UNIVERSITY OF BELGRADE FACULTY OF MEDICINE

Marko V. Aleksić

EVALUATION OF SPLIT TIBIAL TENDON TRANSFERS IN CORRECTION OF VARUS FOOT DEFORMITY AND PREVENTION OF SKELETAL CONTRACTURES IN PATIENTS WITH SPASTIC CEREBRAL PALSY

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Marko V. Aleksić

EVALUACIJA USPEŠNOSTI PARCIJALNIH TIBIJALNIH TETIVNIH TRANSPOZICIJA U KOREKCIJI VARUSNOG DEFORMITETA STOPALA I PREVENCIJI KOŠTANIH KONTRAKTURA KOD PACIJENATA SA SPASTIČNOM FORMOM CEREBRALNE PARALIZE

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SUPERVISOR:

Prof. dr Zoran Baščarević, M.D., PhD, University of Belgrade, Faculty of Medicine, Institute for Orthopaedic Surgery "Banjica"

CO-SUPERVISOR:

Prof. dr Marija Đurić, M.D., PhD, Anatomy Institute, University of Belgrade, Faculty of Medicine

BOARD MEMBERS:

- 1. Prof. dr Marko Bumbaširević, M.D., PhD, University of Belgrade, Faculty of Medicine, Orthopaedic Clinic, Clinical Centre of Serbia
- 2. Prof. dr Lukas Rasulić, M.D., PhD, University of Belgrade, Faculty of Medicine, Neurosurgery Clinic, Clinical Centre of Serbia
- 3. Prof. dr Đorđe Gajdobranski, M.D., PhD, Univeristy of Novi Sad, Faculty of Medicine, Pediatric Surgery Clinic, Institute for Children and Youth Health Care of Vojvodina

Thesis defence date:

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EVALUATION OF SPLIT TIBIAL TENDON TRANSFERS IN CORRECTION OF VARUS FOOT DEFORMITY AND PREVENTION OF SKELETAL CONTRACTURES IN PATIENTS WITH SPASTIC CEREBRAL PALSY

SUMMARY

Introduction. Cerebral palsy (CP) is neurodevelopmental disorder characterized mainly by the motor, movement, and muscle tone abnormalities. Spastic CP if often accompanied by the foot deformities, which can be treated with orthoses, muscle tone management and surgical interventions.

Aim. The aim of the thesis was to determine the results of split tibial tendon transfers in patients with spastic varus foot deformity due to CP, and to determine whether surgical outcome depended on other parameters aside from surgical technique, as well as to determine the incidence of postoperative skeletal contractures.

Methods. Patients were categorized into four groups depending on the surgical intervention performed. One group was treated with split anterior tibial tendon transfer (SPLATT) procedure; second group of patients was treated with split tendon transfer of tibialis posterior muscle (SPOTT) procedure, the third group was operated with the combination of those two procedures (SPLATT+SPOTT), and the fourth group of patients was treated with the modified SPOTT intervention. Gross motor function was assessed preoperatively by the expanded and revised Gross Motor Function Classification Scale (GMFCS). Functional mobility was assessed before and after the operation using the Functional Mobility Scale (FMS). Postoperative change in FMS was categorized as positive, equal, or negative change in relation to preoperative values. Weight-bearing radiographs were used to determine postoperative valgus/varus foot deformity (<5° - neutral, 5° - 10° - mild, and >10° - severe). Postoperative outcomes were assessed using Hoffer's clinical criteria in procedures involving tibialis anterior, and by Kling's clinical criteria for surgical procedures involving tibialis posterior.

Results. In 14 patients (16 feet), SPLATT procedure was performed. Preoperative GMFCS level I was recorded in 8 cases (50%), while 5 cases (31.3%) had level II and 3 cases (18.8%) had level III. In the majority of cases (13 cases, 81.2%) there was a positive change in FMS values, while in remaining 3 cases (18.8%) postoperative FMS values remained unchanged. After median follow up of 7 years (IQR 6.24 – 11.00) excellent/good results were achieved in 14 (87.5%) cases, while poor results were obtained in two (12.5%) cases. There was no significant correlation between the surgical outcome and patients' age at the time of the surgery (ρ =0.136, p=0.615). Postoperative fixed skeletal contracture was recorded in only one patient treated with SPLATT, where triple arthrodesis was performed.

Sixty-six patients (76 feet) were treated with SPOTT procedure. The majority of patients had GMFCS level II (36 cases, 47.4%) and level III (34 cases, 44.7%). Positive postoperative change in FMS was noted in 52 (68.4%) cases, while no change was recorded in 24 (31.6%). Median follow-up was 8 years (IQR 6 – 11 years), with good/excellent results obtained in 65 (85.6%) feet, and poor postoperative outcome recorded in 11 (14.5%) feet. There was no statistically significant difference in surgical outcome compared to patients' gender (χ 2=0.773, p=0.514), and no significant correlation with the patients' age at the time of the surgery (ρ =-0.094, p=0.418). Postoperative fixed skeletal

contracture was recorded in only one patient treated with SPOTT, where triple arthrodesis was performed.

There were 18 patients (18 feet) in SPLATT + SPOTT group. The majority of patients had preoperative GMFCS level III (8 cases, 44.4%) and level II (6 cases, 33.3%). Positive change in FMS was recorded in 16 patients (88.9%), while 2 patients (11.1%) had no change in FMS score before and after the surgical intervention. After the median follow-up of 8.50 years (IQR 4.75 – 10.00), good/excellent results were seen in 13 (72.8%) patients, while 5 patients (27.8%) had poor postoperative outcomes. Postoperative fixed skeletal contracture was recorded in only one patient treated with SPLATT +SPOTT procedure, in which case a triple arthrodesis was performed.

Modified SPOTT was performed in 10 patients (12 feet). There was equal distribution of patients regarding level I, level II, and level III on preoperative GMFCS (4 patients in each group). FMS change was positive in 6 (50%) cases, and equal in other 6 (50%). After median follow-up of 8.5 years, all patients had good/excellent postoperative outcome. There were no postoperative fixed skeletal contractures in patients treated with the modified SPOTT technique.

Conclusion. Split tibial tendon transfers (SPOTT, SPLATT, SPOTT+SPLATT, and modified SPOTT procedure) in patients with spastic foot deformity due to CP, showed good/excellent results in majority of operated patients during the long-term follow-up. Patients treated with modified SPOTT procedure, which was developed by the team led by an experienced surgeon from our institution, showed good/excellent postoperative results in all cases, with no poor postoperative outcomes and no postoperative fixed skeletal contractures.

Keywords: cerebral palsy, varus foot deformity, split tibial tendon transfer, SPOTT, SPLATT

Research area: Medicine

Research field: Skeletal biology

EVALUACIJA USPEŠNOSTI PARCIJALNIH TIBIJALNIH TETIVNIH TRANSPOZICIJA U KOREKCIJI VARUSNOG DEFORMITETA STOPALA I PREVENCIJI KOŠTANIH KONTRAKTURA KOD PACIJENATA SA SPASTIČNOM FORMOM CEREBRALNE PARALIZE

REZIME

Uvod. Cerebralna paraliza (CP) je neuromišićni poremećaj koji se primarno karakteriše poremećajem motorike i mišićnog tonusa. Spastična forma CP često uključuje poremećaje deformacije stopala koje se mogu lečiti ortozama, kontrolom mišićnog spazama i hirurškim intervencijama.

Cilj. Cilj teze je bio određivanje uspešnosti parcijalnih transfera tibijalne tetive kod pacijenata sa spastičnim varusnim deformitetom stopala, i ispitivanje povezanosti ishoda sa drugim parametrima osim same hirurške tehnike, kao i određivanje incidence postoperativnih koštanih kontraktura.

Metode. Operisana stopala su klasifikovana u grupe na osnovu hirurškog metoda korekcije deformiteta. Prvu grupu činila su operisana stopala na kojima je obavljena parcijalna transpozicija prednjeg tibijalnog mišića (SPLATT). Drugu grupu činila su stopala na kojima je obavljena parcijalna transpozicija zadnjeg tibijialnog mišića (SPOTT). Treću grupu činila su stopala na kojima su obavljene obe procedure (SPLATT i SPOTT), dok su četvrtu grupu činila stopala na kojima je obavljena modifikacija SPOTT procedure. Procena preoperativnog opšteg motornog statusa učinjena je na osnovu sistema klasifikacije motornih funkcija (engl. *expanded and revised Gross Motor Function Classification Scale, (GMFCS)*). Ambulatorni status pacijenata pre i nakon operacije procenjivan je na osnovu skale funkcionalne mobilnosti (engl. *Functional Mobility Scale, FMS*). Postoperativna promena vrednosti FMS je opisana kao pozitivna, nepromenjena ili negativna u odnosu na preoperativnog varusnog ili valgusnog deformiteta (manje od 5° - bez deformiteta, od 5° do 10° - blagi deformitet, i preko 10° - izraziti deformitet). Postoperativni ishod je procenjivan na osnovu Hoferovih kliničkih kriterijuma kod procedura vezanih za prednji tibijalni mišić.

