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Surgical orthopaedic management of cerebral palsy in adults: literature review and preliminary analysis of our treatment experience

O.I. Gatamov[1], G.M. Chibirov[1], D.Yu. Borzunov[1,2], D.A. Popkov[1]

[1]Russian Ilizarov Scientific Center for Restorative Traumatology and Orthopaedics, Kurgan, Russian Federation
[2]Tyumen State Medical University, Tyumen, Russian Federation

IIntroduction Improving the quality of care has led to an increase in the life expectancy of patients with cerebral palsy and in the number of adult patients suffering from cerebral palsy. However, functional motor limitations aggravate after their physiological growth completion and the risk of pain increases. The **aim of this work** was to study the literature on the problem of surgical orthopedic treatment in adolescents and adults with cerebral palsy belonging to GMFCS levels I-IV of motor disorders as well as to make a preliminary analysis of the surgical orthopedic treatment performed in this category of patients at our institution. **Materials and methods** The results of multi-level single-event interventions were studied in 165 patients older than 16 years. The sample for analysis included cases that met the following criteria: age of 16 years and older, spastic types of cerebral palsy, GMFCS levels I–IV. In addition, some patients underwent botulinum therapy during the stages of surgical treatment. **Results** The maximum functional effect was manifested 12–24 months after the surgery if proper early and subsequent rehabilitation was provided. According to the Gillette Functional Assessment Questionaire, motor abilities improved in 81.3 %. Multilevel interventions included 2.3–3.5 elements on average during one surgical session. Current literature postulates the implementation of multi-level single-event interventions and indications for surgery and follow-up control are studied at a motion analysis laboratory. **Conclusion** Multi-level orthopedic interventions are indicated for patients who have completed physiological growth. Techniques of such interventions should provide early functional activity. Surgical orthopedic treatment in adult patients with cerebral palsy should be performed by the staff and at an institution that specialize in neuro-orthopedics.

Keywords: cerebral palsy, adults, multi-level orthopedic interventions, botulinum therapy

Cerebral palsy (CP) is a static non-progressive encephalopathy, accompanied by impaired control of motion and posture [1-4]. Secondary CP manifestations that have a progressive nature and are associated with aggravation of motor disorders and quality of life are orthopedic complications: contractures, deformities, dislocations, early degenerative changes in articulating surfaces, accompanied by severe pain [5-14]. These orthopedic complications of cerebral palsy can be prevented, partially or fully compensated with adequate and timely therapeutic measures, including botulinum therapy, kinesitherapy, rational orthotics among other methods [15-21].

Improvement in the quality of care has led to an increase in the life expectancy of patients with cerebral palsy (CP) and in the number of adult patients suffering from CP with multi-component orthopaedic pathology [5, 6, 13, 22–25]. It was shown that upon completion of physiologiac growth (after 16 years of age) functional motor limitations aggravate and the risk of pain increases [6, 26, 27].

The aim of this work was to study the available literature on surgical orthopedic treatment in adolescents and adults with cerebral palsy belonging to GMFCS levels I–IV of motion disorders as well as, on the basis of the literature findings, to make a preliminary analysis of surgical orthopedic treatment performed in CP patients at our institution's clinic of neuro-orthopadics.

MATERIALAND METHODS

According to the literature analyzed, 225 Russian literature sources were devoted to cerebral palsy, among which 198 (88.1 %) reported on rehabilitation of children and only 27 (11.9 %) to treatment of adult patients. Among 893 foreign sources, the ratio of children-to-adult sources was 840/53, which is 96 % and 4 %, respectively [28].

We retrospectively analyzed the medical records of all patients with cerebral palsy who were treated at our institution in specialized neuro-orthopedic units from 2014 to June 2017.

The sample for analysis included cases that met the following criteria:

– age of 16 years and older;

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– spastic CP types;

– GMFCS levels I–IV [29];

– patients with multi-level single-event reconstructive interventions on the lower limbs.

We studied the types of surgical interventions, stages of reconstructive operation, average number of surgical elements per operation. Patients' ability to move in the pre- and postoperative period which follow-up was 1.5–2 years after the intervention was assessed using the Gillette scale [30, 31].

