



Assessment of the state of patients with spastic cerebral palsy at transition to adult medical institutions: a cross-sectional study

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Abstract

Introduction The purpose of this cross-sectional study was anamnestic evaluation of the treatment performed in patients with GMFCS levels I, II, III spastic cerebral palsy and to assess their motor status, quality of life and anatomical disorders in the lower extremities in the age range from 17 to 18.5 years. **Material and methods** A cross-sectional study was conducted in patients with spastic types of cerebral palsy at the age of 17-18.5 years. The completeness and consistency of previous treatment (according to medical documentation), motor abilities, quality of life, radiographic anatomy of the lower extremities were studied. **Results** We selected 73 patients meeting the inclusion criteria out of 201 cases. Insufficient systematic implementation of complex therapeutic measures was revealed, which was reflected in a high rate of orthopedic interventions (93.1 %). Fibromyotomies, triceps lengthening surgeries performed at an early age (6-7 years) contribute to the development of an iatrogenic crouch gait pattern, a decrease in motor abilities and quality of life by the age of 17-18 years. The probability of maintaining a positive result and improving motor ability by the end of the childhood period is significantly higher in the subgroup of multilevel orthopedic interventions than in patients after fibromyotomies or who did not undergo orthopedic treatment; the differences are significant as the chi-square test for binary samples shows ($p = 0.012$). **Conclusions** The severity of impairment of motor abilities, quality of life, incidence of the crouch gait pattern in patients with spastic types of cerebral palsy are close between subgroups at the time of transition to adult medical and diagnostic institutions. The quality of life and motor status after multi-level intervention improves and remains stable by the end of the childhood. Early surgical interventions for lengthening the triceps, fibromyotomy reduce motor potential in the long term, cause the development of the iatrogenic crouch gait pattern, and decrease the quality of life of adolescents with mild neurological disorders.

Keywords: spastic cerebral palsy, adolescents, young adults, orthopedic complications, crouch gait, healthcare organization

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INTRODUCTION

Cerebral palsy remains the main cause of movement disorders that occur in early childhood and accompany the patient throughout the life. The average rate is 1/500 newborns [1]. An increase in the life expectancy of persons with cerebral palsy has been achieved due to improved medical care [2, 3]. At the same time, the majority (up to 99 %) of patients with the level of impairment of global motor functions of the Gross Motor Function Classification System (GMFCS) I, II, III reach 18 years of age and are referred to the adult network of medical institutions for further treatment [4, 5].

The basis for the treatment of children with cerebral palsy is the combination of spasticity control with physical rehabilitation, conservative orthopedic treatment

and surgical orthopedic operations (if orthopedic complications occur) [1, 6-8]. The combination of therapies involves the development of motor skills and tolerance, which must be maintained for a long time, throughout life [6, 7]. Therefore, it is so important to assess the condition of patients aged 17-18 years at their transfer for observation and treatment to adult medical institutions, given the systemic nature of the disease [4, 5, 9, 10].

Purpose Anamnestic assessment of the treatment performed in patients with spastic GMFCS levels I, II, III, as well as assessment of their motor status, quality of life and anatomical disorders of the lower extremities in the age range of 17-18.5 years.

MATERIALS AND METHODS

The selection of patients for the study was determined by the availability of complete medical records at the Republican Children's Clinical Hospital of the Republic of Bashkortostan, who were under regular supervision of an orthopedist, neurologist

and physical therapist. The inclusion criteria were the age of 17-18 years, the diagnosis of cerebral palsy (G80.1, G80.2) in patients without critical disorders of global motor functions (GMFCS I, II, III). After the selection based on medical records, patients

were called for a clinical examination, a sociological study, and radiography. The study included only patients whose medical documentation was complete (stages and details of treatment reflected), and clinical, sociological and radiographic studies were carried out.

The study was conducted in the period January-April 2022.

The following parameters were studied: the severity of spastic types of cerebral palsy according to the GMFCS scale [11], the regularity of antispastic therapy, rehabilitation measures and orthotics throughout childhood, orthopedic surgical interventions performed (volume and type), incidence of the crouch gait pattern, quality of life (PedsQL) [12].

Parents or relatives caring for patients evaluated the treatment and its result according to the criteria: improvement or preservation of a positive result by this age, or worsening and loss of the result.

Radiographic parameters of the position of the patella [13] and relationships in the ankle joint and midfoot were studied: tibio-talar angle, coverage of the head of the talus with the navicular bone, Shade line [14].

