THE CURRENT VIEW ON THE BODY MASS AND BODY MASS INDEX OF CHILDREN WITH SPINE DEFORMITY DUE TO CEREBRAL PALSY: A SYSTEMATIC REVIEW

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Background. Currently, several authors believe that one of the main risk factors for the development of postoperative complications after surgical correction of scoliosis due to cerebral palsy (CP) is the body weight, changes in it, and the body mass index (BMI). However, a unified approach for the analysis of these indicators in children with CP remains unclear.

Aim. Analysis of the available data in the modern literature on the issues of body mass and BMI in children with spine deformity due to CP.

Materials and methods. A systematic search of the literature was conducted in open electronic databases of the scientific literature PubMed, Web of Science, Scopus, MEDLINE, eLIBRARY, Russian Index of Scientific Citation (RISC), and bibliography of key articles. The criteria of inclusion were systematic reviews, meta-analyses, multicenter studies, controlled cohort studies, uncontrolled cohort studies of children with spine deformities due to CP, and age of the CP patient <20 years. The criteria of exclusion were clinical cases, observations, materials of conferences, the patient’s age >20 years, and neuromuscular scoliosis of another etiology.

Results. The review primarily included 156 articles with the publication date of 1990–2020. Among them, 25 publications met the following criteria of inclusion: 3 systematic reviews and meta-analysis, 3 population studies, 1 multicenter study, 11 controlled cohort studies, 6 uncontrolled cohort studies, and 1 case-control study.

Conclusion. Body mass and BMI correlate with the functional activities of children with scoliosis due to CP. GMFCS stratified growth graphs of children with CP are the most appropriate reference indicators for assessing body mass and BMI of children with CP. Underweight body and low BMI (below the 10th percentile) are important factors that contribute to high risk of complications after scoliosis surgical correction. In the future, it will be necessary to develop national special centile tables for the optimal assessment of the anthropometric indicators in children with CP.

Keywords: cerebral palsy in children; the spine deformities; body mass; body mass index.
Обоснование. Многие авторы считают, что одним из факторов риска осложнений после хирургической коррекции сколиоза вследствие детского церебрального паралича является масса тела, ее изменения, индекс массы тела. Однако однозначного подхода к анализу данного показателя у детей с церебральным параличом в настоящее время не сложилось.

Цель — анализ данных в современной литературе по вопросам массы тела, индекса массы тела у детей с деформацией позвоночника вследствие церебрального паралича.

Материалы и методы. Системный поиск литературы проведен по базам данных PubMed, Web of Science, Scopus, MEDLINE, eLIBRARY, РИНЦ, библиографии ключевых статей. Критерии включения: систематические обзоры, метаанализы, мультицентровые исследования, контролируемые когортные исследования, неконтролируемые когортные исследования детей с деформациями позвоночника вследствие церебрального паралича, возраст пациентов с детским церебральным параличом <20 лет. Критерии исключения: клинические случаи, наблюдения, материалы конференций, возраст пациентов >20 лет, нейромышечный сколиоз другой этиологии.

Результаты. В обзор первично было включено 156 статей, из них критериям включения соответствовали 25 публикаций: 3 систематических обзора и метаанализ, 3 популяционных исследования, 1 мультицентровое исследование, 11 контролируемых когортных исследований, 6 неконтролируемых когортных исследований, 1 исследование случай – контроль.

Заключение. У детей со сколиозом вследствие церебрального паралича вес и индекс массы тела зависят от функциональной активности. Наиболее подходящими референсными показателями при оценке веса и индекса массы тела детей с церебральным параличом являются стратифицированные GMFCS графики роста детей с церебральным параличом. Недостаточный вес и сниженный индекс массы тела (меньше 10-го процентиля) — факторы, повышающие риск осложнений после хирургической коррекции сколиоза. В перспективе для более оптимальной оценки антропометрических показателей детей с церебральным параличом необходима разработка национальных специальных центильных таблиц.

Ключевые слова: детский церебральный паралич; деформация позвоночника; масса тела; индекс массы тела.

Cerebral palsy (CP) is one of the most common diseases that limits the life activity and significantly deteriorates the quality of life of the affected children [1–5]. The incidence of this disease is 2–9 cases per 1,000 newborns [3, 4].

