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FUNCTIONAL STATUS OF PERIPHERAL BLOOD FLOW AND NEUROMUSCULAR SYSTEM OF THE LOWER EXTREMITIES AFTER SURGICAL TREATMENT OF CONGENITAL FALSE JOINT OF THE LOWER LEG

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The aim of this study was to evaluate the clinical and functional state of the neuromuscular system and the blood supply to the lower limbs of children with congenital pseudarthrosis of the tibia (CPT) after consolidation. Material and Methods. A total of 100 patients with CPT were analyzed. We performed a clinical examination of patients, panoramic X-ray of the lower extremities, electroneuromyogram, and reovasography. Results and Conclusions. The primary complaints of patients with CPT after the consolidation of the non-union were lameness, deformations of lower extremities, and pain in the local joints. The electromyoneuromyogram data of the lower limbs of patients with CPT exhibited a decrease of the contractility of the muscles of the lower limbs, and neuropathy of the peroneal nerves of both lower limbs. The reovasography data of the lower limbs of patients with CPT displayed improvement in blood circulation in the lower extremities after the consolidation of the tibia. These data promote the current methods of treatment of patients with CPT; however, the temperature, degree of limb lengthening, and deformity correction should be considered in the future.

Keywords: congenital pseudarthrosis of the tibia, correction deformity, neuropathy, shortening.

Introduction:

The issue of the possibility of deformities correction in patients with congenital false joint of lower-leg bones (CFJLL) after consolidation of the pseudarthrosis is still open [1, 2, 3, 4]. Multi-level and multi-component deformities of the lower extremity require complex corrections as well as the use of modern bone-holding devices. However, these surgical interventions are accompanied by the risk of pseudarthrosis re-formation and loss of weight bearing for the affected lower extremity [1].

Study objective:

This study was designed to evaluate the clinical findings and functional status of affected lower extremities in children with CFJLL of different origins after successful surgical treatment of false joint before and after deformities correction.

Study materials and methods:

The study included 100 children and adolescents (50 males and 50 females) aged 3–18 years with CFJLL of different origins. They had previously been followed up and were successfully treated for lower-leg false joint consolidation at the Turner Institute for Children. Patients were admitted for deformities correction.

All patients underwent an orthopedic examination to determine the range of motion in the joints of lower extremities. Panoramic radiography, electroneuromyography (ENMG), and rheovasography (RVG) of the lower extremities were also performed.

All patients had a combination of lower-leg shaft deformation and shortening. According to the classification of A.P. Pozdeev (1984), based on etiological factors of CFJLL, the majority of patients had neurofibromatosis (62 patients, 62%), 26% (26 patients) had myelodysplasia and incontinence,

and 12% (12 patients) had CFJLL secondary to fibrous dysplasia. The distribution of patients by sex and age is presented in Table 1.

The age group 9–14 years had the most number of patients (46%), whereas the group 15–18 years had the least (17%). The 3–8 years group accounted for 37% of patients. The number of boys and girls in each age group was approximately equal.

The functional status of the neuromuscular system for lower extremities was studied using ENMG. We conducted an electromyography (EMG) of lower leg muscles (anterior tibial, gastrocnemius, and peroneal muscles) for shortened and contralateral extremities. Electroneurography (ENG) (peroneal and tibial nerves) allowed us to evaluate the neurological deficit and the level of peripheral nerve damage in the lower extremity.

To study the blood supply in the affected lower extremity after surgical (often multiple) treatment and restoration of continuity in lower leg bones, we performed RVG of the lower extremities. This method was developed to estimate the perfusion of the extremity, flexible and elastic properties of blood vessels, and conditions of capillary blood flow and venous outflow. We analyzed the following parameters: rheovasographic, bisferious, and diastolic indices and assessed the tone of the main vessels.

Table 1

Distribution of patients with CFJLL by sex and age

Patient age groups	Patient sex		Total
	male	female	
	%	%	%
3–8 years	18	19	37
9–14 years	23	23	46
15–18 years	9	8	17
Total	50	50	100

Results

From the analyses of subjective data, including complaints and medical history, we could determine the frequency of the most typical complaints for this group of patients. They were, in descending order as follows: the presence of deformities in the lower extremity (100%), lameness and gait disorders (98%), and pain in adjacent joints (10%). There were no complaints in 6% of the children.

Detailed orthopedic examination allowed us to evaluate the degree of severity of abnormalities in the affected lower extremities and disorders of the contralateral extremities and other body segments. Data on the nature of orthopedic pathology in patients with CFJLL before deformity correction are presented in Table 2.