The study did not include patients with GMFCS levels IV-V, non-spastic types of cerebral palsy, aged less than 16 years or older than 18.5 years, or cases of incomplete medical records.

Due to the diversity of the severity of the pathology and some elements of surgical orthopedic treatment

that are crucial for motor prognosis in the long term, we tried to compare groups of patients, dividing them into hemiplegic and diplegic types and within each group a subgroup of patients without orthopedic surgery (1), patients who performed surgical interventions that critically weaken the triceps of the leg (fibromyotomy) (2), patients with multi-level classical operations (3).

The AtteStat 12.0.5 program was used for statistical data processing. In descriptive statistics, the mean value of the indicator and its standard deviation were used. To compare the occurrence of a deterioration sign in motor abilities by the age of 17-18 years and pathologically low values of the tibio-talar angle, a chi-square test was used for binary samples. Differences were considered significant at p less than 0.05.

The approval of the Ethics Committee of the Federal State Budgetary Institution Ilizarov Center No. 2(57) dated May 17, 2018 was obtained. The studies were carried out in accordance with the ethical standards of the Declaration of Helsinki of the World Medical Association "Ethical principles for conducting scientific medical research involving human subjects" as amended in 2000, "Rules of Clinical Practice in the Russian Federation", approved by Order Ministry of Health of the Russian Federation dated June 19, 2003 No. 266. Patients or parents of patients, authorized employees of social institutions confirmed their consent to conduct the study and publish the results without identifying the person.

RESULTS

From the list of 201 potential patients, 73 patients were included in the study group.

The average age at the time of examination and assessment of motor and X-ray parameters was 17.8 ± 0.7 years. The gender distribution was as follows: 25/48 (female/male). According to the International Classification of Diseases (10th revision), the distribution was as follows: G 80.1 – 54 patients, G80.2 – 19 patients.

Throughout the childhood, botulinum therapy was carried out: regularly (at least 4 courses) in 31 children (botulinum therapy was initiated before the age of 10 years in 30 cases), sporadically or unsystematically in 3 children, and was not carried out in 39 persons.

Orthotic appliances were used regularly in combination with botulinum therapy only in two cases (Ankle Foot Orthosis (AFO) was used in both cases), rarely or unsystematically in four patients; in other cases, orthotic means were not used.

Physical therapy (postural management, strengthening of antagonist muscles, development of selective control of movements) on a regular

basis (at least 2 sessions per week) was performed on an outpatient basis in 35 cases; it was conducted rarely in 16 cases (not more than once per week) and not carried out at all in 22 children. Therapeutic measures with hospitalization in rehabilitation centers were carried out in 15 cases (at least 1-2 courses per year), rarely in 11 cases, and not carried out in 46 patients. Ten subjects underwent sanatorium-resort treatment on a regular basis annually, 10 persons sporadically, and the rest never had such treatment.

Sixty-one patients underwent surgical orthopaedic treatment. Among the surgical interventions performed, there were 22 cases of various options for lengthening the Achilles tendon (according to Bayer or open Z-shaped); that this approach was used both in patients with hemiparesis (5 cases) and diplegia (17 cases). Multilevel surgical interventions were performed at the Ilizarov Center or by the surgeons of the Republican Children's Clinical Hospital of the Ministry of Health of the Republic of Bashkortostan.

More detailed information on the subgroups is presented in Tables 1-4.

Table 1

Hemiplegic types: treatment variants

Criterion	Subgroup		
	No surgery	Fibromyotomy	Multilevel orthopaedic interventions
GMFCS: I/II/III	3/2/0	3/2/0	3/3/3
Botulinum therapy: systemic/ sporadic / not carried out	3/1/1	1/0/4	4/0/5
Rehabilitation: systemic/ sporadic / not carried out	3/2/0	2/1/2	5/1/3
Age at operation (years)	–	7.2 ± 4.3	9.5 ± 3.4

Table 2

Diplegic types: treatment variants

Criterion	Subgroup		
	No surgery	Fibromyotomy	Multilevel orthopaedic interventions
GMFCS: I/II/III	4/2/1	1/8/8	0/13/17
Botulinum therapy: systemic/ sporadic / not carried out	5/1/1	4/0/13	14/2/14
Rehabilitation: systemic/ sporadic / not carried out	4/3/0	7/4/6	17/2/11
Age at operation (years)	–	6.3 ± 3.3	11.5 ± 3.46