Children with CP are at an increased risk of scoliosis [6]. The incidence of scoliosis in this group of pediatric patients varies significantly (from 6% to 100%) [7], averaging 20%–25% [6, 8, 9]. The formation of spinal deformity is directly related to the level of motor functions with reference to the GMFCS classification [6, 8], with the incidence rate of 6%–25% with GMFCS I and almost 100% in GMFCS V [10]. The highest incidence of scoliosis (up to 70%) was also recorded in patients with spastic CP. The incidence of scoliosis is 16%–39% in the dyskinetic form and 6%–50% in the atonic-astatic and mixed forms [11–13].

In the absence of treatment, neuromuscular scoliosis reduces the quality of life [10]. The main method of treatment in this case is surgical correction of the spinal deformity. In the surgical treatment of CP-induced scoliosis, the incidence of postoperative complications is high. In pediatric patients with CP, who underwent spinal fusion, serious perioperative complications were registered in 39.4% of the cases [14].

In addition, patients with CP after posterior instrumental correction of scoliosis are at a greater risk of infection at the surgical site (1.1%–15.2%) in comparison to adolescents with idiopathic scoliosis (0.9%) [15, 16].

Several authors believe that one of the risk factors for complications after surgical correction of scoliosis due to CP is body weight, its changes, and body mass index (BMI) [14, 17–20]. However, a unified approach to the analysis of this indicator in patients with CP remains to be established.

This work aimed to analyze the data in the modern literature on the body weight and BMI in pediatric patients with spinal deformity due to CP.

Materials and methods

A systematic literature search was performed in the open electronic databases PubMed, Web of Science, Scopus, MEDLINE, eLIBRARY, and RSCI, according to the bibliography of key articles for the publication period January 1990 to December 2019.

Inclusion criteria were systematic reviews, meta-analyses, multicenter studies, controlled cohort studies, and uncontrolled cohort studies of pediatric patients with spinal deformities due to CP and age of patients with CP of <20 years.
Search keywords: cerebral palsy, spinal deformities due to cerebral palsy, body weight of children with cerebral palsy, and body mass index of children with scoliosis due to cerebral palsy.

Exclusion criteria were clinical cases, monitoring studies, conference proceedings, patients’ age >20 years, and neuromuscular scoliosis of other etiology.

Results

This review initially included 156 articles, of which, 25 met the inclusion criteria. These 25 articles included 3 systematic reviews and a meta-analysis, 3 population studies, 1 multicenter study, 11 controlled cohort studies, 6 uncontrolled cohort studies, and 1 case-control study. Table 1 presents the information about the main publications that meet the inclusion criteria. The geography of publications was sufficiently wide. The sample sizes of patients with CP included in each study ranged from 47 to 25,545 people. The age of the patients ranged from 0–2–8 to 18–20 years in 15 publications, 2–12 years in 1 article, 2–16 years in 1 article, 1–10 years in 1 article, and 3–5 years in 1 article. In 2 articles, the exact age of the pediatric patients with CP was not indicated.
Discussion

BMI in children and adolescents (aged 2–18 years) was calculated as in the adults. The ratio of weight and height was calculated using the following formula: BMI (kg/m²) = body weight (kg)/height² (m²). However, this formula was only suitable for people aged >18 years, since children are constantly growing and their anthropometric data change. To obtain more accurate outcomes when determining the BMI of children, it is necessary to know the child’s gender and age. The results of calculating BMI were compared with the indicators of centile tables, which mostly objectively reflected the range of the percent distribution of anthropometric indicators among healthy children [21]. When estimating BMI using percentile tables, it was determined whether a child or adolescent showed deviations from the parameters of development of an average children [21–23]. BMI indicators of healthy children were categorized into 4 groups: 1) ≥5th to <85th percentile in the normal-weight group; 2) <5th percentile in the underweight group; 3) ≥85th to <95th percentile in the overweight group; and 4) ≥95th percentile in the obese group [22, 24].