Rezultati. Kod 14 pacijenata (16 stopala) sprovedena je SPLATT procedura. Kod 8 pacijenata (50%) preoperativni GMFCS je bio nivo I, dok je kod 5 pacijenata (31,3%) bio II, a kod 3 pacijenta (18.8%) je iznosio III. U većini slučajeva (13 stopala, 81,2%) bilo je pozitivne promene u vrednosti FMS, dok su u preostala 3 slučaja (18,8%) postoperativne vrednosti FMS ostale nepromenjene. Nakon prosečnog praćenja od 7 godina (IQR 6,24 – 11,00), odlični/dobri rezultati su postignuti kod 14 (87,5%) stopala, dok su loši rezultati notirani kod dva (12,5%) stopala. Nije uočena značajna korelacija između hirurškog ishoda i uzrasta pacijenta u vreme procedure (ρ =0,136, p=0,615). Postoperativna skeletna kontraktura zabeležena je kod jednog pacijenta nakon SPLATT procedure, koji je lečen triplom artrodezom.

Kod 66 pacijenata (76 stopala) primenjena je SPOTT procedura. Većina pacijenata je imala GMFCS nivo II (36 stopala, 47,4%) i III (34 stopala, 44,7%). Pozitivna postoperativna promena u vrednosti FMS registrovana je kod 52 (68,4%) slučaja. Prosečno praćenje pacijenata iznosilo je 8 godina (IQR 6 – 11 godina) sa odličnim/dobrim ishodom kod 65 stopala (85,6%), dok je loš ishod notiran kod 11 (14,5%.) slučajeva. Nije uočena statistički značajna razlika u hirurškom ishodu u odnosu na pol (χ 2=0,773, p=0.514) i uzrast pacijenata u vreme hirurške intervencije (ρ =-0,094, p=0,418).

Postoperativna skeletna kontraktura registrovana je kod jednog pacijenta nakon SPOTT procedure, koja je lečena triplom artrodezom.

Ukupno 18 pacijenata (18 stopala) lečeno je SPLATT + SPOTT procedurom. Većina je imala preoperativni GMFCS nivo III (8 pacijenata, 44.4%) i nivo II (6 pacijenata, 33.3%) Pozitivna promena u vrednosti FMS zabeležena je kod 16 pacijenata (88,9%), dok su dva pacijenata (11,1%) imala neizmenjen FMS nakon operacije. Prosečno vreme praćenja pacijenata iznosilo je 8,5 godina IQR 4.75 – 10.00). Odlični/dobri rezultati su registrovani kod 13 (72,8%) pacijenata, dok je kod 5 pacijenata (27,8%) postoperativni ishod okarakterisan kao loš. Postoperativna skeletna kontraktura postojala je kod jednog pacijenta nakon SPLATT + SPOTT procedure, koja je lečena ukočenjem stopala i skočnog zgloba (triplom artrodezom).

Modifikovana SPOTT procedura obavljena je kod 10 pacijenata (12 stopala). Postojala je jedanka raspodela pacijenata po GMFCS nivoima I, II i III (po 4 pacijenta). Pozitivna promena u vrednosti FMS je zabeležena kod polovine pacijenata (n=6), dok je kod preostalih 6 pacijenata vrednost FMS ostala nepromenjena nakon operacije. Nakon prosečnog praćenja pacijenata od 8,5 godina, kod svih pacijenata je registrovan odličan/dobar ishod lečenja. Nije bilo fiksiranih skeletnih kontraktura kod pacijenata u ovoj grupi.

Zaključak. Parcijalni tetivni transferi (SPOTT, SPLATT, SPLATT+SPOTT i modifikovana SPOTT procedura), pokazali su se kao uspešne procedure za rešavanje varusnog deformiteta stopala kod većine pacijenata sa spastičnom formom CP nakon dugoročnog praćenja. Pacijenti kod kojih je primenjena modifikovana SPOTT procedura, razvijena od strane hirurga naše ustanove, pokazala se kao odlična/dobra kod svih pacijenata, bez zabeleženih loših ishoda i bez pojave fiksnih, skeletnih kontraktura zgloba.

Ključne reči: Cerebralna paraliza, varusni deformitet stopala, parcijalni tetivni transfer, SPOTT, SPLATT.

Naučna oblast: Medicina

Užanaučna oblast: Biologija skeleta

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1. INTRODUCTION

1.1. Cerebral Palsy – Definition, Etiology and Classification

Cerebral palsy (CP) is one of the most common neuromuscular abnormalities in children affecting 1 – 5 per 1,000 newborns (1). It is can be defined as a permanent, non-progressive encephalopathy resulting in heterogeneous musculoskeletal and articular manifestations. These abnormalities are often accompanied by the sensation, perception, cognition, communication and behaviour disturbances, by epilepsy and secondary musculoskeletal disorders (2). Standard definition of CP taken from the International consensus and adoption in 2006 states that: "Cerebral palsy describes a group of permanent disorders of the development of movement and posture, causing activity limitation, that are attributed to non-progressive disturbances that occurred in the developing fetal or infant brain" (2). Although the underlying CP encephalopathy is static and non-progressive, the motor, musculoskeletal and articular problems can evolve and worsen over time, and, in a way, be "progressive" in that sense (3).

Etiology of CP is diverse and many etiological factors can act individually, but more often act together, leading to the brain damage. These factors have shown variations over time and in the different geographic areas. Traditionally, etiologic factors can be divided into three categories, depending on the affection time (4). Prenatal causes include congenital brain deformations, intrauterine infections, and chromosomal abnormalities. The most common perinatal etiologic factors are: premature birth, fetal hypoxia and ischemia, infections of central nervous system (CNS), fetal stroke, birth trauma, as well as low birth body weight. Postnatal causes of CP include head trauma, CNS infections, and neonatal stroke (4).

In general, the most significant risk factors for CP include: prematurity, followed by multiple gestation, preeclampsia, chorioamnionitis, abnormal placental pathology, and intrauterine growth restriction. In exception of infrequent disorders, such as familial spastic paraparesis and congenital ataxia, no known genetic factor is present in the etiology of CP (4).

Classification of CP depends on the 4 major elements (2). First is the presence of motor abnormalities, where the nature and type of the motor disorder are considered, along with the functional motor abilities. The second determinant covers the accompanying musculoskeletal and/or non-motor neurodevelopmental abnormalities (e.g. sensory problems, cognitive deficit etc.). The third important component for the CP classification is the anatomic distribution and the neuro-imaging findings. The fourth determinant reflects the cause of CP (known/unknown) and the timing of its affection (prenatal/perinatal/postnatal). Since CP is largely characterized by motor abnormalities, it is important to assess muscle tone abnormality or involuntary movement in each patient. Depending on the predominant type of the motor disorder, patients are grouped into spastic, dyskinetic, ataxic or mixed types, where the "mixed" category is reserved for patients where none of the three major types dominate (5). In routine clinical practice, CP is often categorized depending on the type of the muscle tone abnormality combined with the anatomic region that is affected (6). Therefore, patients are usually categorized into spastic diplegic, spastic hemiplegic, spastic quadriplegic, dyskinetic, dyskinetic, dystonic, ataxic, and mixed types (the terms "plegia" or "plegic" and "paresis" are used interchangeably in the literature) (7). In recent time, other classifications have been developed that

are function-based. These functional classifications systems are able to grade individuals based on their abilities, instead of their deficiencies which is in concordance with the World Health Organization's International Classification of Functioning, Disability, and Health, which focuses on activity and participation. The first widely accepted functional classification was the Gross Motor Function Classification System (GMFCS). Initially described by Palisano et al. (8), this five-level ordinal grading system has been found to be a reliable and stable method of classification and prediction of motor function for children under the age of 12 years (Figure 1). Its' expanded and revised form now includes children that are 12 to 18 years of age (9, Figure 2). It also takes into account the need for assistive devices, such as walkers and wheelchairs, and the quality of movement is age-based. The emphasis of this scale is on voluntary movement and walking as well as on sitting function. The GMFCS level only serves as a guideline for prognosis of the children's long-term functional outcome (10,11).

