, medication intake, hypoxemia and a number of other factors associated with CHDs, also affect the physical development of children. This concept combines the process of growth, weight gain, development of the muscular system and other organ systems, which, in turn, depends on the maturity and functional abilities of the nervous system.

The **aim** of this study to systematize the available data on the peculiarities of the physical and motor development of children with congenital heart defects and on the underlying factors, and to justify the importance of a more in-depth examination of children at all stages of their development and the need to correct the identified disorders from the moment of their diagnosis.

Since the physical and motor development of children are closely related to each other and to the development of the nervous and muscular systems, and muscle strength and balance are integral components of some motor skills [4], each of these parameters is described in a separate section of the article.

Physical development of children with CHD

According to the literature, patients with congenital heart diseases are characterized by a lag in height and body weight, which is associated with malnutrition [5, 6]. Stunting in children with CHDs is associated with tissue hypoxia, increased metabolism, impaired absorption of nutrients, low concentrations of insulin-like growth factor-1, decreased cardiac output, pulmonary hypertension, repeated respiratory infections, intake of medical drugs, and genetic characteristics [5]. Growth disorders in infants with acyanotic heart diseases are believed to be proportional to the severity of hemodynamic disorders, and the presence of cyanosis in infants with a heart disease exacerbates the evidence of malnutrition: according to statistics, 'pale' defects are to a larger extent associated with disorders in the weight gain, whereas children with 'blue' CHDs are more likely to lag behind in growth and body weight [5]. Another important factor is nutrition. Mothers of children with CHDs often complain of problems with feeding due to poor appetite or refusal of food by the child [7]. It should be noted that difficulties with feeding of a child with a CHD may be not only a sign of heart failure, but also of a concomitant neurological pathology [8], which may be a consequence and/or be aggravated by tissue hypoxia in conditions of disordered hemodynamics.

One of the first publications on this problem was J Benn's data on the height and body weight of school-age children with the open arterial duct: according to statistics, the body weight of boys and girls was lower than that of their peers, and at the same time, girls were statistically significantly higher than their peers [9].

M Campbell, et al. analyzed 200 patients (0–35 years; 88.5% — children aged 0–14 years) with various CHDs, with half of them having Fallot's tetralogy. Scientists have found out that the more severe cyanosis, to a more extent a CHDs is reflected on the patient's height; the mean height in the group of acyanotic defects made 98% of normal values, and 94% in the group of 'blue' defects. The scientists showed that CHDs mostly affect the body mass of patients. Thus, the average body mass in the studied cohort made 85% of the average norm. At the same time, the severity of cyanosis was inversely proportional to the body mass: in 'pale' and severe 'blue' defects the body mass made 91% and 77% of the average norm, respectively. It was also found that with patients getting older, the body mass increasingly becomes dependent on the age and less on the height. It is noteworthy that in patients with Fallot's tetralogy, deviations from the norm in both height (97.1% vs. 94.5%) and body weight (87.2% vs. 78.3%) were significantly less pronounced than in other cyanotic CHDs, in which connection the authors concluded that the lag in the development of children in CHDs is not only due to cyanosis [10].

In the literature of the mid-XX-century, a number of assumptions were made about other probable causes of deviations in the physical development of children with CHDs. H Taussig in her book 'Congenital Heart Diseases', first published in 1947, noted that better physical development of children with Fallot's tetralogy may be due to the absence of overload of the pulmonary circulation, which is noted in an unoperated open arterial duct, atrial septal defect, or transposition of the great arteries (TGA) [11]. It has also been noted long ago that, besides the lack of subcutaneous fat, children with CHDs are characterized by very poorly developed muscle mass, which is aggravated by the deficit of normal physical activity and cannot but affect the child's body weight [10].