Literature on this topic was searched in the databases of PubMed, Scopus, and RINC. Mean values and standard deviation were calculated using Microsoft Excel 2016 to present quantitative data.

RESULTS

During the study period, 1267 patients were surgically treated at our institution. All of them underwent orthopedic operations on the extremities. The group of patients aged 18 years and older included 252 individuals (19.8 %).

Patients in the age of 16 years and older, related to the levels of motor activity I–IV, who underwent multi-level single-event interventions on the lower limbs and who were included in this study, were 165 individuals.

In total, 228 types of reconstructive interventions were performed (1.38 types per case). The average age of patients at the start of treatment was 21.5 ± 6.4 years.

Table 1 shows the distribution of patients into groups depending on the level of their motor activity, their mean age, number of single-event stages of reconstructive treatment performed at the Ilizarov Center, as well as previous interventions. The latter parameter will be interesting in the study of elements of surgical treatment, performed at our institution.

Half of the patients with severe cerebral palsy (GMFCS levels III and IV) had at least two open surgical reconstructive interventions. Operations on the extremities were sequentially performed due to a large volume of interventions, as well as the need to preserve the possibility of active verticalization of patients and their mobility, taking into account the age and particularities of patient's care.

Table 2 presents the distribution of elements of the surgical intervention based on the severity of general motor functions disorders.

As can be seen from the data presented in Table 2, the largest number of surgical elements per intervention was used in the management of patients with GMFCS levels II to IV. The elements of the operation performed reflected multi-component orthopedic problems that required surgical correction.

In addition, eight patients underwent botulinum therapy on the retracted muscles simultaneously with the surgical intervention by administration of the Dysport preparation into the muscles, the spasticity of which caused restrictions of the motion in the joints of the lower limbs. The target muscles were m. rectus femoris (6), m. gracilis (4), m. adductor longus (4), m. semitendinosus (3), and m. semimembranosus (3).

The maximum functional effect was manifested 12 to 24 months after the intervention, if a proper early and subsequent rehabilitation was provided. According to the Gillette Functional Assessment Questionaire, motion abilities improved in 91 out of 112 cases (81.3 %). Table 3 shows the change in the motor abilities of patients.

Table 1

Patients' distribution according to GMFCS level

GMFCS	Number of patients	Mean age	Number of single-event stages per patient	Previous interventions	Fibromyotomies or staged fibromyotomy, number of cases (%)
I	7	24.1 ± 6.3	7/1	none	1 (14 %)
II	65	21.9 ± 6.0	81/1.25	4 percutaneous lengthenings of the Achilles tendon 2 triple arthrodesis	6 (9.2 %)
III	76	21.2 ± 6.7	114/1.50	6 open lengthenings of the Achilles tendon 1 femur detorsion	23 (26.3 %)
IV	17	19.8 ± 6.3	25/1.47	2 open lengthenings of the Achilles tendon	5 (29.4 %)

Table 2

Elements of surgical interventions

Intervention element and number of its application	GMFCS			
	I	II	III	IV
Aponeurotomy of m. psoas	0	0	2	2
Proximal tenotomy of m. rectus femoris	0	0	1	2
Distal transfer of m. rectus femoris	0	6	2	0
Detorsion osteotomy of the femur	2	21	15	5
Lengthening of femoral adductors	1	16	17	2
Lengthening of the medial knee flexors (hamstring group)	1	55	70	19
Bringing the patella down	0	26	50	13
Supracondylar extension osteotomy of the femur	0	12	33	3
Detorsion osteotomy of the tibia	0	10	6	1
Aponeurotomy of m. gastrocnemius	5	46	39	5
Percutaneous lengthening of the Achilles tendo	2	6	3	5
Lengthening of the calcaneous according to Evans	1	4	2	0
Tripple arthrodesis of the foot	0	25	65	11
Subtalar arthrodesis	0	7	11	5
Talonavicular arthrodesis	0	1	0	1
Arthrodesis of the 1st matatarsophalangeal joint	0	6	19	2
Hemitransfer of the anterior tibial muscle	1	2	2	0
Transfer of peroneal tendons on the calcaneal bone	0	3	6	2
Shortening of the Achilles tendon	0	8	12	0
Reconstructive correction of hallux valgus	1	19	31	4
Shortening of the posterior tibial muscle	1	9	8	3
Deformity correction in toes II–V (plasty of tendons and muscle, interphalangeal arthrodesis)	1	2	2	2
Average per 1 intervention	2.29	3.51	3.47	3.48