The Functional mobility scale (FMS) is a classification system specifically devised for ambulation status in CP patients. It has a proven ability to detect change following surgery (12). Ambulation is categorized for three walking distances (5, 50 and 500 m). It includes assessment of the need for walking aids and assistance for ambulation on different surfaces, using a six-degree ordinal scale for each walking distance (13). Postoperative FMT value is then characterized as a positive or negative change in comparison to a preoperative value (14).

GMFCS E & R between 6th and 12th birthday: Descriptors and illustrations

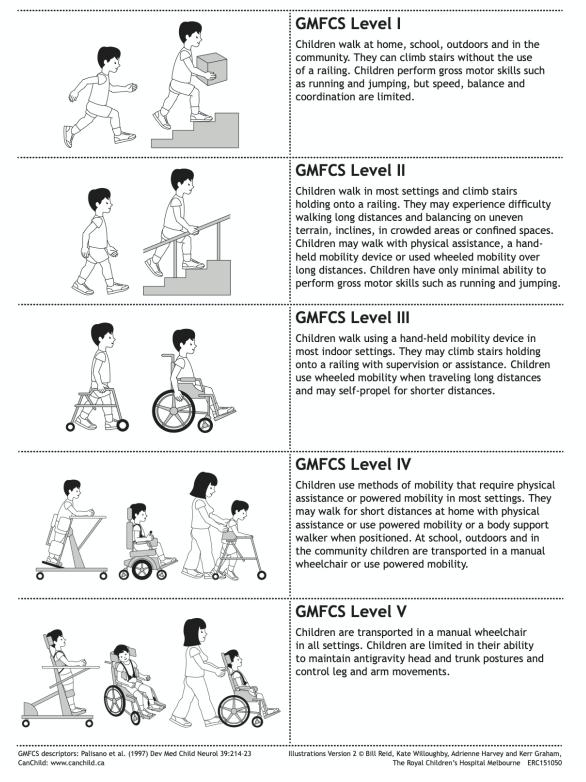


Figure 1. GMFCS E&R between 6th and 12th birthday: Descriptors and illustrations. (Taken from: CanChild: www.canchild.ca)

GMFCS E & R between 12th and 18th birthday: Descriptors and illustrations

	GMFCS Level I Youth walk at home, school, outdoors and in the community. Youth are able to climb curbs and stairs without physical assistance or a railing. They perform gross motor skills such as running and jumping but speed, balance and coordination are limited.
	GMFCS Level II Youth walk in most settings but environmental factors and personal choice influence mobility choices. At school or work they may require a hand held mobility device for safety and climb stairs holding onto a railing. Outdoors and in the community youth may use wheeled mobility when traveling long distances.
	GMFCS Level III Youth are capable of walking using a hand-held mobility device. Youth may climb stairs holding onto a railing with supervision or assistance. At school they may self-propel a manual wheelchair or use powered mobility. Outdoors and in the community youth are transported in a wheelchair or use powered mobility.
	GMFCS Level IV Youth use wheeled mobility in most settings. Physical assistance of 1-2 people is required for transfers. Indoors, youth may walk short distances with physical assistance, use wheeled mobility or a body support walker when positioned. They may operate a powered chair, otherwise are transported in a manual wheelchair.
	GMFCS Level V Youth are transported in a manual wheelchair in all settings. Youth are limited in their ability to maintain antigravity head and trunk postures and control leg and arm movements. Self-mobility is severely limited, even with the use of assistive technology.
GMFCS descriptors: Palisano et al. (1997) Dev Med Child Neurol 39:214-23 CanChild: www.canchild.ca	Illustrations Version 2 © Bill Reid, Kate Willoughby, Adrienne Harvey and Kerr Graham, The Royal Children's Hospital Melbourne ERC151050

Figure 2. GMFCS E&R between 12th and 18th birthday: Descriptors and illustrations. (Taken from: CanChild: www.canchild.ca)

1.2. Diagnosis of CP

In children younger than 2 years, diagnosing CP can be exceedingly difficult. This is even more pronounced if the child can walk by that age. This can lead to many false positive diagnoses. Deonna et al. (15) showed that over half of the patients diagnosed with CP by the age of one, did not qualify for the same diagnosis by the age of 7. Transient dystonia of prematurity can often be confused with CP. This condition can mimic CP as it can also lead to increased tone in the lower extremities. It usually manifests itself in the first or second year of life. Unlike CP, transient dystonia is self-limiting and it resolves with no specific treatment needed (16).

The specific clinical features of CP are best recognized at the age of 3 to 5 years, yet some suggestive features can be present in infancy. CP is best diagnosed with the combination of anamnesis, clinical examination, neuroimaging techniques and developmental assessment tools. Patient's anamnesis should be thorough, especially with the details about the pregnancy and delivery (10). Differential diagnosis of CP includes many neurodegenerative and developmental abnormalities, other neuromuscular disorders, movement disorders and neoplasms, which should be carefully considered when diagnosing certain CP forms (17,18). Additional diagnostics, including imaging (CT scan, MRI, and PET) are used occasionally, especially to determine the exact type and extent of the condition (19).

It is essential to be familiar with physiological motor developmental milestones as well as primitive reflexes that occur during childhood. This allows for much easier identification of patients with delayed motor development. Cephalad-to-caudal pathway is usually the normal direction of motor development. It starts with swallowing and sucking. These signs are present at birth. The next milestone occurs usually at two and a half years of age and is characterized by gaining sphincter control (20). Primitive reflexes of motor activity gradually disappear as a part of the normal developmental process. In children with neuromuscular disorders, specifically cerebral palsy, primitive reflexes can last longer than normal, and in some cases can even become permanent. Development of more mature motor patterns needed for the development of walking function can be considerably delayed or even absent. Patient's neurological age can be determined by taking into account all the primitive and mature motor reflexes. Neurologic quotient is calculated by comparing the chronologic age to the neurologic age. It is used to determine prognosis as well as to guide treatment (20).

1.3. Prognostic Factors

Determining prognostic factors is essential to guide treatment in patients with CP. When tonic neck reflexes are present, patients are less likely to have independent standing balance and have the ability for independent ambulation. If a patient is sitting independently by 2 years of age, there is a good chance that the patient will walk without assistance (21). If a child cannot sit without aid by 4 years of age, possibility of unassisted ambulation is highly doubtful. Independent walking will likely never happen if a child is not ambulatory by the age of 8 (21).

Bleck et al found several poor prognostic signs which included: persistent Moro reflex, an imposable asymmetric tonic neck reflex, absence of normal parachute reaction after 11 months of age, persistent

neck-righting reflex as well as strong extensor thrust on vertical suspension. If these primitive reflexes remain, they are usually linked with severe brain damage leading to a poor prognosis for independent walking, and inability to do other basic and essential activities of daily life. (22)

Clinical observation is still the most important element in diagnosing gait abnormalities in children with CP. This is done by watching the patient from all sides during his walking cycle, studying one component of gait at a time. Attention is given to the movements, position and rotational alignment of the trunk, pelvis and major joints of the leg and foot, as well as to stride length and cadence (23).

Quantitative gait analysis is a system that uses special, high-speed video capture devices that record from different viewpoints, sensors on the surface of the skin are placed on bony landmarks, while the patient is walking on a force platform. It can measure many valuable gait components. The acquired data is presented in a waveform that represents the joint movement in all three dimensions during the gait cycle (24). Dynamic electromyography (EMG) is used to record the activation of various muscles during the gait cycle. It is also used to determine which muscles are firing in a normal pattern and which are firing out of phase (25). Combined with dynamic foot pressure measurements (pedobarography) and oxygen consumption measurements, it can give detailed information about all gait components (26). Although quantitative gait analysis provides objective data, interpretation of that data is subjective. It presents only low to moderate interobserver reliability. There are substantial differences in diagnosis and treatment recommendations based on quantitative gait analysis in different hospitals, so its role in the treatment of CP patients remains controversial (27). More studies are needed to determine if its use leads to improved clinical outcomes.

1.4. Spastic CP

Spastic form represents the most common subtype of CP, accounting for 80-90% of all cases (5). This form is characterized by the upper motor neuron signs, reflecting as muscle hypertonia, hyperreflexia, clonus and pathological reflexes and muscular movement resistance (28). Spasticity is defined as an increased resistance to the joint movement, intensified with movement velocity (29). Additionally, muscle resistance differs with the direction of joint movement (30).