Despite commonly timely correction of CHDs in recent years, the problem of lagging in the physical development of children with CHDs still remains relevant. In a Chinese study of the beginning of the XXI century, when comparing preschoolers with CHDs and their healthy peers, statistically significant differences in height and body weight were revealed: children with CHDs were slightly shorter and significantly lighter than their peers. Severe physical retardation (height and body weight below the 3rd percentile) was observed in 13% of children with CHDs and in 1% of healthy children [12]. C Chien, et al. when comparing children of early and preschool age with CHDs with healthy children of the same age, also noted a lower weight at birth, lower height/age and body weight/age ratios in the group with heart diseases [7]. And in a recent study of young children with CHDs, S. Maya, et al. found a deficit of body weight (less than -2 standard deviations (SD)) and height (less than -3SD) in 42.3% and 38.5% of children with 'blue' defects, respectively, and in 11.5% of children with 'pale' defects in both parameters [6]. C Rego, et al. found malnutrition in 6.3% of children with CHDs aged 6–15 years by the weight/age index, and in 18.5% by the ratio of body mass index to age. Low height was noted in 15.6% of the examined children [13].

Muscle strength

A deficit in the development of muscle mass in children with CHD cannot but affect muscle strength. The results of studies on this topic confirm a decrease in muscle strength in children with CHDs. Thus, in schoolchildren with heart diseases, the strength of femoral quadriceps and of the hand muscles was 21.4% and 17.7% lower than in the control group [4]. In a study comparing the muscle strength of the hand in 569 children with CHDs with 2,551 healthy children, it was found that this parameter in the main group was significantly lower than in the comparison group (20.8 kg vs. 24.5 kg). Scientists also found that in children with complex CHDs, the lowest values of hand muscle strength were 19.8 kg, with moderate CHDs — 20.7 kg, with simple CHDs — 22.5 kg [14]. Thus, the degree of cyanosis in CHDs is reflected not only on the weight and height of the child, but also on the development of muscle strength. According to recent data, children with complex CHDs, especially with single-ventricle defects, are at increased risk of reduction of muscle strength [15], which was confirmed by M Meyer, et al. who revealed the lowest muscle strength parameters in the group of children after Fontan surgery [14].

In a Brazilian study including 32 children with CHD from 6 to 15 years old, a decrease in hand muscle strength was found in 96.9% of patients. Due to the fact that the authors revealed a low functional reserve of muscles in 43.8% of children with muscle strength below normal, they assumed that this cohort of patients may have an increased risk for development of sarcopenia [13]. The data of a systematic review and meta-analysis published in October 2022, confirmed that children and adolescents (under 18 years of age) with CHD have reduced muscle strength compared to the control group (-34.07 N•m), and although the meta-analysis did not reveal any significant difference in hand muscle strength between these two groups (0.08 N•m), it showed a decrease in muscle strength of limbs in the group of children with CHDs. The authors also noted that patients with CHDs are characterized by lower strength of respiratory muscles [16].

There is no doubt that physical activity is impossible without muscle work. Regular use of muscle mass in physical activity leads to an increase in both muscle strength and endurance and physical performance, however, it should be borne in mind that the strength of the hand muscles is poorly related to the physical performance of the patient [14], therefore these parameters require separate assessment and correction.

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Motor development

The work of the muscles of the body provides voluntary motor activity. Motor development determines the accuracy and speed of motor activity, and, consequently, the achievement of its aim. Motor skills are a set of coordinated human actions aimed at performing precise movements.

Lagging in motor development is considered one of the main problems in the development of children [5]. Motor disorders can have a negative impact on various areas of a child's life: on the daily activities (for example, dressing, eating), participation in games with other children, school activities, sports, the child's self-esteem and his position in peer society. All this can lead to a sedentary lifestyle in the future and exacerbate health problems [4].

Over the past two decade, scientists have been paying increasing attention to the development of motor skills in children with congenital heart defects, although as early as in 1949, M Campbell, et al. pointed to the lag in walking skills in this cohort of patients [10]. At the moment, a delay in motor development is known to be typical for 30%–60% of children with CHDs [8,17] in approximately equal proportions at different age periods [17].

According to statistics, impaired motor skills in children with CHD are detected from an early age. In long-term studies, the lag in the development of gross motor skills in this category of children is noted from the age of 2 months [18]. According to the data published by S Fourdain, et al. 79% of infants with CHDs at the age of 4 months lag in motor development and need restorative treatment [18].