Table 3

Motion abilities according to Gillette Functional Assessment Questionnaire

Level	Before treatment	1.5–2 years after treatment
3 – Walks during exercise but not for household distances, needs help for moving	28	12
4 – Walks for household distances, but makes slow progress. Does not use walking at home as preferred mobility	19	9
5 – Walks more than 4.5-15 m for household distances routinely at home and/or school. Indoor walking only	22	15
6 –Walks more than 15-50 m outside the home but usually uses a wheelchair for community distances or in congested areas	26	27
7 –Walks outside for community distances, but only on level surfaces (cannot perform curbs, uneven terrain, or stairs without assistance of another person)	11	34
8 –Walks outside the home for community distances, is able to get around on curbs and uneven terrain in addition to level surfaces, but usually requires minimal assistance or supervision for safety	6	10
9 –Walks outside the home for community distances, easily gets around on level ground, curbs, and uneven terrain but has difficulty or requires minimal assistance or supervision with running, climbing, and/or stairs		5

It should be noted that the global goal of surgical orthopedic treatment in neurological disorders that was specific for each patient was to restore their motor abilities and quality of life, decreased or lost due to the development of orthopedic complications natural for the pathogenesis of CP spastic types. Another important goal of treatment was the elimination or reduction of pain caused by early arthrosis in the femoro-patellar joint and/or joints of the foot.

Finally, patients in who "gradual fibrotomies" or lengthening of the Achilles tendon were performed in early childhood, iatrogenic orthopedic complications associated with the development of excessive passive range of joint motion (primarily, pathologically excessive dorsal foot flexion) and a decrease in arbitrary muscle contraction strength were revealed. In particular, the combination of triple foot arthrodesis with shortening of the Achilles tendon and/or transferring of the peroneal muscle to the calcaneal bone was aimed at overcoming the weakness of the triceps.

Quantitative and detailed assessment of the operation outcome and subsequent rehabilitation require a separate in-depth analysis based on the study of gait parameters in the pre- and long-term postoperative period and the study of the quality of life. It was not a purpose of this work.