Spasticity most often derives from several pathophysiologic pathways (4). The most common mechanisms include loss of control in major motor pathways, such as corticospinal, reticulospinal, and vestibulospinal pathway (31).

Longstanding skeletal muscles spasms can result in the loss of their elasticity, marked reduction in the muscle growth and the loss of the muscle volume (32). Spastic muscle bellies appear shorter and thinner, while their tendons are elongated, and often thickened. Continuing spasticity can lead to muscle retraction, interstitial degeneration, and fibrosis, which in turn lead to contractures, articular deformities, skeletal deformities, and degenerative changes of articular cartilage (33,34).

Spastic CP can be categorized by the geographical pattern of involvement as: (1) hemiplegic (or hemiparesis, one-sided) CP, and (2) both-sided – diplegic (diparesis) and quadriplegic (or quadriparesis) CP. There are also less common patterns like monoplegic, biplegic, paraplegic, bilateral hemiplegic and triplegic (2). It is often not easy to completely designate the precise pattern

of involvement geographically. This is because some extremities can be only mildly affected and patient's true pattern of involvement can change during growth and development (2).

1.4.1. Hemiplegic CP

Hemiplegic CP is characterized by the spastic paresis of the one side of the body, affecting one third of patients with spastic form of CP (2). Usually the upper extremity is more severely affected than the lower extremity on the same side, with the right-sided hemiplegia being more prevalent than the affection of the left side of the body. The most common abnormalities seen in these patients include impaired extension of the wrist, supination of the forearm, dorsiflexion and aversion of the foot, flexion of the elbow and wrist, equinus position of the foot. Sensory abnormalities are frequently present in the affected limbs (in around 70% of patients) (35). Epilepsy, cognitive impairment, visual defects, and facial nerve palsy is often seen in these patients (36).

Spastic hemiplegia is usually associated with the infarction or trauma of the brain, and periventricular haemorrhagic infarction resulting in cystic encephalomalacia (4).

1.4.2. Diplegic CP

Spastic diplegia is characterized by the bilateral affection particularly of the lower limbs. Upper extremities are affected to a lesser extent that can be observed when performing fine motor functions (2). Prevalence of diplegic CP is between 25 - 50% of spastic CP. This form of spastic CP was first described in 1853 by the surgeon W.J. Little, and is historically known as Little's disease (Morbus Little) (37).

In patients with spastic diplegic cerebral palsy, there is a characteristic gait pattern including increased knee flexion in the stance phase of the gait cycle (38). There is usually a weakness in the three major groups of muscles responsible for resisting the tendency toward the development of a crouch gait, but the majority of younger individuals walk with a reasonably upright posture in early childhood (39).

Deformities (termed lever arm deformities) that are frequently seen in adolescents with spastic diplegia are excessive femoral anteversion, hip subluxation, patella alta, excessive external tibial torsion, and pes valgus (40). Spasticity of the lower limbs is often accompanied by multiple deformities such as adduction, flexion and internal rotation of the hip, external rotation of the lower leg, equinus, and varus of the foot (41). These motor abnormalities are often accompanied by speech impairment, problems with visual perception, convergent strabismus, and epilepsy (42).

The main causes of diplegic CP are the prematurity and low birthweight, as well as other perinatal and postnatal etiologic factors (43). The most common neuroimaging feature of diplegic CP is the periventricular leukomalacia (44).

1.4.3. Quadriplegic CP

Quadriplegic CP represents the most severe form of CP, characterized by the involvement of all four extremities. Prevalence of this form of spastic CP is about 3 - 35%. Usually the upper extremities are

more severely affected, with the abnormalities similar to those seen in hemiplegic CP (2). The majority of these patients are not able to walk individually. Neuroimaging shows diverse and extensive abnormalities, accompanied by the cystic encephalomalacia. This form of CP is the most frequently associated with intrapartum hypoxia. Patients often experience seizures and optic atrophy, while cognitive impairment is present in almost all cases (45). The most severe CP patients are those with total body involvement. In addition to quadriplegia, they also have loss of head and neck control as well as many severe cognitive deficits (46). These patients usually require full-time assistance for overcoming challenges of daily living and special seating systems that include head and neck support.

1.5. Treatment of spastic CP

Aim of CP treatment is to improve patients medical, social, and psychological aspects of life, and to provide educational support. Management of the cerebral palsy patient mandates a multidisciplinary approach by different physician subspecialists; physical, occupational, and speech therapists, and nursing staff. The orthopedic surgeon's role is to assess the biomechanical aspects of spasticity and address malalignments, deformities, or contractures. The neurosurgeon's role is to modulate movement disorders such as spasticity and dystonia at the level of the brain or spinal cord. Any surgical intervention must be done in concert with other spasticity management specialists and appropriate postoperative therapy (47).

Spasticity can be managed in several ways. Oral medications (such as benzodiazepines, baclofen, and tizanidine) are appropriate for children with generalized spasticity in cases where the mild tone reduction is needed (48). Injection of botulinum toxin acts by the chemo denervation, namely by the inhibition of the presynaptic release of acetylcholine which results in the muscle tone decrease. This approach is useful in children without fixed contractures, in order to relieve their local spasticity (49). Intrathecal baclofen administration results in higher drug concentration in cerebrospinal fluid than after the oral administration. Since the way of drug administration is very delicate, this approach can be associated with serious complications (50).

Despite the medical treatment, children with spastic CP often require surgical interventions. Therefore, the spastic form of CP is encountered most often in orthopedic surgeons' routine clinical practice.

1.6. Foot deformities in spastic CP

Foot and ankle deformities are very often seen in children with CP, as they comprise up to 90% of cases (51). Abnormal foot position and deformities are caused by the spasticity of certain muscles or muscle groups that overpower their normal strength or weakened antagonists. Deformities of the foot can be mild and resolve using nonoperative techniques (such as orthosis and physical therapy with stretching exercise), but they can also be severe, rigid, and complicated, and therefore be challenging for the orthopedic surgeon (52). As the deformities progress, they can compromise walking and standing ability by affecting the stability of foot. During the early phase, foot deformities in CP are often dynamic and flexible. However, when left untreated, such deformities can progress and lead to

osseous changes, with severe joint deformities that are fixed (52). These deformities are more impairing for the patient, harder to treat, usually needing bone surgery, like corrective osteotomies and arthrodesis. These treatments also have a higher rate of complications compared to soft tissue surgeries (53).

The most common deformities of the foot and ankle region are the equinus deformity, valgus and varus foot deformity, as well as the combinations of these deformities, such as equinovalgus and equinovarus (54). Toe deformities are often present in patients with CP, namely hallux valgus, hallux flexus, adduction hallux deformity and flexion deformities of toes.

Treatment of these deformities is aimed to relieve any symptoms of pain or skin irritation, and to optimize function. Foot deformities can be treated with several approaches which include orthoses (braces), tone management and surgical interventions (52). Orthoses are applied in order to improve gait in patients with CP. The most commonly used braces are the ankle-foot orthoses. Tone management can be obtained by the local approach (reducing muscle tone in specific muscle, e.g. intramuscular injection of botulinum toxin), and the global approach (medications) (55). Surgical interventions are aimed to treat specific spastic muscles or muscle groups and are developed to correct specific deformities.

1.7. Varus foot deformity in patients with CP

Varus foot deformity refers to an overly medial forming angle of the foot or it's inward angulation in relation to the body's midline axis (56). This deformity is frequently observed in children with CP. In cases where the angulation is directed toward the midline of the longitudinal axis of the calcaneal tuberosity, it is referred as hindfoot varus. Forefoot varus is defined as the elevation of the medial ray of the foot (56).

Varus foot deformity is more functionally impairing for patients, and is more difficult to treat with nonoperative methods. Although varus foot deformity is more challenging for the orthopedic surgeon than the valgus foot, postoperative results are better than in cases of valgus foot deformity (21). Varus foot deformity results from the disbalance between the tibialis muscles (tibialis anterior and tibialis posterior), and peroneal muscles (peroneus longus and peroneus brevis). When the varus foot deformity is predominantly caused by the spasticity of tibialis posterior, accompanied by the spasticity of the gastrocnemius muscle (and Achilles tendon shortening), it is clinically presented as equinovarus (57). When the spasticity of tibialis anterior is the predominant cause of the deformity, there is usually no equinus of the foot, with the varus being present in the middle and anterior part of the foot (32). When both tibialis muscles are affected, the whole foot is then in the varus position.