Without correction of motor disorders in infancy, especially in the case of a severe CHD, the problem continues to the early childhood. The results of a data analysis of 294 children with CHD showed that only 52% of them learned to walk by the age of 18 months, and 73% by the age of 2 years. In this connection, the researchers concluded that cyanosis/decreased blood saturation with oxygen and poor development of muscle mass, have a direct effect on the time of starting walking. Thus, in the group with 'pale' defects, 76% and 92% of children learned to walk by the age of 1.5 and 2, whereas in the cohort of 'blue' defects only 47% and 69%, respectively [10]. According to recent data, the late onset of walking even in healthy preschool children is a predictor of disorders in such parameters of motor development as fine motor abilities and balance [19].

A number of studies have been devoted to the examination of children with CHDs of preschool age. According to A Nasiruzzaman, et al. 60% of children with heart defects (n = 50) aged 3.04 (± 2.01 SD) years had a lag in the development of gross motor skills, and 54% had disorders of fine motor skills [5]. A Swiss study included 233 children with CHDs, 64 of them with a confirmed genetic disease, who underwent surgery with application of AC. The mean age of the studied cohort was 6.3 years (5.1–6.8 years). Comparison of the main parameters of motor development of children with CHD without genetic pathology with the norm showed statistically significant disorders in motor development ($p < 0.01$), to this end, pronounced deviations in motor skills were noted with a frequency of 21.2%–41.1% (depending on the studied skill) [20]. A recent study by C. Chien, et al., also revealed a lag in children with CHDs under 6 years of age in general motor development with the most pronounced disorders in gross motor skills [7].

In the international literature, problems with motor function have been reported for older children as well. According to S Dordel, et al. 75% of children with cyanotic and 42% of children with acyanotic CHDs of school age had moderate (25% and 26.3%, respectively) or severe (50% and 15.8%, respectively) motor development disorders [21]. According to the results of another study, after surgical correction of TGA in preschool and school-age children, deviations in gross and fine motor skills were noted in 23.4% and 22.1% of cases, respectively [22]. When comparing children with CHDs aged 7–17 years with healthy peers, R Liamlahi, et al. obtained statistically significantly worse results in all areas of motor development in the main group ($p \leq 0.001$) with the most pronounced deviations in gross motor skills ($p < 0.001$). Scientists also noted that 54% of children with deviations in motor function had behavioral disorders [23]. A group of researchers led by I. Holm found motor disorders of varying severity in children with CHDs from 7 to 12 years in 42.5% of cases [4].

Motor development disorders detected at an early age are unlikely to level out on their own with age [4]. This is confirmed by the above-mentioned studies, where motor disorders were detected in children with CHDs of different ages without previous rehabilitation, as well as by a study of B Bjarnason–Wehrens, et al. in which more pronounced motor skills disorders were recorded in older children (11–15 years versus 5–10 years; $p < 0.01$) [24]. A survey of adolescents with CHDs conducted by K Easson, et al. showed that 42.4% of them had moderate to severe motor development disorders (less than -1SD), and 18.2% had severe motor development disorders (less than -2SD) [25], which also confirms the persistence of this problem in adolescence.

Nevertheless, a number of authors report normalization of motor parameters in children with CHDs without genetic pathology by the age of 12 months and older [18, 26]. However, in these studies, of attention is the fact that from 74% to 100% of children with developmental delay received rehabilitation treatment, which confirms the fact that timely detection and treatment of such disorders contributes to a significant improvement in the prognosis of motor development in children with CHDs.

The authors of the systematic review concluded that severe motor disorders (less than -2SD) are characteristic mainly of younger age, which suggested that motor skills may improve as a child with a CHD grows up [17]. This corresponds to the data that motor abnormalities are most often (4%–74%) detected before the age of 12 months, then their frequency gradually declines to 3.5%–6.7% at 24–36 months (but remains higher than the population average) [27]. However, opposite results were obtained in a long-term study by M Sprong, et al. most children with impaired gross motor skills (≤ -1SD) at the age of 9 months had the same (≤ -1SD) or more severe problems with motor skills (≤ -2SD) by 18 months [2].