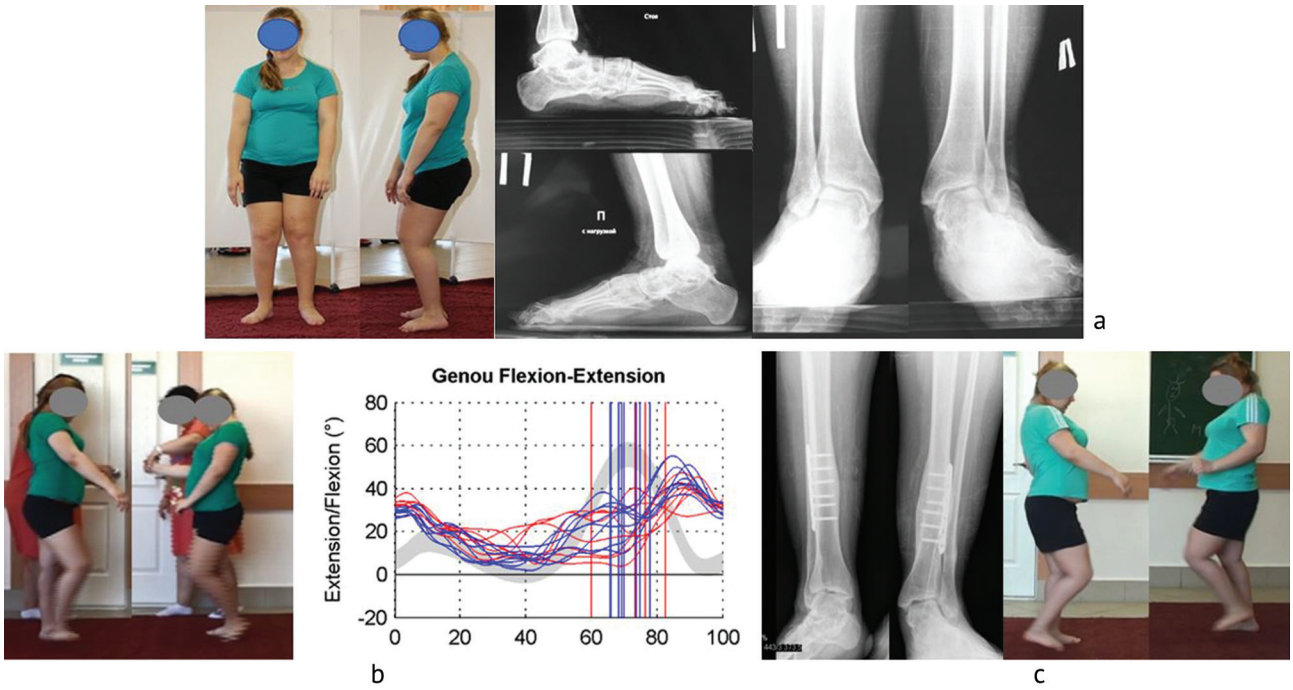

Fig. 1 Patient, 16 years old, GMFCS level II, stiff knee gait, had previous bilateral triple arthrodesis of the feet: **a** – photo of the patient and radiographs of the feet show an excessive external rotation of the feet due to torsional deformity of the lower legs, flexion in the knee joints; **b** – photo of the patient and kinematogram that revealed that the maximum angle of flexion in the knee joint in the non-support phase varied between 38° and 45°, the onset of the flexion peak in the knee joint exceeded 80 % of the step cycle; **c** – patient underwent simultaneous bilateral detorsion osteotomy of the tibias, plate osteosynthesis as well as distal transfer of m. rectus femoris on the semitendinosus muscle and bringing the patellae down; 1.5 years after the operation, the patient is able to move independently; the maximum angle of flexion in the knee joint in the non-support phase of the step cycle varies between 60 and 68°