Incidence of varus foot deformity is around 23%. It is most common in spastic hemiplegia with almost 94% of patients reported to have some degree of this deformity (58). In diplegic and quadriplegic spastic forms, it is considerably rarer in comparison to the valgus foot deformity (36% vs. 64%).

Varus foot deformity is an important impairment factor for patients who are able walk. During gait, varus foot deformity is complicated by painful callosities at the lateral part of the foot (59). Such callosities can progress into ulcerations, or be accompanied by the hypertrophy of the base of the fifth metatarsal bone (58).

Trampling, falling and injury often happen in patients with CP and varus foot deformity, especially if the deformity is severe and rigid. With time, the foot often becomes shortened, accompanied by hypotonia of lower leg muscles, tibial torsion, or even internal rotation of the hip (60). As these progresses, it can lead to painful degenerative changes in the joints, followed by even more walking difficulties.

The treatment of varus foot deformities can be both conservative and operative. Non-operative treatment is indicated in mild deformities that are flexible. It is also used as an adjunct to operative treatment to ensure long lasting effects of surgical correction. Most commonly used conservative treatment options include Botulinum toxin injections that target tibial muscles, physical therapy, foot and leg orthosis, special shoes, and their combinations which usually give the best results (32).

1.8. Surgical treatment of varus foot deformity in patients with CP

Surgical treatment is required in ambulatory patients with correctable deformity with marked impairment in stance and swing phases of the gait. Surgery is also recommended in patients with fixed, skeletal deformities of the joint, as well as in patients where non-operative techniques did not result in improvements (61).

Varus foot deformities can be treated with soft-tissue and osseous techniques. Correctable deformities are treated with soft-tissue surgical techniques, while the rigid deformities require both surgical techniques.

Varus foot deformity surgical procedures can be grouped based on the predominant location of deformity: forefoot, midfoot and hindfoot varus, and the affection of whole foot by this deformity (61).

1.8.1. Hindfoot varus and SPOTT procedure

Hindfoot varus is characterized by the varus position of the calcaneus which is adducted and rotated under the talus. The underlying cause in CP is the spasticity of tibialis posterior. The surgical treatments include:

- 1. Elongation of tibialis posterior. This procedure is achieved by three techniques:
 - a. Lengthening of the tibialis posterior tendon (61)
 - b. Tibialis posterior tenotomy at the tendon-muscle junction (62)
 - c. Partial tenotomy of tibialis posterior (63)
- 2. Partial transposition (,,split transfer ") of tibialis posterior tendon (SPOTT) on the peroneus brevis tendon (64,65)

The SPOTT procedure was initially described Kaufer at al. (64), and further developed by Green et al. (65) and Kling et al. (66). The purpose of this intervention is to transfer the split tendon of the tibialis posterior to the tendon of the short peroneal muscle. Initially, Kaufer et al. reported the successful postoperative outcome in 29 of 90 feet (96%) (64). Green et al. (65) performed this intervention in 16 children with equinovarus deformity due to spastic CP. They achieved correction of the varus foot deformity in all treated patients, without the recurrence of the deformity in the

minimum two years follow-up. Another group of authors reported excellent postoperative results in 35 of 37 treated children with equinovarus deformity and spastic CP (66).

Retrospective analysis of the SPOTT technique success in 38 feet with spastic varus deformity showed excellent results in 20 feet (according to Kling's clinical criteria), good in 14 feet, and poor outcome in 4 feet (67). Another group of authors (68) also evaluated the SPOTT technique's success in their cohort of 25 patients (34 operated feet). According to Green's criteria, very good postoperative results were obtained in 67.6%, good results in 23.6% of cases, and poor outcome in 8.8% of cases.

Modification of SPOTT accompanied by the Achilles tendon lengthening was reported by Mulier et al. (69). This modified technique was performed in 17 children (total of 21 procedures), resulting in excellent/good result in 19 cases (90.5%), with two poor results after mean follow-up of 29 months. Another modification of SPOTT was described by Medina et al. (70). This simplified technique by the attaching the split posterior tibial tendon more proximally to the peroneus brevis than previously described. This intervention was performed in 13 patients with spastic equinovarus deformity. After the mean follow-up pf 21 months, excellent/good results were reported in eleven patients, two patients had fair results, with no poor results of complications after this intervention.

1.8.2. Midfoot/forefoot varus and SPLATT procedure

Midfoot and forefoot varus is caused by the spasticity of anterior tibialis muscle. This deformity is treated with the split anterior tibial tendon transfer (SPLATT) to the cuboidal bone. This procedure was first described by Hoffer et al. (71). Surgical intervention consists out of transposition of the laterally split tendon of the tibialis anterior to the cuboid bone, which results in the corrected supination and neutralisation of the midfoot varus deformity. The same author (72) reported 10-year follow-up results of the SPLATT procedure performed on 27 feet. All included patients had equinovarus deformities of the hind part of the foot. In 6 feet only SPLATT procedure was performed, while 13 patients required additional Achilles tendon lengthening due to fixed deformities. Long-term follow-up proved that the SPLATT procedure was effective and safe in patient with CP. Vlachou et al. (67) performed SPLATT intervention on 11 feet with forefoot and midfoot deformity due to spastic CP. According to Hoffer's clinical criteria, excellent postoperative results were achieved in 8 cases, satisfactory in 3 feet, with no procedure failure noted.

In another study, SPLATT procedure was performed in 44 children (68 feet) with equinovarus foot deformity due CP (73). Long-term follow-up showed excellent results in 48 feet (70%), good results in 10 cases (15%), with the poor outcome in 10 cases (15%).

Barnes et al. (74) used the combination of SPLATT with intramuscular lengthening of the posterior tibial tendon. After the median 6.2 years of follow-up, postoperative results were excellent in 14 feet, good in 4 cases, and poor in 4 cases.

1.8.3. Combination of SPLATT and SPOTT technique

Varus deformity affecting the whole foot is induced by the spasticity of tibialis posterior and tibialis anterior. In such cases, surgical technique includes procedures performed on both affected muscles (67). Vlachou et al. (67) combined SPLATT and SPOTT technique in three cases (feet) of the spastic varus foot deformity. Simultaneous procedures gave one excellent and two satisfactory results.

Osseous surgical treatment can be used in concordance with soft-tissue operating techniques. These procedures are indicated in rigid varus foot deformity, and in cases of failure or inadequately achieved deformity correction with soft-tissue surgical techniques. Hindfoot varus deformity can be treated with wedge calcaneal osteotomy (75). In cases of midfoot varus deformity, a cuboid bone correction is indicated, with a lateral closing wedge osteotomy (76). Triple arthrodesis is indicated when the whole foot is in varus position, with the wedge-shape resection area (77).

Several clinical criteria can be used to evaluate the success of these surgical interventions. For the success of surgical correction of hindfoot varus, Kling's clinical criteria (66) or Chang's clinical criteria (59) can be used. For the evaluation of the surgical interventions of the midfoot/forefoot varus deformity, Hoffer's clinical criteria can be used (70).

2. AIMS

The aims of this thesis were to determine the results of split tibial tendon transfers, as well as to determine whether surgical outcome depends on other parameters aside from surgical technique. Research goals include:

- 1. Analysis of preoperative and follow up patient history and clinical examinations;
- 2. Determination of preoperative gross motor function using The expanded and revised Gross Motor Function Classification System scale (GMFCS)
- 3. Analysis of postoperative weight-bearing radiographs of foot and ankle (varus, valgus and equinus angle measurements);
- 4. Determination of postoperative fixed skeletal contracture incidence;
- 5. Evaluation of surgical outcome:
 - a. Assessed by Hoffer's clinical criteria for surgical procedures involving anterior tibialis,
 - b. and Kling's clinical criteria for surgical procedure involving posterior tibialis
- 6. Analysis of preoperative and postoperative functional mobility by using the Functional Mobility Scale (FMS).
- 7. To evaluate the association between surgical outcome and CP form (type of neurological deficit), gender, age in time of surgery, GMFCS, comorbidities, as well as other surgical procedures performed on the foot and ankle.

3. METHODS

The study was designed as a retrospective cohort study. The analysed data were obtained from the medical archives of the Institute for Orthopaedic Surgery "Banjica". The sample was selected from a group of patients with CP who have undergone spastic foot surgery performed by a single surgeon between 1986 and 2014 (153 patients, 178 feet). The process of patient selection upon defined inclusion and exclusion criteria was based on data provided in medical records of each patient.