The analysis of BMI of children with CP revealed that the body weight was normal in 26.3% of the patients, overweight in 5.4%, obese in 11.5%, and underweight in 56.8% [25]. The incidence of low body weight was greater in children with moderate (GMFCS III) and severe (GMFCS IV and V) degrees of CP (52.7% and 53.8%, respectively). In the group of children with minor CP (GMFCS I, II) who could walk, the number of obese children was greater (7.1%). Based on a past study [25], the BMI indicator influenced functional independence (according to the WeeFIM scale) and the quality of life (with reference to the CHQ-PF50 questionnaire) associated with the health of children with CP. This effect was more pronounced in patients with severe CP (GMFCS IV and V) and in those with low BMI values (Table 2).

Similar results were reported by Ryabykh et al. [5] who demonstrated that, in patients with low BMI (<10th percentile of the growth graph of healthy children) and severe movement disorders (GMFCS IV and V), the incidence of pathology of the cardiovascular and genitourinary systems and anemia increases [5].

Cohort studies on BMI of children with CP of GMFCS I–III conducted by Pascoe et al. [26] demonstrated that 7% of the patients, who could walk, were underweight, 73.6% had normal weight, 7.3% were overweight, and 12.1% were obese. This observation corresponded to the distribution of the indicator among healthy children. Notably, the level of motor functions GMFCS I was recorded in the greatest number of children with normal BMI values and in the majority of obese children with GMFCS III [26].

Hurvitz et al. reported that patients with CP who could walk (GMFCS levels I and II) tended to have a higher prevalence of being overweight (22.7%) when compared with children who could not (GMFCS IV and V, 9.6%). The underweight condition was most common among children who could not walk [27].

According to Feeley et al. [28], in a population of patients with CP who could walk and in children with quadriplegic CP, BMI was significantly lower when compared with that in children with diplegic and hemiplegic CP. However, the fact that BMI was significantly lower in patients with GMFCS III than in those with GMFCS I and II contradicts the results of Pascoe et al., who reported that the level of motor functions in the majority of obese children was GMFCS III [26].

Such inconsistencies in these observations may be attributed to the fact that the data for weight and BMI were obtained from children from different countries and from different continents (Australia, USA) who possessed certain ethnosocial characteristics.

When analyzing a sample of patients with CP in Western China, Wang et al. concluded that growth abnormalities and decreased BMI were significantly more typical for patients with severe movement disorders. In addition, reduced weight was mainly noted among older children (aged 12-18 years), while being overweight and obesity was registered in the group of children aged 2-12 years [29]. However, according to the study by Almuneef et al. [30], one factor that was associated independently with low BMI was the age of ≤5 years. Nevertheless, most authors agree that patients with CP with disorder levels of GMFCS IV and V are more often recorded with lower height, weight, and BMI compared to children with GMFCS I–III [25, 29, 31–38].
Thus, we can conclude that weight and BMI are associated with the functional activity of patients with CP (Table 2). That is, patients with lower functional status have a significantly lower BMI [28, 37]. The underweight condition is most common among children who cannot walk [27, 36, 39]. This observation raises the question of what parameters of the body component composition decrease the weight and BMI in CP children with the level of motor functions GMFCS IV and V. The study of the body composition of children with CP at various levels of motor functions has revealed that patients with GMFCS IV and V have significantly lower indicators of lean mass, skeletal muscle mass, cell body mass, and bone mineral content when compared with children with GMFCS I–III and control children [31–38, 40–43]. In addition, according to Rakhmaeva [43], as motor disorders increases in grade (GMFCS I → GMFCS II → GMFCS IV), the number of patients with a deficiency of fat mass increases.

Anthropometric studies in children with CP have revealed that the measurement of height can be hindered by contractures, scoliosis, and muscle spasticity. If height cannot be measured directly (for example, in children with GMFCS IV and V), other

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<td>Day S.M. et al. [57]</td>
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<td>Hurvitz E.A. et al. [27]</td>
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<td>Egenolf P. et al. [56]</td>
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<td>Baranek E.S. et al. [62]</td>
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calculation methods are used [43–45]. In all cases, when the height in the standing position cannot be determined, the method of segmental measurements is employed, which considers the sum of the lengths of different body segments [45].