Despite the variability in the results of examinations of younger children, the frequency of lag in motor development increases by the school age, which may be associated with increasing requirements to the child, as well as with the complexity of the necessary motor skills [25]. A systematic review by M Bolduc, et al. confirms that children and adolescents with CHDs requiring open-heart surgery are characterized by deviations in motor development by ~ 1SD for the worse compared to healthy peers, and without rehabilitation treatment children do not 'outgrow' these disorders [17].

The degree of motor function disorders is largely predetermined by the severity of a congenital heart disease. J Stieh, et al. revealed a significant deficit in the development of gross and fine motor skills in children with cyanotic CHDs, and not in children with 'pale' defects [28]. According to S Dordel, et al. motor development disorders were 1.8 times more common in children with 'blue' CHDs [21].

The most severe CHDs are single-ventricle (SV) defects. According to a systematic review and meta-analysis, in children with single-ventricle CHDs, development parameters were expectedly lower than in those with two-ventricle circulation [15]. Other authors, when comparing 9- and 18-month-old children, revealed statistically significantly poorer results ($p \leq 0.05$) in gross and fine motor skills in children with single ventricle compared with peers with other complex CHDs. And when evaluated in dynamics, the rate of severe disorders of gross motor skills (less than 2SD) in children with single-ventricle circulation, increased with patient's age: at 3, 9 and 18 months, it was 8%, 30.6% and 32.2%, respectively [2]. According to M Ricci, et al. motor disorders in children with single- and two-ventricle critical CHD at the age of 4.5 years are detected in 32.4% and 11.8% ($p < 0.001$), respectively [29].

A number of articles focus on the features of the motor development of children with various CHDs with a single ventricle. D Williams, et al. found that children after Fontan surgery for hypoplastic left heart syndrome (HLHS) are characterized by pronounced motor development disorders [30]. These data are confirmed by A Sarajuuri, et al. who compared 30-month-old children with HLHS with children with other single-ventricle defects of the same age: patients with HLHS had significantly worse parameters of motor development [31]. However, O Khalid and T Harrison revealed a lag in motor development in 55% of children with single-ventricle circulation without HLHS and in 33% of children with HLHS at the age of 12 months, but the differences were statistically insignificant [32].

Since the parameters of motor development in children with SV are significantly worse than in children with two-ventricle CHDs, assumptions can be found in the literature about children with simpler (two-ventricle) heart defects gradually 'outgrowing' their initial developmental disorders [15]. As discussed above, this issue remains controversial, however, one of the factors that may affect the development of motor skills is the duration of hypoxemia [28], which persists throughout life in children with one ventricle, despite surgical interventions.

Surgical correction of CHDs reduces or levels off the hypoxemic condition of the patient, so it can have a beneficial effect not only on the cardiovascular system, but also on the overall development of the child, including motor skills. Thus, J Stieh, et al. noted that after surgical treatment of 'blue' defects, the number of patients with moderate and severe motor dysfunctions decreased from 39% to 21% and from 46% to 33%, respectively [28]. It is important to note that children who underwent surgery under 2 years of age, had significantly higher motor development parameters than children operated on at a later age. According to B Bjarnason–Wehrens, et al. motor disorders were more pronounced in children who underwent an open-heart surgery compared with children with CHDs who did not require any treatment or underwent endovascular intervention [24]. Both examples confirm the assumption that the less pronounced hypoxia and/or the shorter the period of exposure to hypoxia, the less the neuromotor function is affected, and there is a higher chance for a child to recover from the existing abnormalities. However, even in this case, everything is not that unambiguous, since C Limperopoulos, et al. found no improvements in the cohort of children studied. They published data of 42% of children with CHDs ($n = 131$) having a lag in motor development (gross and/or fine motor skills) within 12–18 months after the open heart surgery [33]. After 5 years, these disorders were detected with the same frequency (according to a re-examination of 94 children) [34]. Nevertheless, the most reasonable seems to be the opinion of B Bjarnason–Wehrens, et al. that timely correction of CHDs creates the optimal conditions for successful 'catching up' with growth and normalization of physical development [24].