3.1. Study population

Study population included patients with spastic CP treated at the Institute for Orthopaedic Surgery "Banjica" from 1986–2014.

Inclusion criteria for this study were:

- 1. Spastic form of CP;
- 2. Varus foot deformity during stance and swing phase of gait;
- 3. Performed split tendon transfer surgery of anterior tibialis and/or posterior tibialis;
- 4. Patients ambulatory or potentially ambulatory prior to surgical intervention;
- 5. Follow-up of at least three years.

Exclusion criteria for this study were:

- 1. Spasticity caused by factors other than CP;
- 2. Presence of fixed, skeletal contractures prior to surgery;
- 3. Incomplete medical records (absence of detailed clinical examination and patient history preoperatively and on follow-up check-ups, absence of neurological status and inadequate radiographs).

3.1. Patients' Characteristics

Characteristics of included patients were collected anamnestically, by clinical examination and by reviewing patient's medical records. Collected data included:

- Demographic data: birth date, gender;
- Clinical characteristics: CP form (type of neurological deficit: spastic hemiparesis, spastic paraparesis, spastic triparesis, and spastic quadriparesis), affected side of the body (right or left), ambulation status (walking ability, gait pattern, distance, need for walking aids and assistance, stair climbing, use of railing), presence of comorbidities, foot surface contact during gait, age when the surgery was performed, type and severity of deformity, presence of fixed, skeletal deformities and callosities, type of footwear, need for orthotics, patient/parent satisfaction with the procedure outcome, other surgical procedures performed on the foot and ankle, and revision surgery performed;
- Preoperative GMFCS;
- Postoperative change in FMS;
- Postoperative weight-bearing radiographs;
- Procedural success

3.2. Surgical treatment

Surgical treatment consisted of split tendon transfer of anterior (SPLATT) and posterior (SPOTT) tibial muscles, separately or in combination, as well of the modified SPOTT technique, developed in our institute by the team led by prof. dr Goran Čobeljić (32). SPOTT and SPLATT procedures include longitudinal tendon splitting, transfer, and attachment of the lateral part of the split tendon to the bony insertion on the outer part of the foot (64,71,72,78). The purpose of split tendon transfers lies in the change of muscle-tendon-bone complex, in order to gain balance between foot invertors and evertors, while maintaining foot stability and flexibility (67).

The operated feet were classified in to four groups according to the type of procedure performed. First group included feet surgically treated with the SPLATT procedure. Second group consisted of feet surgically treated with the SPOTT procedure. The third group included feet that underwent both procedures simultaneously (SPLATT + SPOTT). Fourth group consisted of feet surgically treated with our modification of the SPOTT procedure.

3.2.1. SPLATT

SPLATT procedure was first described by Hoffer et al. (71). Aim of this intervention is to correct the forefoot and midfoot varus caused by a spastic anterior tibialis muscle.

The principle behind the SPLATT is the movement of the lateral half of the anterior tibial tendon to the cuboid, neutralizing the varus pull of the anterior tibial muscle. The patient is in a supine position on the operating table. Two incisions are used. The first incision is made dorsomedial over the medial cuneiform and base of the first metatarsal, continuing proximally over the anterolateral aspect of the ankle, up to the distal third of the tibia (Figure 3).



Figure 3. The first incision and the exposure of tibialis anterior tendon during SPLATT.

After the anterior tibial tendon is identified, its sheath is opened longitudinally from the musculotendinous junction proximally, to just proximal of its distal insertion in the first metatarsal bone (Figures 4 and 5).



Figure 4. The longitudinal splitting of the anterior tibial tendon into two halves: medial and lateral.



Figure 5. The lateral half of the tendon is detached near its distal insertion, the sutured and tagged

The lateral half of the tendon is detached near its distal insertion and provisional suture was placed at the tendon tip (absorbable, synthetic suture size 0 or 1). A second incision is made in a longitudinal direction over the dorsum of the cuboid (Figures 6 and 7). The tagged, lateral half of the anterior tibial tendon is passed subcutaneously into the second incision (Figure 8). A hole is then drilled into the cuboid (Figure 9). Care is taken to preserve the root of the bone. The lateral slip of the tendon is introduced into the hole and fixed with an absorbable anchor with the ankle in slight dorsiflexion and the hind part of the foot in eversion (Figures 10 and 11). Postoperative management required short leg walking cast. The cast is removed after 6 weeks and replaced by the orthosis for 6 months (72).



Figure 6. A second incision in a longitudinal direction over the dorsum of the cuboid.



Figure 7. The long haemostatic forceps is then passed from a second incision to the first incision.



Figure 8. Lateral half of the anterior tibial tendon is passed subcutaneously into the second incision.

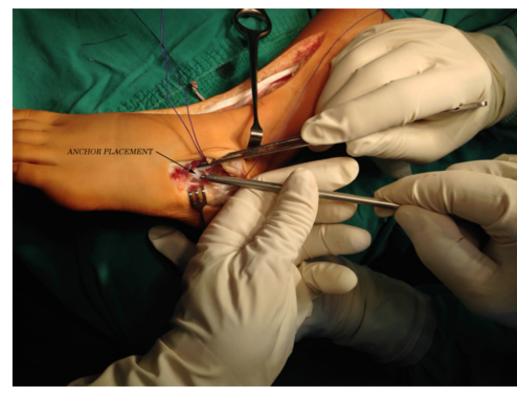


Figure 9. Small hole drilled into the cuboid.



Figure 10. The lateral slip of the tendon is introduced into the hole and fixed with an absorbable anchor suture.



Figure 11. The foot is corrected with an ankle set in slight dorsiflexion and the hind part of the foot in slight eversion.

3.2.2. SPOTT

SPOTT technique was first described by Kaufer at al. (64), and further developed by Green et al. (65) and Kling et al. (66). The procedure produces a neutral hindfoot alignment while maintaining adequate plantar flexion.

Patient is placed in a prone or supine position. A curved incision is made along the anterior edge of triceps surae muscle, around 10 cm above the medial malleolus, going behind and below it, and continuing up to the navicular tuberosity (Figure 12).



Figure 12. First incision and exposure of the posterior tibial muscle tendon. The tendon sheath is opened longitudinally throughout the length of the exposure up to the level of the medial malleolus.

A sharp dissection is carried through the subcutaneous layer, cutting through the superficial crural fascia, and exposing the posterior tibialis tendon. The tendon sheath is opened longitudinally throughout the length of the exposure with the exception of the part bellow the medial malleolus, being careful not to damage the deltoid ligament. The exposed tendon is then split longitudinally into its anterior and posterior halves (Figure 13). The cut is made from the inferior margin of the muscle belly proximally, and close to the navicular tuberosity distally (Figure 14). The posterior half is detached distally, and then introduced proximally, going underneath the deltoid ligament. Flexor tendons and neurovascular bundle are carefully mobilized from the posterior aspect of the tibia, creating an opening foot the transfer of the detached part of the posterior tibialis tendon to the lateral side of the foot (Figure 15).



Figure 13. Exposed tendon of the posterior tibialis muscle is split longitudinally into its anterior and posterior halves.



Figure 14. The cut is made from the inferior margin of the muscle belly proximally and close to the navicular tuberosity distally.

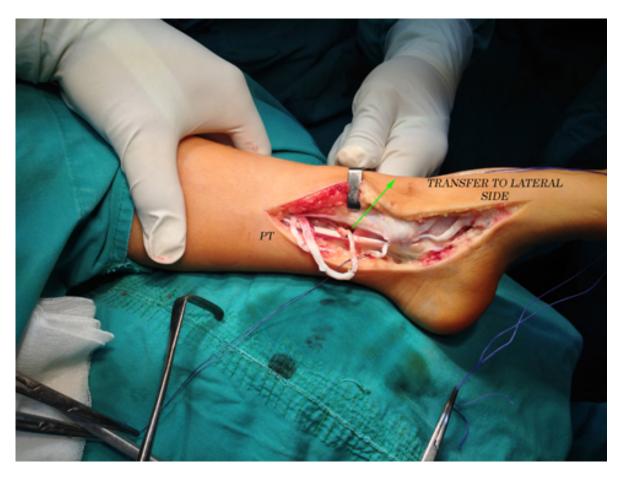


Figure 15. Flexor tendons and neurovascular bundle mobilized from the posterior aspect of the tibia, creating an opening for the transfer of the detached part of the posterior tibialis tendon to the lateral side of the foot.