Table 3

Hemiplegic types: total treatment outcome

Criterion	Subgroup		
	No surgery	Fibromyotomy	Multilevel orthopaedic interventions
Assessment of the patient's condition before the transition to the adult medical institutions (improvement or preservation/deterioration); number of cases	4/3	5/11	24/6*
Total result PedsQL; points	49.9 ± 16.5	54.1 ± 17.4	52.8 ± 18.1

Table 4

Diplegic types: total treatment outcome

Criterion	Subgroup		
	No surgery	Fibromyotomy	Multilevel orthopaedic interventions
Assessment of the patient's condition before the transition to the adult medical institutions (improvement or preservation/deterioration); number of cases	4/3	5/11	24/6*
Total result PedsQL; points	49.9 ± 16.5	54.1 ± 17.4	52.8 ± 18.1
Crouch gait rate at the study time, %	33.3	29.4	36.7

Note: * – significant difference between the groups of fibromyotomies and multilevel interventions by chi-square test for binary samples ($p = 0.012$)

A trend towards a decrease in the frequency of surgical interventions and the predominant use of conservative management in GMFCS I patients can be noted. The general trend for this sample is a rare systemic treatment that combines rehabilitation measures, botulinum toxin therapy and orthotics (50 % or less). The older age of performing multilevel interventions than fibromyotomies was observed.

The quality of life achieved was similar among all groups, despite the fact that more severe motor dysfunction according to GMFCS was noted in the group of multilevel interventions. Interestingly, the higher quality of life was reported by patients of the group of hemiplegic complications who underwent

surgery, regardless of the type of surgery performed, than in patients without intervention

In the spastic diplegia group, there is a clear incidence of positive outcomes after multilevel interventions as assessed by parents and a clear indication of deterioration in outcomes after fibromyotomies by the time of transition to adult institutions (by Chi-square test).

There are several aspects to consider by interpreting radiographic data:

– the occurrence of radiographic anatomic foot disorders and pathologically high values of the Caton-Deschamps index, characteristic of the crouch gait pattern, is close to the occurrence of this gait disorder for diplegic types;

– in the hemiplegic type, there are no pronounced radiographic anatomic disorders and there is no development of the crouch gait pattern in this subgroup, which is explained by the peculiarities of the biomechanics of walking and vertical posture in this type of neurological disorders, when a healthy

limb prevents decompensation of adaptive position on the affected side;

– pathologically low values of the tibio-talar angle are found only in the subgroup of fibromyotomies, which is characterized by weakness of the triceps of the leg after this type of surgery.

Table 5

Hemiplegic types. Radiographic findings

Criterion	Subgroups		
	No surgery	fibromyotomy	SEMLS
Tibio-talar angle, less than 90°/ 90°-105° / more than 105°; number of cases	0/4/3	0/1/4	0/2/7
Coverage of the talar head by the navicular bone; %	83.2 ± 14.4	84.1 ± 7.9	86.4 ± 9.3
Incidence of the talar head coverage with the navicular bones less than 80 %; number of cases	1/7	0/5	1/9
Shade line; 0°-9° / 10° and more; number of cases	6/1	5/0	9/0

Table 6

Diplegic types. Radiographic findings

Criterion	Subgroups		
	No surgery	Fibromyotomy	SEMLS
Tibio-talar angle, less than 90°/ 90°-105° / more than 105°; number of cases	0/10/14	12/6/16*	0/12/48
Coverage of the talar head by the navicular bone; %	73.4 ± 12.6	86.7 ± 16.2	68.1 ± 19.1
Incidence of the talar head coverage with the navicular bones less than 80 %; number of cases	18/6	4/30	22/38
Shade line; 0°-9° / 10° and more; number of cases	15/9	23/7	27/33
Caton-Deschamps index; less than 1,2/1,2 and more; number of cases	7/17	8/26	18/42

Note: * – significant difference between the groups of fibromyotomies and multilevel interventions by chi-square test for binary samples ($p = 0.041$).

DISCUSSION

Our study revealed the lack of consistency in the complex treatment and prevention of orthopaedic complications in this cohort of cerebral palsy patients. The reasons for the difficulties in organizing and implementing an integrated approach to the treatment of children with spastic types of cerebral palsy in the countries with low and middle incomes of the population are, in addition to the low availability of medical care, the insufficient knowledge of doctors about high-evidence treatment methods, late diagnosis of the disease and insufficient awareness of the parents about the nature of disease and the effectiveness of various treatments [15, 16]. Al Jabri et al. [17] showed that the first step towards overcoming the shortcomings of the system of care for children with cerebral palsy is the creation of national registries of patients that consider the severity of the disease, its possible causes, and allow planning medical care in different regions of the country.