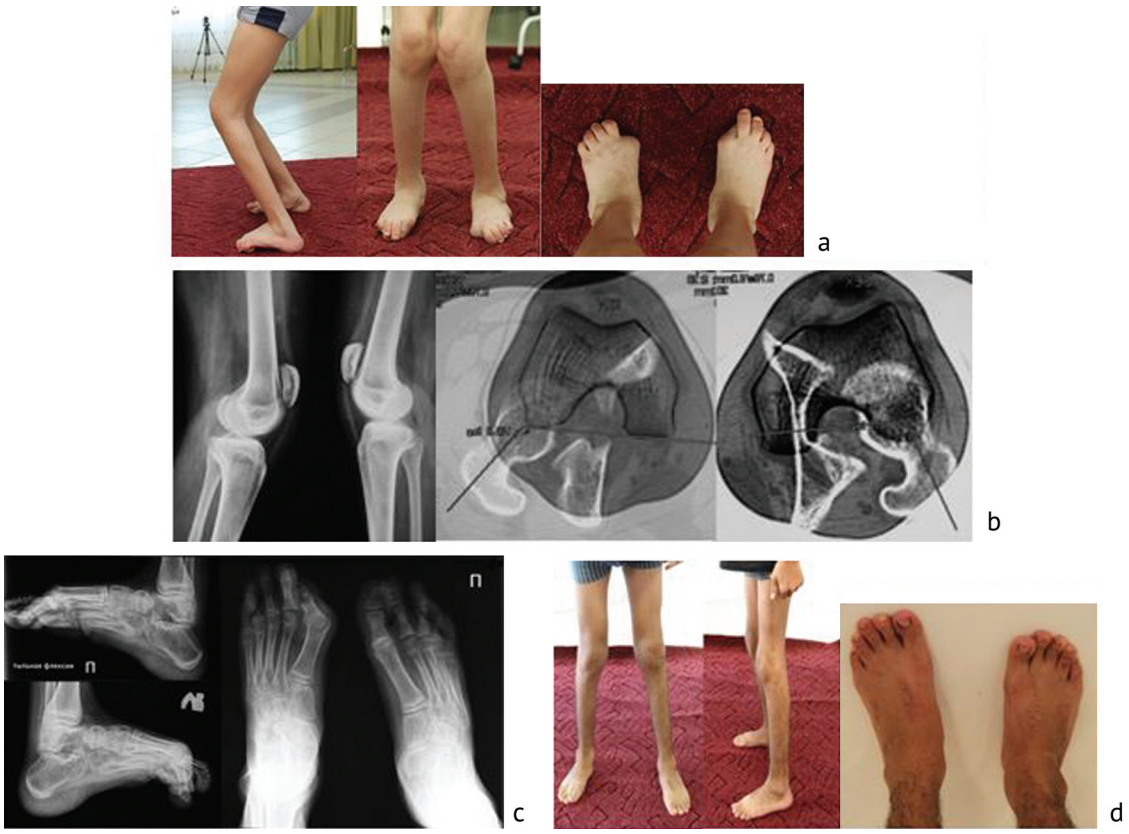

Fig. 2 Patient D., 18 years old, GMFCS level II, had previous "staged fibrotomies", after which he developed weakness of the triceps muscles of the lower legs, excessive dorsal flexion of the feet and secondary flexion contractures of the knee joints: **a** – photo of the lower limbs before the operation; **b** – radiographs of the knee joints and CT of the femurs showing the pathologically high position of the patella, torsional deformities of the femurs; **c** – radiographs of the feet; **d** – photo of the patient's lower limbs one year after performing bilateral intervention involving a supracondylar detorsion extension osteotomy of the femurs, bringing the patellae down, shortening of the Achilles tendon, and transfer of the long peroneal muscle to the calcaneal bone, arthrodesis of the first metatarsophalangeal joint

Continuation of Fig. 2 Patient D., 18 years old, GMFCS level II, had previous "staged fibrotomies", after which he developed weakness of the triceps muscles of the lower legs, excessive dorsal flexion of the feet and secondary flexion contractures of the knee joints: **e** – radiographs of the feet after arthrodesis of the first metatarsophalangeal joint, a year after the detorsion extenson osteotomy of the femurs and bringing the patellae down

DISCUSSION

Currently, cerebral palsy is the most common cause of motor disorders that occur in early childhood and are present throughout the life of a person [9, 32]. Its incidence is one case per 500 newborns. There are more than 17 million people with cerebral palsy in the world [1, 2, 9, 33–36]. The incidence of this pathology has been growing since the 90s of the last century and is associated with an increase in the survival rate of premature babies [9, 11, 25, 37]. In addition, patients with cerebral palsy live nowadays longer due to improved medical and, in particular, surgical care [7, 19, 32, 38, 39]. It is known that 99 % of children with cerebral palsy but without severe functional impairment (GMFCS levels I-III) become adults [11, 22].

Despite that cerebral palsy is not a progressive neurological disease, functional impairment in maturing patients develops in the natural course of the disease. Motor abilities, self-care, socialization, quality of life due to progression of orthopedic problems and pain associated with early degenerative arthrosis deteriorate [5, 24].

Adult patients with cerebral palsy complain of pain, fatigue, decreased functionality as the main factors reducing their quality of life. In the study of Andersson C et al. (2001) [26], the factors worsening the health in 221 adults suffering from cerebral palsy were contractures in 80 % of cases, pain in 18 %, and progressive deterioration of walking ability in 35 % of cases. Among 562 French adults with cerebral palsy from the province of Brittany in average age of 36 years, 82 % suffered from mechanical pain located in the joints of the lower extremities and the spine [27]. Finally, Benner JL et al. (2017), in a survey of 49 adults with cerebral palsy (Netherlands) aged 35 to 45 years found that progressive pain, fatigue by walking and limitation of self-care are the most important reasons of impairment of quality of life and motor abilities in 32 % of cases [6].

In a review article on orthopedic complications of cerebral palsy in adult patients, K.P. Murphy identified four groups of problems [24]:

1) patella alta – a pathologically high position of the patella and the entire range of associated disorders combined in the abnormal gait (crouch gait): torsional deformities of the femur and tibia, external deviation of the foot, planovalgus foot, permanent dorsal foot flexion during walking that condition the dysfunction of the levers in a weight-bearing step phase, as well as femoro-patellar arthrosis

2) dislocation of the femur, early coxarthrosis, loss of verticalization in patients with severe neurological disorders;

3) spondylolysis, most common at L5–S1 level;

4) cervical spine stenosis.