Accordingly, height prediction equations that include segmental limb measurements have been developed to determine the precise height in people with motor impairment. For instance, Chumlea et al. [46] developed an equation for predicting height using the knee height (Table 3) for people with limited physical capacities or disabilities. However, these measurements and calculations were performed during examination of a group of healthy children (aged 6–18 years), and it can be presumed that the use of these equations on analyzing anthropometric data of children with limited physical capacities can increase the errors in predicting the height.

Stevenson [47], in an effort to consider the specific growth abnormalities in CP, developed regression equations for the lengths of limb segments in a population of patients with CP (Table 3). However, in this work, the level of GMFCS was not considered, and the patients with scoliosis or contractures were excluded from analysis. The patients whose data were used to formulate the predictive equations were younger and presented with less severe neurological impairments when compared with the general population of patients with CP.

Another commonly used equation was proposed by Gauld et al. [48] on a large group of healthy Australian schoolchildren (Table 3); this equation was tested on a small sample of children and adolescents with Duchenne muscular dystrophy.

Haapala et al. [44] analyzed the correspondences between the calculated height (considering the lengths of the limb segments) using the above equations and the measured height (actual height) in a heterogeneous, clinically representative sample of patients with CP. The actual height was determined either in the standing position in patients who could stand or in the supine position in patients who could not. According to these authors [44], the average difference between the measured and calculated heights was smallest when the knee height calculations and equations by Stevenson and

<table>
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<th>Height prediction equations with segment lengths [44]</th>
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<tr>
<td><strong>Using the formula of Chumlea [46] (healthy children, age 6–18 years)</strong></td>
</tr>
<tr>
<td>Caucasian boy</td>
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<tr>
<td>( H = (2.22 \cdot \text{KH}) + 40.54 )</td>
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<tr>
<td>Black boy</td>
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<td>( H = (2.18 \cdot \text{KH}) + 39.60 )</td>
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<tr>
<td>Caucasian girl</td>
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<tr>
<td>( H = (2.15 \cdot \text{KH}) + 43.21 )</td>
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<tr>
<td>Black girl</td>
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<td>( H = (2.02 \cdot \text{KH}) + 46.59 )</td>
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<tr>
<td><strong>Using the formula of Stevenson [47] (children with cerebral palsy, age 0–12 years)</strong></td>
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<tr>
<td>Knee height</td>
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<tr>
<td>( H = (2.68 \cdot \text{KH}) + 24.2 )</td>
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<tr>
<td>Tibia length</td>
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<tr>
<td>( H = (3.26 \cdot \text{TL}) + 30.8 )</td>
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<tr>
<td><strong>Using the formula of Gauld [48] (healthy children, DMD children, age 7–18 years)</strong></td>
</tr>
<tr>
<td>Boys</td>
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<tr>
<td>Ulnar bone length</td>
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<tr>
<td>( H = 4.605U + 1.308A + 28.003 )</td>
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<tr>
<td>Knee height</td>
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<tr>
<td>( H = 2.423KH + 1.327A + 21.818 )</td>
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<tr>
<td>Tibia length</td>
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<td>( H = 2.758T + 1.717A + 36.509 )</td>
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<tr>
<td>Girls</td>
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<tr>
<td>Ulnar bone length</td>
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<tr>
<td>( H = 4.459U + 1.315A + 31.485 )</td>
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<tr>
<td>Knee height</td>
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<tr>
<td>( H = 2.473KH + 1.187A + 21.151 )</td>
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<tr>
<td>Tibia length</td>
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<tr>
<td>( H = 2.771T + 1.457A + 37.748 )</td>
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Note. \( H \) — estimated height; \( \text{KH} \) — knee height; \( \text{TL} \) — tibia length; \( U \) — ulnar bone length; \( A \) — age; \( \text{DMD} \) — Duchenne muscular dystrophy.
Gauld et al. (5.4 ± 1.8 and 6.4 ± 0.2 cm, respectively) were used. In cases where the calculations considered the length of the ulna as per the Gauld equation, there was a tendency of increased calculated height in comparison with the measured height (averagely by 6.9 cm). Similar data were reported during anthropometric studies by Amezquita and Hodgson [49] and Iñiguez et al. [45]. Notably, in CP with the level of GMFCS I–III, the difference between the calculated value of height (according to the equations of Gauld et al. and Stevenson) and the actual one is (on an average) close to zero. However, in patients with the levels of abnormalities of GMFCS IV and V, the calculated height was less than the measured one [44]. For patients with GMFCS levels IV and V, when calculated using the Gauld et al. equation, the calculated height differed less from the actual height than when using the Stevenson equation. The height parameters obtained using the equation of Gauld et al. considered that the length of the ulna was greater than that with direct measurements, both in patients with GMFCS I–III and in those with GMFCS IV and V [44].