Table 2

Orthopedic conditions in patients with CFJLL before epithesis

Orthopedic Conditions	%
Knee joint disease: (instability, contractures)	10
Ankle joint disease: – Limitation of dorsal extension – Pseudoankylosis	50 26 24
Foot deformities: – Valgus – Varus – Intoeing – Normal	 66 8 10 18
Foot shortening: Average	95 2 ± 1 sm
Postural abnormalities	100

Other musculoskeletal disorders was observed in all patients with shortened extremities, but this was not a primary complaint in most cases. Postural disorders and deformations of segments of affected lower extremities were observed in all patients. A combination of deformities with shortened lower extremities was diagnosed in 92 patients (92%) with CFJLL of different origins.

The disorders of adjacent joints were as follows: knee joint (10%), ankle joint on the affected side (50%), valgus at the level of talar and subtalar joints (66%), and foot shortening (95%). These were the most prevalent but were probably not primary disorders because of the lack of adequate weight bearing on the affected extremity and multiple surgical treatments. The average length of foot shortening was 2 ± 1 cm.

We also analyzed the magnitude of lower extremity shortening in children with CFJLL before epithesis.

Patients with shortenings of < 4.5 cm were the most prevalent in this study. This group accounted

for 40% of the children who were admitted to the hospital before epithesis. Patients with shortenings of > 9.5 cm accounted for 17% of all patients, those with shortenings of 8.6–9.5 cm accounted for 7%, and those with shortenings of 5.6–6.5 cm and 7.6–8.5 cm accounted for 8% each. Shortening of the affected lower extremity was associated with the following: bone tissue resection during multiple surgical interventions, injury to the distal growth zone of the tibia, and the lack of continuity and support ability of the tibia.

Surface ENG of the lower extremity in patients with CFJLL before epithesis revealed the presence of fibular neuropathy in most cases. There were changes both in the affected lower extremity and on the contralateral side. In most cases, neuropathy was manifested as an axonopathy.

Depending on the CFJLL origin, the highest incidence of fibular neuropathy on the affected side was present in patients with myelodysplasia (75%). Contralateral nerve injury in this group of patients occurred in 25% of patients. Fibular nerve injury on the affected side in patients with neurofibromatosis was observed in 52% of the patients and in 24% of patients on the contralateral side. In patients with fibrous dysplasia, fibular neuropathy was seen in 43% of the patients and only on the affected side. ENG data are shown in Table 4.

Tibial neuropathy was seen only in patients with CFJLL secondary to neurofibromatosis and myelodysplasia and only on the affected side. Neuropathy was not a primary condition; it was the result of multiple surgical interventions. Incidence in both groups was 4% and 9%, respectively.

Contractility studies for the lower leg muscles during EMG showed a decrease in contractility both on the affected and contralateral sides in patients with CFJLL. A decrease of muscle contractility on the affected side was observed in patients with CFJLL secondary to neurofibromatosis (76%) and fibrous dysplasia (64%).

In all patients with CFJLL secondary to myelodysplasia, we noted a decrease of muscle contractility on both lower extremities. The assessment of motoneuron activation in the lumbar and sacral spinal segments showed an injury in an average of 50% of children with CFJLL of different origins both on the affected side and on contralateral lower extremity.

The analysis of blood supply in the lower extremity did not reveal significant abnormalities in RVG parameters relative to normal standards and appropriate parameters of the contralateral lower extremity segments in patients with CFJLL secondary to neurofibromatosis and myelodysplasia. RVG data for these studies are shown in Tables 5–7. Peripheral hemodynamics was quite stable, capillary blood flow was not impeded, and venous outflow was not delayed.

Thus, RVG data showed the absence of marked abnormalities in the circulation of affected lower extremity segments in patients with CFJLL secondary to neurofibromatosis and myelodysplasia. This indicates an adequate body response to the restoration of the continuity of affected extremity bones and the ability of the musculoskeletal and central nervous systems of patients to support rehabilitation of the extremity after surgical treatment.