The problem of motor development in children with CHDs becomes even more relevant when comparing this group of children with healthy peers. In Norway, the data from a survey of 120 children from 7 to 12 years old who underwent several surgical interventions in the first year of life for complex CHDs were compared with a control group of 387 healthy peers. In the CHD group, 16.7% of children with awkward movements were identified, 25.8% had severe motor disorders, whereas in the comparison group these indicators were 4.9% and 2.3%, respectively. Scientists have calculated that the risk of motor disorders of any degree in children with CHDs is 5.8 times higher, and of severe motor problems 11 times higher [4]. In another study, which compared children with CHDs from 5 to 15 years of age with a control group of healthy children, almost 3/5 of the sample of the main group showed disorders in motor development, a third of children with CHDs had severe problems with gross motor abilities [24]. Chien C, et al. published the data on the average z-scores of gross, fine motor skills and general motor development: they appeared negative in children with CHDs up to 6 years old compared with healthy peers, with gross motor skills being most affected. Scientists also found that a shorter duration of hospitalization and a higher body weight/age ratio were characteristic of better development of gross motor skills [7].

An analysis of the literature on motor development in children with CHDs showed that a number of studies have revealed disorders in both gross and fine motor development [2, 4, 5, 7, 20, 22, 23, 28, 33], and often the authors did not differentiate which motor skills suffered. However, according to a meta-analysis of 2021, this cohort of children is characterized by more pronounced disorders in gross motor abilities [27]. M Sprong, et al. (2022) did not find (0%) significant problems (≤ -2SD) with fine motor skills in children with CHDs at the age of 18 months, whereas severe disorders of gross motor skills were observed in 12% of children of this age [2]. Thus, more research is required with an emphasis on what motor skills are disordered in a particular patient, since this will determine the management tactics for the child and the range of potential difficulties in the future for him in the absence of timely correction.

Balance

Balance is the main component of motor skills [29], as it helps stabilize the body and exercise movement control. The balance can be static and dynamic. The difference is in the position of the body in which the posture is maintained and controlled: standing or sitting in the first case, in motion in the second.

A very small number of articles describe the state of balance in children with CHDs. According to I Holm, et al. the static balance index measured in 120 children with CHDs aged 7–12 years turned out to be 27.5% worse than in 387 healthy peers [4]. In the study by C Rego and C Sabino Pinho, the data of children with CHDs aged 7–17 years and of the control group were analyzed, and the most pronounced disorders in the main group were found in the static balance ($p < 0.001$) [13]. According to the results of a Swiss study, the same body balance component was significantly ($p < 0.01$) worse in children with CHDs of 10.4 ± 2.5 years old [35]. At the same time, N Naef, et al. pointed out in their work that for children with CHDs, most difficult turned out to be the maintenance of dynamic balance [20].

It is expected that when comparing patients with critical CHDs, children with single-ventricle circulation showed more difficulties when performing tests for balance compared with children with two-ventricle defects ($p = 0.001$) [29].

DISCUSSION

As follows from the review of scientific publications, the problem of disorders in the physical and motor development of children with CHDs remains relevant up to date. Motor disorders are identified on average in 6%–10% of children's population [4], about 5%–6% of children suffer dyspraxia [36], while the cohort of children with CHDs is characterized by a more pronounced and frequent delay in motor development.