The second incision is made on the outer part of the foot from the base of the fifth metatarsal to the tip of the lateral malleolus, exposing the distal part of the peroneus brevis tendon. A small slit is then made in the tendon, just above its distal insertion. The split posterior tibialis tendon half is then transferred laterally, behind the tibia, moved through the slit in the peroneus brevis tendon and then sutured to it under controlled tension with the foot in a slightly overcorrected position (Figures 16 and 17).

The final position and tensions are checked. If there is a marked equinus component in the deformity, elongation of the Achilles tendon can then be done using the proximal part of the medial incision. The wounds are then closed in anatomical layers. Postoperatively, a long-leg cast is applied with the foot in slight valgus position. After eight weeks, a short leg walking cast is applied for 8 more weeks. The cast is then removed and rehabilitation protocol is initiated.



Figure 16. The split tibialis posterior tendon half transfer into the second incision, laterally, behind the tibia.



Figure 17. The tendon is moved through the slit in the peroneus brevis tendon and then sutured to it under controlled tension with the foot in a slightly overcorrected position.

3.2.3. Modified SPOTT technique (procedure)

Modified SPOTT procedure was developed by the team led by an experienced orthopedic surgeon from our institute, prof. dr Goran Čobeljić (32). This intervention was introduced in the Institute for Orthopaedic Surgery "Banjica" in 1986, and since then has been successfully performed in patients with spastic foot deformity due to CP. It was first used in cases with severe anterior tibialis tendon spasm, and it showed good initial results in other cases as well.

For the modified SPOTT procedure, tibialis posterior tendon was exposed using an L-shaped incision starting 10 cm proximally from the medial malleolus and continuing along the front edge of triceps surae muscle, curving along the ankle, and ending at the level of navicular bone tuberosity. The *tibialis posterior* tendon sheet was opened longitudinally from the top of the wound, down to the tip of the medial malleolus where it was left intact with part of the deltoid ligament, approximately 1 cm from its insertion. The tendon was split longitudinally in two equal halves (anterior and posterior) from the origin of the muscle belly proximally to its insertion in the navicular, distally. The posterior half was detached from the navicular insertion, and provisional suture was placed at the tendon tip (absorbable, synthetic suture size 0 or 1). It was then pulled through the remaining part of the deltoid ligament into the proximal aspect of the wound. The anterior half of the tibialis posterior tendon was incised in a Z-shape fashion 5 to 7 cm in length (Figure 18).

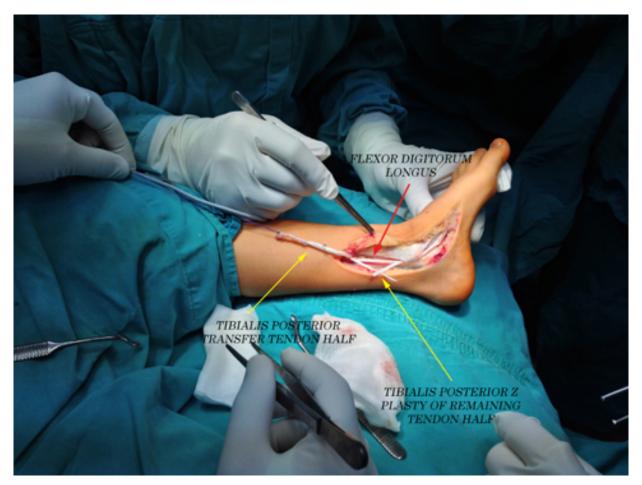


Figure 18. The anterior half of the tibialis posterior tendon was incised in a Z-shape fashion 5 to 7 cm in length. The posterior half of the tendon is sutured and tagged. Flexor digitorum tendon is identified and protected.

The second incision was made 1 cm below the tip of the lateral malleolus and continued distally, along the dorso-lateral side of the foot, ending at the base of the fifth metatarsal. Peroneus brevis tendon insertion and cuboid bone was exposed. From the proximal part of the medial incision, passing below and behind the medial malleolus, posterior half of tibialis posterior tendon was pulled toward the peroneus brevis tendon using a long, blunt forceps. Rear part of the foot was held in a slight valgus position after which the transposed tendon was pulled through a previously made longitudinal split in the peroneus brevis tendon and sutured under tension using three or four absorbable sutures. Finally, the remaining part of tibialis posterior tendon was elongated with Z-plasty and sutured with the foot in a corrected position (Figure 19 and 20).



Figure 19. After the tibialis posterior tendon transfer is completed, parts of the remaining tendon are sutured in slight tension.



Figure 20. Completed Z-plasty with the foot in a corrected position.

All wounds were closed in anatomical layers. For all patients, a short walking cast was applied for six weeks after surgery. Later, the cast was removed and rehabilitation protocol was initiated (32,78).

3.3. GMFCS – Expanded & Revised

Gross motor function of included patients was assessed by using the Gross Motor Function Classification System. It was first introduced in 1997 to describe and categorize children and youth with CP (8). This classification system was expanded in 2007 (9) categorizing children and youth up to 18 years old into five levels, depending on their present abilities and limitations. Children are grouped according to their age (0 - 2 years, 2 - 4 years, 4 - 6 years, 6 - 12 years, and 12 - 18 years). General features of this five-level classification system refer to:

- Level I walking without limitations
- Level II walking with limitations
- Level III walking with the use of hand-held mobility device
- Level IV present self-mobility with limitations, when children may use powered mobility
- Level V transport in a manual wheelchair.

For each classification level, separate abilities and limitations are described thoroughly for each age group (9).

In all included patients GMFCS level was assessed preoperatively.

3.4. Functional mobility scale

Functional mobility in included patients was assessed by using Functional Mobility Scale (FMS) (12-14). FMS is used to classify functional mobility in children, and is helpful in following change over

time, especially after orthopaedic intervention in children. This scale rates walking ability of a child at three different distances: 5 meters, 50 meters and 500 meters. These distances represent the mobility at home, at school, and in the community setting, respectively. Functional mobility is rated by numbers from 1 to 6 that best represent child's current ability or need for assistance.

- Rating 1 Uses wheelchair; may stand for transfer and may do some stepping supported by another person or a walker/frame.
- Rating 2 Uses a walker or frame, without help from another person.
- Rating 3 –Uses crutches, without help from another person.
- Rating 4 Uses sticks (one or two), without help from another person.
- Rating 5 Independent on level surfaces. Child does not use walking aids or help from another person, but requires a rail for stairs.
- Rating 6 Independent for all surfaces. Child does not use any walking aids or need any help from another person when walking over all surfaces, including uneven ground, curbs etc. and in a crowded environment.

FMS value was assessed preoperatively and postoperatively in all patients. Postoperative change compared to preoperative FMS values was noted and categorized as a positive, equal, or negative change.

3.5. Weight-bearing radiographs

On postoperative follow-up weight-bearing front and lateral foot radiographs, varus, valgus and equinus angles were measured using a standard goniometer (59). The degree of deformity was classified based on the varus/valgus angles of the foot. Foot alignment was considered neutral when there was less than 5° of varus or valgus angle. Slight varus deformity was considered when an angle ranged from 5° to 10° degrees of varus and slight valgus deformity when there was 5° to 10° degrees of valgus) foot deformity was considered when there was more than 10° of varus or valgus angle present (78).

3.6. Postoperative fixed skeletal contracture

The incidence of fixed skeletal contractures was determined based on last follow up patient examinations. If significant postoperative varus or valgus deformity was still present, in some patients, a fixed, skeletal contracture of the joint occurred, resulting in a permanent varus or valgus of the foot. These rigid deformities had to be treated with bony surgery, namely the triple arthrodesis procedure, and were considered as failed results regardless of other parameters.

3.7. Evaluation of surgical outcomes

Surgical outcome was evaluated using previously published clinical criteria by Hoffer et al. (71,72) in surgical procedures involving tibialis anterior, as well as by Kling' clinical criteria for surgical procedures involving tibialis posterior tendon (66).

According to Hoffer's clinical criteria, the postoperative results were considered excellent if there was no deformity postoperatively, when a patient could walk with complete foot surface contact during gait and could wear standard footwear. Results were considered satisfactory when there was postoperative varus, valgus or equinus deformity of less than 10° , incomplete foot contact during gait, and a necessity for overnight orthosis. The results were regarded as poor when postoperative foot deformity was more than 10° , with and a necessity for all day orthosis wearing (71,72).