Table 3

Lower extremity shortening: patient distribution before epithesis

Magnitude of shortening (cm)	Incidence of shortening (%)			
	3–8 years	9–14 years	15–18 years	Total
Up to 4.5	22	12	6	40
4.6–5.5	–	6	–	6
5.6–6.5	3	3	2	8
6.6–7.5	3	3	–	6
7.6–8.5	–	6	2	8
8.6–9.5	2	3	2	7
> 9.5	–	12	5	17
Total	30	45	17	92

Table 4

Results of surface EMG of the lower extremities in patients with CFJLL before epithesis

Parameter	Fibular neuropathy		Tibial neuropathy		Decrease of muscle contractility in lower leg		Dysfunction of motoneuron activation in lumbar and sacral spinal enlargement	
	Affected side	Contralateral extremity	Affected side	Contralateral extremity	Affected side	Bilateral injury	Affected side	Contralateral extremity
	%	%	%	%	%	%	%	%
CFJLL secondary to neurofibromatosis	52	24	4	–	76	56	44	56
CFJLL secondary to myelodysplasia	75	25	9	–	62	62	59	41
CFJLL secondary to fibrous dysplasia	43	–	–	–	64	21	57	43

Table 5

Rheovasographic data in patients with CFJLL secondary to neurofibromatosis

Parameter	Segment			
	Hip		Lower leg	
	Affected extremity	Contralateral extremity	Affected extremity	Contralateral extremity
Rheovasographic index (rel.u.)	0,8 ± 0,05*	0,9 ± 0,03	1,6 ± 0,05	1,4 ± 0,04
Major vessel tone (V_{max})	0,9 ± 0,12	1,0 ± 0,09	2,4 ± 0,12	2,1 ± 0,08
BIS (%)	46,7 ± 5,6	44,5 ± 2,9	31,5 ± 5,3	30,8 ± 3,6
DIA (%)	42,3 ± 6,08	38,1 ± 2,96	36,5 ± 4,2	38,8 ± 3,6

Table 6

Rheovasographic data in patients with CFJLL secondary to myelodysplasia

Parameter	Segment			
	Hip		Lower leg	
	Affected extremity	Contralateral extremity	Affected extremity	Contralateral extremity
Rheovasographic index (rel.u.)	0,7 ± 0,05*	0,7 ± 0,03*	1,3 ± 0,05*	1,3 ± 0,04*
Major vessel tone (V_{max})	0,9 ± 0,02*	0,8 ± 0,05*	2,1 ± 0,02*	2,1 ± 0,04*
BIS (%)	41,4 ± 5,2	41,7 ± 2,0	29,2 ± 5,7	29,3 ± 3,9
DIA (%)	33,7 ± 6,8	34,9 ± 2,9	31,9 ± 4,6	31,6 ± 3,7

Table 7

Data on longitudinal RVG of the lower extremities in patients with CFJLL secondary to fibrous dysplasia

Parameter	Segment			
	Hip		Lower leg	
	Affected extremity	Contralateral extremity	Affected extremity	Contralateral extremity
Rheovasographic index (rel.u.)	1,0 ± 0,05	1,0 ± 0,03	1,9 ± 0,05	1,5 ± 0,04
Major vessel tone (V_{max})	1,2 ± 0,12	1,0 ± 0,09	2,5 ± 0,12	2,1 ± 0,08
BIS (%)	57,0 ± 2,5*	48,8 ± 2,3*	23,7 ± 5,3	26,7 ± 3,6
DIA (%)	48,2 ± 3,5*	38,6 ± 1,4*	24,4 ± 2,0*	33,5 ± 1,6*

The symbol * indicates significantly varying parameters with confidence levels of $p < 0.05$ in comparison with similar parameters of the contralateral segment.

Despite preserved blood flow in patients with CFJLL secondary to fibrous dysplasia, abnormalities in the peripheral hemodynamics both at the level of the hip and the lower leg of the affected extremity in this group of patients were identified. At the hip level this condition was manifested by impeded capillary blood flow, as evidenced by a significant increase in the bisferious index to 57.0% ± 2.5% compared with the unaffected segment (48.8% ± 2.3%), ($p < 0.05$).

In addition, there was delayed venous outflow in the hip of the affected side where the average diastolic index increased significantly to 48.3% ± 3.5% compared with that of the contralateral segment (38.6% ± 1.4%), ($p < 0.05$). Conversely, at the level of the lower leg on the affected side, parameters of capillary blood flow and venous outflow indicated the tendency for the shunting of blood flow to the area of interest (partial discharge of blood from arterial to venous bed, bypassing the capillary network). This is evidenced by the reduction of bisferious and diastolic indices. Moreover, a median resistance of the capillary network was reduced insignificantly up to 23.7% ± 5.3% compared with the value of the contralateral side, which was 26.7% ± 3.6%, ($p > 0.05$). However, in general, the venous outflow was reduced significantly to 24.4% ± 2.0% as against 33.5% ± 1.6%, ($p < 0.05$).