Each child is individual, but it is obvious that children with CHDs have much more factors predisposing for the formation of neuromotor disorders than their healthy peers. Such factors include:

- preoperative factors (intrauterine circulatory disorder [3], structural abnormalities of the brain [4], acidosis [37], duration of hypoxemia [28], extracardiac concomitant pathology);
- intraoperative factors (duration of AC [4, 20, 35, 37], intraoperative seizures [3], repeated operations [27]);
- postoperative factors (hyperlactatemia in the first 5 days after surgery [29], acute cerebrovascular accident [3], seizures [8, 20], low cardiac output [4], restriction of physical activity [18], duration of stay in the intensive care unit [20], duration [35,38] and the frequency of hospitalizations [38], muscular hypo- or hypertonia [8], the need for extracorporeal membrane oxygenation, installation of an artificial heart ventricle);
- unmodified factors (a genetic disease, gestation age, prematurity, low birth weight [38], low body weight and head circumference at birth [20], severity of a CHD, blood saturation with oxygen below 85% [27]);
- social (overprotection, restrictions on physical activity and communication with peers [12]);
- socio-economic (family income [38]).

A number of authors dispute the importance of certain factors in the development of neuromotor disorders. For example, M von Rhein, et al. believe that intraoperative factors play a less important role in the consequences for a child with a CHD than postoperative complications [35]. However, it should be taken into account that the risk factors for disorders in neuromotor development at an early age may differ from those in the long-term period [20]. Thus, if in the early postoperative period, especially in the early childhood, the most important are postnatal factors and factors related to surgery, then at an older age and/or several years after surgery, social factors and features of children's physical development will come out on top.

There is evidence that even in healthy children, deviations from the norm in motor development have a significant impact on psychological, emotional and cognitive development [36]. Children with impaired gross motor abilities may seem awkward, have balance difficulties, and are clumsy in daily physical activity [4], which may cause them to have problems interacting with peers, for example, during games, which may lead to behavioral problems. In addition, overprotection on the part of parents and teachers may limit the child's participation in situations of social interaction in which certain behavioral and motor skills could be formed, which further aggravates the situation. There is also evidence that children and adolescents with dyspraxia have low self-esteem and a higher level of anxiety compared to their peers [39]. Uncorrected motor disorders can have long-term consequences related to daily activities, self-determination and general well-being [17]. Moreover, it is important to separate the assessment of gross and fine motor skills [2], because problems with gross motor skills lead to a sedentary lifestyle and social isolation from peers, and disturbance of fine motor skills cause learning difficulties. Children with CHDs often cannot cope with everyday tasks using fine motor skills, for example, turning the pages of books in early childhood, tying shoelaces and buttoning in preschool age, which in turn can lead to difficulties with the formation of reading and writing skills and subsequent lag in mastering the school curriculum [40].

In their work, S Fourdain, et al. identified a modifiable factor of lag in neuromotor development which is early stimulation to the formation of motor skills, which was confirmed by the results of their study [18]. In addition, ergotherapy, physical therapy and classes with a psychologist can help, which proved effective in children with dyspraxia who had similar disorders in motor development as children with CHD [17]. It is extremely important to encourage children with motor disabilities to be physically active, to acquire experience of participating in a variety of activities, communicate with healthy peers and not feel anxious about the success of doing anything.

According to M Unverdorben, et al. children with CHDs, regardless of the severity of un underlying disease, who were exempt from physical education classes at school, had significantly more pronounced problems with motor function compared with children who attended these classes [41]. However, according to B Bjarnason–Wehrens, et al. physical education classes at school cannot compensate for motor disorders, which most likely stem from the preschool age, because 92.3% of children with CHDs in the study group attended physical education classes, and more than half of them had pronounced problems with motor skills [24]. Therefore, it is extremely important to involve children with CHDs not only in sports activities at school (in the absence of contraindications), but also to organize additional specialized training for them aimed at improving motor skills. According to the available research results, children who begin regular physical activity under the supervision of specialists show evident positive dynamics after a few months. The participation of children with CHDs in a program aimed at improving psychomotor development within 8 months or in a specialized swimming program within 3 months led to a decrease in the proportion of children with motor disorders from 54.8% to 29.0% [37].

The American Association of Cardiology and the American Academy of Pediatrics identify the following groups of children with CHDs who have a high risk of developmental delay (Class I, evidence level A) [3]:

1) newborns and infants requiring open heart surgery (cyanotic and acyanotic defects, for example, HLHS, TGA, tricuspid valve atresia, etc.);

2) children with other types of cyanotic heart defects that do not require open heart surgery during neonatal period and infancy (for example, some types of Fallot's tetralogy, Ebstein anomaly);

3) children with factors aggravating CHDs, such as prematurity, extracorporeal membrane oxygenation and cardiopulmonary resuscitation in history, intraoperative seizures, organic brain changes, etc.