Among 105 adult patients able to walk independently (GMFCS levels I-III) with or without support tools, H.M. Horstmann et al. [11] identified the following orthopedic problems in the lower limbs which required surgery: foot equinus (contracture of the ankle), planovalgus deformity, equinovarus foot deformity, valgus deformity of the first toe, severe flexion contracture of the first metatarsophalangeal joint, flexion contracture of the knee joint, pathologically high position of the patella, torsional deformities of the femur and/or tibia, contracture of the hip joint. These problems were complicated by early arthritic changes in the joints, osteochondromalacia

of the patella and progressive pain. In our study, adolescents and adults with cerebral palsy had the same elements of orthopedic pathology requiring surgical correction.

Horstmann H.M. et al. [11], Ławniczak D. et al. [40], Gannotti M.E. et al. [41], Opheim A. et al. [42], Lee S.Y. et al. [43], Lehtonen K. et al. [44], Novacheck T.F. et al. [45], Putz C. et al. [46, 47] formulated and presented the basic principles and approaches to the surgical treatment of adults with cerebral palsy.

Among the principles of planning and performing orthopedic surgical treatment, the authors focus on the mandatory 3D gait analysis with the study of kinematic, kinetic parameters and dynamic EMG and the implementation of single-event multi-level interventions, taking into account the systemic nature of the damage to the motor apparatus in cerebral palsy. The most common types of gait disorders in adults are stiff knee gait, crouch gait, apparent equinus gait and asymmetric gait. It is generally accepted that goal-oriented surgery is aimed at eliminating orthopedic disorders, increasing mobility, patient's self-care, and improving care for them. These goals are achieved through correction of deformities to improve the biomechanics of movements, eliminate joint contractures to improve daily physical activity and increase functionality, reduce energy waste associated with biomechanical disorders, relieve pain caused by arthritis, deformities, reduce or eliminate problems of using shoes and orthosis products. Finally, it is recognized that the principles of orthopedic surgical treatment in adult patients should be used from the moment the natural growth of the segments is completed and the physes of the bones of the lower extremities close.

3D gait analysis becomes most important when planning the correction of orthopedic deformities in patients with stiff knee gait and crouch gait. The key element of the intervention in the first case is the distal transfer of the rectus femoris to the flexors of the knee joint to bypass the pathological activity of the muscle in the non-weight-bearing phase of the step. This contributes to an increase in the angular and linear velocity of movements in the knee joint and brings the gait parameters closer to those of a healthy adult [40]. Currently, the indications for this operation in children have been defined: peak flexion of the knee joint (PKF) less than 64°, range of knee joint motion (KROM) less than 53°, moment of maximum knee joint flexion (TiPKF) later than 80 % of the step cycle [48]. These indicators could be used at this stage when planning an intervention in adult patients. Lee SY et al. [43] showed, during the observation period of more than two years after this type of intervention in 290 pediatric patients, a statistically significant increase in PKF, KROM, onset of TiPKF early by 5.4 %. Results were better in patients with GMFCS levels I and II than for GMFCS III.

Correction of torsion deformities, isolated or in combination with extension supracondylar osteotomies, when correcting leverage dysfunction in the stance phase of a step in patients with crouch gait [40], improves overall motor activity and social integration of patients [49]. Putz C. et al. [47] studied long-term results of treatment in 63 adults using a laboratory of motion analysis and found a significant improvement in the rotational position of the thigh in the stance phase of the step, in the orientation angle of the axis of the foot by walking, in motion parameters in the knee joint throughout the step cycle, as well as in clinical indicators of hip joint movements. The authors noted that the results are more predictable in adults than in children by planning and performing correction of torsion deformities. It is known that there is a weak correlation between the anatomical values of torsion angles and gait analysis parameters [50].