These changes indicate that in patients with CFJLL secondary to fibrous dysplasia, moderate circulatory disorders of the entire affected lower extremity persisted after surgical repair. These disorders were characterized by abnormalities in peripheral blood circulation that ultimately manifested with reduced microcirculation mainly in the area of interest.

In general, the circulation of the affected lower extremities in children with CFJLL of different origin was satisfactorily compensated, which suggests the long term benefits of surgical interventions.

Discussion

The data obtained during a comprehensive study of the lower extremities in children with CFJLL after consolidation of the lower leg bones indicate the involvement of the entire lower extremity in the pathological process, either as a result of disease or as a result of multiple surgical interventions. These results should be considered at the stage of elongation and epithesis of the lower extremities to avoid the development of circulatory disorders of the lower extremity or neuropathies.

Conclusions

On the basis of this study we may conclude the following:

1. Common complaints of patients with CFJLL of different origin after lower leg bone continuity were lameness (in 98% of patients), affected lower extremity deformities (in 80% of patients), and pain in adjacent joints (in 10% of patients).

2. For all patients with CFJLL, the presence of concomitant orthopedic conditions was typical with postural disorders, stiffness of the ankle joint of the affected lower extremity in 50% of patients, and foot shortening in 65% of patients.

3. Surface ENMG revealed a decrease in the contractility of lower leg muscles and fibular neuropathy of axonal origin in both lower extremities. Thus at each stage of treatment, the magnitude of elongation and the speed at which epithesis is performed must be considered.

4. Data obtained from RVG analysis indicated improved circulation in the lower extremities after the restoration of the continuity of the lower leg bones, and the possibility of further surgical treatment aimed at epithesis of the affected lower extremity.

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ФУНКЦИОНАЛЬНОЕ СОСТОЯНИЕ ПЕРИФЕРИЧЕСКОГО КРОВОТОКА И НЕРВНО-МЫШЕЧНОЙ СИСТЕМЫ НИЖНИХ КОНЕЧНОСТЕЙ У ПАЦИЕНТОВ С ВРОЖДЕННЫМ ЛОЖНЫМ СУСТАВОМ КОСТЕЙ ГОЛЕНИ ПОСЛЕ УСТРАНЕНИЯ ПСЕВДОАРТРОЗА

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Введение. Вопросы возможности коррекции деформаций нижней конечности у пациентов с врожденным ложным суставом костей голени (ВЛСКГ) после устранения псевдоартроза большеберцовой кости все еще остаются открытыми. На фоне оперативных вмешательств после достижения консолидации костных фрагментов сохраняется опасность рецидивирования ложного сустава и потери опороспособности пораженной нижней конечности.

Цель исследования. Целью исследования было оценить клиническую картину, функциональное состояние нижних конечностей у детей с ВЛСКГ различной этиологии после консолидации псевдоартроза и перед выполнением последующих коррекций деформаций.

Материалы и методы. Были проанализированы результаты обследования 100 детей и подростков с ВЛСКГ различного генеза в возрасте 3–18 лет. Всем пациентам проводился ортопедический осмотр с определением амплитуды движения в суставах нижних конечностей, выполнялась панорамная рентгенография нижних конечностей, электронейромиография и реовазография нижних конечностей.

Результаты и выводы. На основании проведенного исследования были сделаны выводы, что характерными жалобами пациентов с ВЛСКГ различного генеза после восстановления целостности берцовых костей были: хромота, наличие деформаций пораженной нижней конечности и болевой синдром в смежных суставах. Проведенная поверхностная электронейромиография выявила снижение сократительной способности мышц голеней, аксонопатию малоберцовых нервов обеих нижних конечностей, что необходимо учитывать при определении величины удлинения нижней конечности на каждом этапе лечения и скорости выполнения коррекции деформаций. Данные, полученные при анализе реовазограмм, указали на улучшение показателей кровообращения нижних конечностей после этапа восстановления целостности берцовых костей и возможности выполнения дальнейшего хирургического лечения, направленного на коррекцию деформаций пораженной нижней конечности в сравнении с изучением периферического кровотока у пациентов при наличии псевдоартроза.

Ключевые слова: врожденные ложные суставы костей голени, коррекция деформаций, укорочение, невропатия.

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