According to the document compiled by these organizations, for developmental delay to be excluded, such children should be examined at the age of 12–24 months, at 3 to 5 years and at 11 to 12 years (class IIa, evidence level C) [3].

Given the relevance and prevalence of motor disorders among children with congenital heart diseases, medical professionals should conduct screening examinations of children with CHDs for the neuromotor development from an early age in order to timely identify patients with developmental disorders and their timely correction. There are special scales for assessing motor development, but there is no universal method of examining children that could be applied from the period of neonatality to adulthood. This is due to the physiologically predetermined period of mastering certain skills, therefore, when examining children of different ages, different tools are used and different skills are evaluated. A number of scales are recommended for the examination of fine and gross motor skills in children with CHDs [3], however, it should be borne in mind that if the assessment of fine motor skills does not imply a significant load on the cardiovascular system, the examination of gross motor skills may have a number of limitations in children with severe defects. For example, the Bronix-Oseretsky-2 scale (BOS-2) recommended by the American Association of Cardiology and the American Academy of Pediatrics for assessing fine and gross motor skills in children with CHD [3], which is often used in children, includes tests for shuttle running, push-ups and presses, which are extremely undesirable for performing by children with severe heart defects. In this regard, when choosing diagnostic scales, it is necessary to take into account not only the age for which the scale was developed, but also its safety for patients with residual changes. It is known that growth and neuromotor development are closely interrelated [7], therefore, weight and height characteristics and the development of the muscular system should also be taken into account when examining this cohort of children.

Thus, a more thorough examination of children, including school-age children and adolescents, is required for the presence of developmental abnormalities, including motor skills, since older children can adapt and compensate for their disorders, and the difficulties they face may be invisible to an outsider without a targeted examination. However, it should be taken into account that severe developmental delay is not typical for children with CHDs, therefore, if a significant delay is detected, a more careful examination is required to exclude other probable causes [10]. Thus, in most published works on motor disorders in children with heart defects, children with mental disorders and genetic syndromes, which often cause more pronounced deviations, were excluded from the studies.

Realizing the importance of non-cardiologic complications in children with CHDs, in 2019 in Switzerland, data collection began for the Outcome Registry for Children with Severe Congenital Heart Disease (ORCHID) [42], which plans to include 80-100 children annually. Children with severe CHDs who need surgical treatment of the defect within the first 6 weeks of life, will undergo assessment of the neuromotor development at the age of 9–12 months, 18–24 months and 5.5–6 years, after which the obtained data will be analyzed to determine risk factors and protective factors of neuromotor disorders in this cohort of children.

At the moment, there are very few prospective long-term studies of the motor development of preschool, school and adolescent children with complex CHDs. It is necessary that more research be conducted on the characteristics of the motor development of primary school children and adolescents with CHDs, since there is still no reliable information on the degree and severity of motor disorders at school age.

Summarizing the above, it should be emphasized that it is extremely important that rehabilitation programs for children with CHDs include examinations of the motor sphere and methods of correction of detected disorders. Physical rehabilitation programs for children with CHDs should be aimed not only at increasing exercise tolerance, but also at improving control of their own body through the development of balance function, muscle strength and motor skills using individually selected specialized exercises. This can be achieved only through a comprehensive approach to a child with congenital cardiologic pathology, not limited only to cardiorehabilitation.

CONCLUSION

The analysis of the literature has shown that children with congenital heart defects are characterized by features of physical and motor development, which have a significant impact on the child's life and which must be taken into account during the examination and rehabilitation of this category of patients. The development of motor function contributes to the independence and self-sufficiency of the child, and therefore to the improvement of his quality of life. Therefore, at present there is an extremely urgent need to examine not only the cardiovascular system, but also the physical development and motor sphere in children with congenital heart defects, as well as the development of methods for their timely correction.

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