García-Contreras et al. [50] confirmed the usefulness of the Stevenson equation to assess the height of patients with CP with spastic quadriplegia (GMFCS V) as well as that of healthy children. The correlation coefficient between the measured (in healthy patients in the standing position and in patients with CP in the supine position) and the calculated heights was greater in patients with CP than in healthy children. These authors thus also believed that the knee height is the most appropriate parameter for calculating the height of patients with spastic CP and healthy children [50].

The use of estimated height to determine BMI may not always be reliable, since height is squared and therefore any error is exponential [44, 51]. However, despite these shortcomings [44, 51], equations for predicting height using the knee height and lower leg length are useful for calculating the height and BMI of patients with CP; these equations continue to be used by Russian and international researchers [43, 45, 49, 50].

In addition, an equally important problem in assessing the anthropometric parameters of patients with CP is determining the comparison group, the control group, and the reference values.

Medical professionals use growth graphs (centile tables) as a tool to monitor the growth of a child with CP in comparison with that of age- and gender-matched healthy children. The need for examination or treatment in children with weight or height significantly lower than the normal percentile was identified. When analyzing the BMI of patients with CP, several authors are of the opinion that the normal limits of this indicator for patients suffering from severe forms of CP differ from those of healthy children [52], while the BMI indicators <10th percentile of values for a healthy population are considered pathological [39, 53].

However, according to other authors, the reference standard of the World Health Organization (WHO) is not suitable for the assessment of most anthropometric parameters in patients with CP. Therefore, the height of these children should not be compared to that of healthy children. Thus, as a rule, the growth graphs for healthy children overestimate the underweight condition (with reference to the values of tables of centiles of healthy children, the number of patients with CP with underweight condition are higher) [45, 54–56].

The researchers dealing with the problems of CP have gradually come to the conclusion that the growth graphs (special centile tables) developed by Day [57] and Brooks [58] are more effective as reference indicators (Table 2). These authors conducted a major study on patients with CP who had applied to the California Development Department (USA) over a 15–year period (1988–2002). They analyzed the height of 24,920 patients with CP and revealed clear gradients in height and weight between the most and least severe types of motor impairment with reference to the GMFCS [57]. The same group of authors, on the basis of the data obtained, constructed special centile tables of height, weight, and BMI for patients with CP aged 2-20 years [58]. The graphs presenting the percentiles for height–age, weight–age, and BMI–age [59] were modeled using 141,961 measures of height and weight. Special graphs were developed separately for boys and girls for each of the five levels of the motor function classification system (GMFCS I–V). Level V (most severe motor impairment) was additionally stratified by the presence or absence of a feeding tube [58]. On the graphs, the weight indicators of children were determined, at which the risk of mortality was significantly increased. For levels I and II of the GMFCS scale, it was equal to the ≤5th per-
centile, while, for GMFCS III–V, it was <20th per-
centile [58, 59].

Wright et al. [54] decided to test the degree of
the data of British children with CP corresponding
to the growth graphs of Day and Brooks [57, 58]
in comparison with the growth graphs of healthy
children (Great Britain, WHO) [60]. The height of
children with severe CP was significantly reduced
(2nd centile) in comparison with this indicator as
per the tables of centiles of healthy children. When
assessing the anthropometric data with reference to
the growth graphs of patients with CP in the United
States [58, 59], height and weight were found to be
between the 50th and 75th centiles for all GMFCS
levels, while the BMI was approximately at the
50th centile. The data on British children have been
found to be more consistent with the special centile
tables of patients with CP in the United States than
with those of healthy children [54]. With regard to
the height and weight of British pediatric patients
with CP, an increasing trend in these rates was
noted in comparison to those of patients with CP
in the United States [54]. The authors attribute this
observation to the peculiarities of the mixed ethnic
composition of patients with CP in the USA.