Various options for correction of foot deformities are performed in adult patients with cerebral palsy [11]. However, they are part of single-event multi-level interventions in most cases. The objectives of operations on the foot are the restoration of weight-bearing ability, easier wear of shoes and orthotic products, relief or elimination of pain, elimination of trophic lesions of integumentary tissues, improvement of conditions for hygiene, and foot appearance. There is a widespread opinion in the literature that with fixed, rigid deformities, especially in the presence of pain, various types of arthrodesis are indicated or arthrodesis in combination with corrective osteotomies [11, 19, 51–57]. Trehan S.K. et al. [58] studied long-term results of triple joint arthrodesis (26 feet), performed at the average age of 19.4 years. Ten years after surgery, a good anatomical and functional result was maintained in 95.2 % of cases, and 61.9 % of patients did not have any pain. In equinovarus non-rigid foot deformity, hemitransfer of

the anterior tibial muscle tendon on the lateral part of the foot (preferably the cuboid bone) [11, 12, 55, 59] is an option. In adolescents and adults, in arthrosis of the first metatarsophalangeal joint as well as in marked recurrence of valgus deformity of the first toe, arthrodesis of the first metatarsophalangeal joint is used. Its outcomes are excellent in up to 75 % and good in 25 % of cases [11, 20, 60]. These approaches were used by us when planning the treatment in our patients.

In the literature, the problem of restoring motor activity after excessive surgical elongation of the muscle groups and iatrogenic dysfunction of the limbs resulting due to it, including of walking, remains practically unsolved. For example, panarthrodesis of the foot was shown as the only way to restore support after surgical overlengthening of the Achilles tendon in patients with spastic diplegia [20]. We also found a relatively small amount of literature on the use of multi-level interventions in adult patients, where not only the technical aspects of interventions and immediate results were discussed, but the functional result at long term was evaluated both in terms of changes in the motor activity and changes in the quality of life of patients. If surgical treatment is carried out only in adulthood, an issue about the possibility of returning patients to the highest possible functional level that existed before the development of orthopedic complications remains open [41, 44].

Finally, we would like to discuss the organizational aspects of the provision of orthopedic surgical care to adults with cerebral palsy raised in the literature. Similar to the situation with children suffering from cerebral palsy, there is no or there is an extremely small number of specialized institutions and departments. There is a shortage of orthopedic surgeons in the adult network with experience and/or specialization in the field of neuro-orthopedics; orthopedic care is fragmented and is provided by orthopedists along with the care of other pathologies in the general flow of patients [9, 10, 11]. The proposed solutions to this problem are diverse: creation and maintenance of age and technological continuity of providing assistance within a single institution; operations performed by orthopedists specialized in neuro-orthopedics for all age categories of patients; specialization of orthopedists of "adult" network in neuro-orthopedics; assistance to adult patients within the framework of multi-competent centers, for example, like at the Penn Comprehensive Neuroscience Center [11, 28].

CONCLUSION

Thus, orthopedic management of adult patients with cerebral palsy remains a relevant issue. One should expect an increase in the number of such patients in the coming years. Performance of multi-level orthopedic interventions is also justified in patients who have completed their physiological growth. Indications and the scope of interventions require objective methods for assessing motor abilities. It is important to develop techniques to perform interventions to ensure early functional activity after multi-level interventions. It is required to justify the prediction of functional outcomes in patients who are surgically treated in adulthood. Finally, orthopedic surgical treatment in adult patients with cerebral palsy needs the medical personnel and the institution specialized in neuro-orthopedics.

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Information about the authors:

1. Orkhan I. Gatamov, M.D.,
Russian Ilizarov Scientific Centre for Restorative Traumatology and Orthopaedics, Kurgan, Russian Federation
2. Georgy M. Chibirov, M.D., Ph.D.,
Russian Ilizarov Scientific Centre for Restorative Traumatology and Orthopaedics, Kurgan, Russian Federation
3. Dmitry Yu. Borzunov, M.D., Ph.D.,
[1]Russian Ilizarov Scientific Center for Restorative Traumatology and Orthopaedics, Kurgan, Russian Federation,
[2]Tyumen State Medical University, Tyumen, Russian Federation
4. Dmitry A. Popkov, M.D., Ph.D.,
Russian Ilizarov Scientific Centre for Restorative Traumatology and Orthopaedics, Kurgan, Russian Federation,
Email: dpopkov@mail.ru