Indirect evidence of the insufficiency and non-systematic use of botulinum therapy, physical rehabilitation and conservative orthopedic treatment is

a high percentage of patients (93.1 %) in this sample who underwent surgical treatment for orthopedic complications of cerebral palsy. It is known that the timely and regular use of botulinum toxin preparations in combination with physical therapy has reduced the frequency of surgical orthopedic interventions to 47 % in a similar region of the Russian Federation [18].

The study revealed a number of differences between the groups where surgical interventions were performed: fibrotomy or multilevel orthopedic surgery. The first difference is the age for the intervention. Fibromyotomies were performed at an early age, on average, at 6-7 years of age. The average age of multilevel interventions is much higher (older than 11 years). Such an approach (in the subgroup of multilevel operations) reflects the balanced decision-making on surgical intervention as being inevitable, that is only at the time the true orthopedic complications of cerebral palsy has developed. The literature also indicates the optimality of performing orthopedic operations later, at the age of 9-12 years [19, 20]. Svehlik et al. [21] studied the outcomes of multilevel surgeries in a group

of 32 patients in the average age of 10.5 years and a crouch gait pattern and found that the older the child at the time of surgery, the more stable and better was the result in the long-term follow-up that was 10 years.

The success of treatment, including surgery, determines the quality of life of patients [22]. The quality of life of patients with GMFCS levels I-III depends, first of all, on the mobility of patients in the long-term period after performing multilevel interventions [23]. Himpens et al. [24] indicate that after orthopedic interventions, patients rate the improvement in the quality of life higher than their parents.

In our study, the achieved quality of life was similar among the subgroups, despite the fact that more severe motor function impairment according to GMFCS were noted in the group of multilevel interventions. Multilevel interventions enable to maintain or improve the quality of life by the end of childhood (at an average follow-up of 5-6 years). The overall assessment of the result of the treatment by the end of childhood in the subgroups of conservative treatment and fibromyotomy is a significant deterioration in motor abilities for patients with diplegic lesions.

The radiographic anatomy of the feet in patients after fibromyotomies and the development of the crouch gait pattern shows the characteristic features of the pathogenesis of this gait pathology after surgical weakening of the triceps of the lower leg [25, 26]: the development of the iatrogenic crouch gait pattern in the absence of pathology of biomechanical levers associated with foot deformities in the horizontal plane [27]. Obviously, such disorders require the planning and implementation of surgical treatment in adulthood in order to prevent the loss of motor abilities and the development of early arthrosis of the knee, femoropatellar and talo-navicular joints [28].

We understand the shortcomings of the study which lie in the fact that the analysis of the results should be even more detailed. Our study was conducted on the limited amount of retrospective data available to us. Historically, in terms of treatment organization, this cohort was formed at a time when there were no protocols for managing such patients developed on the basis of evidence-based medicine. Changes in the strategy of future studies to a prospective one, the creation of regional registers of patients with cerebral palsy is necessary for conducting a multivariate analysis.

CONCLUSION

The severity of impaired motor abilities, quality of life, incidence of the crouch gait pattern in patients with spastic types of cerebral palsy are close between subgroups at the time of transition to the adult network of medical and diagnostic institutions. This reflects the lack of consistency in their complex treatment.

The quality of life and motor status after multilevel interventions performed in patients with more severe neurological status improves and remains stable by the end of the childhood period of life.

Early surgical interventions for lengthening the triceps and fibromyotomy reduce motor potential in the long term, cause the development of the iatrogenic crouch gait pattern what ultimately reduces the quality of life of adolescents with mild neurological disorders.

The creation of regional patient registries that would allow for the systematic treatment of children with cerebral palsy with proven effective methods remains relevant.

Conflict of interest None.

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Ethical review The approval of the Ethics Committee of the Federal State Budgetary Institution National Ilizarov Center No. 2(57) dated May 17, 2018 was obtained. The studies were carried out in accordance with the ethical standards of the Declaration of Helsinki of the World Medical Association "Ethical principles for conducting scientific medical research involving humans" as amended in 2000, "Rules of Clinical Practice in the Russian Federation", approved by Order Ministry of Health of the Russian Federation dated June 19, 2003 No. 266.

Informed consent Patients or parents of patients, authorized employees of social institutions confirmed their consent to conduct the study and publish the results without identifying the person.

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