Viñals-Labañino et al. [55] also confirmed the
need to employ growth graphs of patients with CP,
developed by Day and Brooks [58, 59] as a guideline
to assess the anthropometric characteristics of
patients with CP (Table 2). They compared the rates
of patients with CP in Mexico and the USA. The
parameters of height and BMI were comparable,
although the weight indices were significantly
different [55].

Egenolf et al. [56] developed a system of height
percentiles for Caucasian children with CP that
depended on the degree of impairment of the motor
function [56]. Based on the results of a study by
these authors, the height of patients with CP was
significantly lower than that of healthy Caucasian
children, and this difference increased with an
increase in the deterioration of physical activities.
In addition, according to Day and Brooks in the
USA, the height of Caucasian patients with CP was
greater than that of patients with CP [57–59]. This
observation was largely concerned with children of
GMFCS levels IV and V of disorders [56]. Differences
in the height between Caucasian patients with CP
and patients with CP from the United States may
be due to the difference in their ethnicity. Thus, the
height rates in the tables of the centiles of healthy
children were higher among the representatives of
the Caucasian race [56] than in the studies of Day
and Brooks (USA), where the ethnic composition of
the population was miscellaneous [57–59]. This
fact necessitates the development of national special
centile tables for patients with CP.

In a pediatric population of children with
CP, BMI and underweight condition increase the
chances of potential complications during osteotomy
and spinal surgery, while being overweight and
obese did not have such an effect [5, 18] (Table 2).

The researchers also considered the tendency
toward decreasing BMI as one of the factors that
increase the risk of complications in patients with CP
who underwent correction of spinal deformity [14].
A lower BMI correlated with a higher risk of
infection at the surgical site owing to poor wound
healing and reduced immunity in this group of
patients [17, 61].

According to Baranek et al. [62], special centile
tables for patients with CP were employed as
a useful tool for identifying patients at increased risk
of infection in the surgical wound site after surgical
correction of spinal deformity. Simultaneously, the
standard CDC growth graphs (Centers for Disease
Control and Prevention) are much less sensitive.
Based on these reports, a significant correlation
existed between BMI <10th percentile on the
stratified GMFCS growth graphs of patients with
CP before surgery and the development of infection
of the surgical site after surgical correction of spinal
deformity. All cases of infection were registered
in patients with levels IV and V according to the
GMFCS [62].

**Conclusion**

In children with scoliosis due to CP, the weight
and BMI depend on the functional activity. Patients
with a lower functional status have a significantly
lower BMI. The condition of being underweight is
most common among children who are unable to
walk. Patients with GMFCS levels IV and V showed
significantly lower lean mass, skeletal muscle
mass, cell body mass, and bone mineral content
when compared with children with GMFCS I–III
and the control group. With the increase in the
motor impairment, the number of patients with fat
deficiency also increases.
Several authors believe that special centile tables for children with CP are more suitable for assessing the anthropometric data of patients with CP as reference indicators when compared with the WHO and CDC standards. Moreover, it is advisable to develop national special centile tables for children with CP. Centile boundaries were identified, at which the risk of mortality increases significantly, to be equal to ≤5th percentile for GMFCS scale levels I and II and <20th percentile for GMFCS III–V.

Low body weight and low BMI are associated with a high risk of complications after surgical correction of scoliosis. A significant correlation was noted between BMI <10th percentile on stratified correction of scoliosis. A significant correlation was with a high risk of complications after surgical and approved the final version before its publica-

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Author contributions

E.N. Shchurova created the concept and design of the study, performed analysis of literary sources, and wrote the text of the article.

S.O. Ryabykh performed analysis of literary sources, generalized the information, and wrote the text of the article.

E.Yu. Filatov analyzed the literature data and wrote a fragment of the article text.

P.V. Ochirova collected and processed the literary sources.

T.V. Ryabykh collected and processed the literary sources.

All authors made significant contributions to the research and preparation of the article and read and approved the final version before its publication.

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