According to Kling's criteria, results were considered excellent when there was complete foot surface contact during gait, without the development of fixed, skeletal contractures of the foot, when patient wore standard footwear with no foot callosities present, and when the patient or the parent was satisfied with the procedure outcome, without the need for foot orthosis. The results were regarded as good when there was varus, valgus or equinus deformity of less than 10°, when patient could walk in standard footwear with no foot callosities, and when the patient or the parent was satisfied with the procedure. Poor results indicated that the postoperative foot deformity was over 10°, when the fixed, skeletal contractures have been developed, with a necessity for foot orthosis, and/or patient/parent was not satisfied with the procedure outcome (66).

Using the described criteria, evaluation of surgical outcome was analysed for all groups based on last follow-up examination.

3.8. Statistical analyses

The descriptive statistics, including numbers and percentages of categorical variables, mean and standard deviation, and median and interquartile ratio for numerical data, were used to characterize the study sample. Differences between variables were tested using Pearson's chi-square test or Fisher's exact probability test, where appropriate. Association between variables was tested using Spearman's correlation. Statistical analyses were performed using SPSS for Windows, version 20. Significance level was set at 0.05.

4. RESULTS

4.1. Study population and clinical characteristics

Our final study population consisted of 108 patients (122 operated feet). SPLATT procedure was performed in 14 patients (16 feet). SPOTT procedure was performed in 62 patients (76 feet), combination of SPOTT + SPLATT intervention was performed in 16 patients (18 feet), while modified SPOTT technique was performed on 10 patients (12 feet).

Preoperative equinovarus foot deformity, more severe on the left foot is presented on Figure 21, with X-ray showing equinovarus foot deformity shown on Figure 22.



Figure 21. Preoperative equinovarus foot deformity, more severe on the left foot.



Figure 22. X-ray showing equinovarus foot deformity.

4.2. SPLATT procedure

SPLATT procedure was performed in 14 patients (16 feet). In two patients both feet were treated with SPLATT procedure. Eight patients (57.1%) were female, while 6 (42.9%) patients were male. CP distribution is presented in Figure 23. The majority of patients were presented as spastic hemiparesis (7 patients, 50%), while 4 (28.6%) had spastic diparesis, 2 (14.3%) had spastic quadriparesis and one patient (7.1%) had spastic paraparesis. Patients had no other comorbidities.

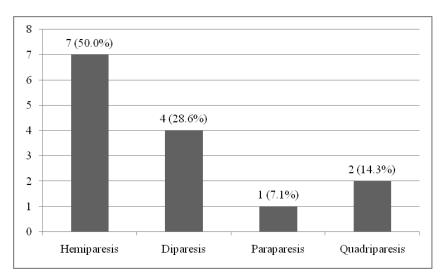


Figure 23. Distribution of CP geographical pattern of involvement in patients treated with SPLATT procedure

The affected side of the body was the right side in 11 (68.8%) feet, and the left side in 5 (31.3%) feet. Mean age at the time of the surgery was 15.07 ± 8.44 years.

4.2.1. Preoperative GMFCS

Distribution of preoperative GMFCS values is presented in Figure 24. The majority of patients had GMFCS level I (8 cases, 50.0%), while level II and level III were recorded in 5 cases (31.3%), and in 3 cases (18.8%), respectively.

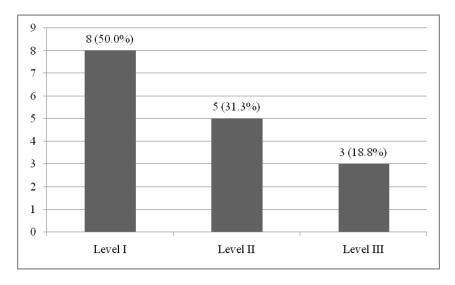


Figure 24. Distribution of preoperative GMFCS levels in patients treated with SPLATT

4.2.2. FMS

Distribution of FMS change postoperatively compared to preoperative values is presented in figure 25. In the majority of cases (13 feet, 81.2%) there was a positive change in FMS values, while 3 cases (18.8%) remained equal FMS values. There were no negative changes in FMS in patients treated with SPLATT technique.

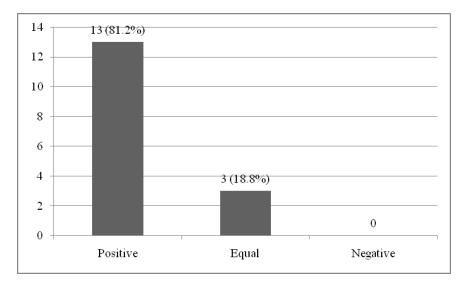


Figure 25. Distribution of FMS change postoperatively compared to preoperative values in patients treated with SPLATT

4.2.3. Postoperative weight-bearing radiographs

Presence of postoperative deformity is shown in Figure 26. In 13 feet (81.2%) there was no significant postoperative foot deformity, while in other three (18.8%) feet there was postoperative varus foot deformity. In two patients (12.5%) the deformity was mild, while one patient (6.3%) had severe varus foot deformity.

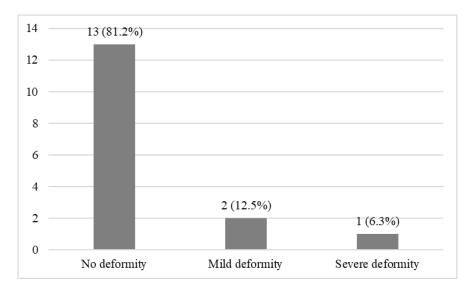


Figure 26. Distribution of postoperative foot deformities in patients treated with SPLATT

Complete foot contact with the standing surface was achieved in 13 treated feet (81.2%). Outer rim contact was achieved in 3 feet (18.8%), with no cases of foot contact only with the inner rim of the foot.

4.2.4. Postoperative fixed skeletal contracture

Postoperative fixed skeletal contracture was recorded in only one patient treated with SPLATT, who had spastic hemiparesis. In this case triple arthrodesis was performed.

4.2.5. Procedural success

Mean follow-up duration was 7.00 years (IQR 6.24 - 11.00, range 4 - 21 years). According to Hoffer's clinical criteria, excellent and good results were achieved in 11 (68.7%) and 3 (18.8%) feet. Poor postoperative result was recorded in 2 (12.5%) feet (Figure 27).

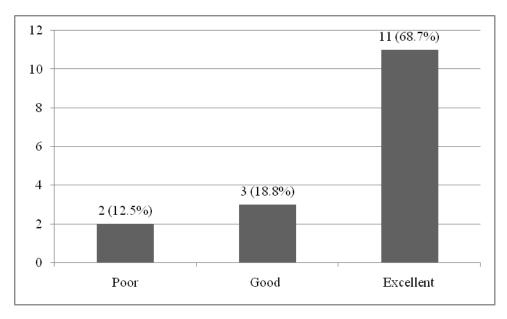


Figure 27. Distribution of postoperative success according to Hoffer's clinical criteria

Distribution of other surgical procedures performed in patients treated with SPLATT is presented in Table 1.

Table 1. Concomitant surgical procedure performed with SPLATT intervention.

Additional surgical procedure	Number of patients (%)
Hoke procedure	9 (56.3%)
Hoke procedure + elongation of flexor digitorum muscle	1 (6.3%)
Tenotomy of flexor digitorum muscle	1 (6.3%)
None	5 (31.3%)

Thirteen patients (81.2%) used standard/normal shoes after the procedure, while 3 patients (18.8%) required special shoes. The majority of patients did not require orthoses (n=13 patients, 81.2%), one patient (6.3%) was wearing orthoses during the night, while 2 patients (12.5%) required orthoses during throughout the entire day. One patient (6.3%) had callosities postoperatively. Patients were satisfied with the SPOTT outcome in 13 cases (81.2%).

Distribution of surgical outcome regarding the patients' CP form is presented in Figure 28. Good/excellent postoperative results were obtained in 6 (85.7%) cases with spastic hemiparesis, in all cases with spastic paraparesis and triparesis, and in one (50.0%) case with spastic quadriparesis.

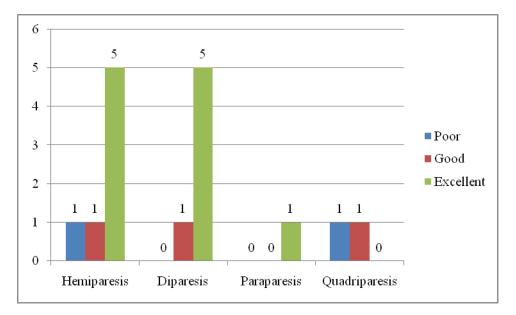
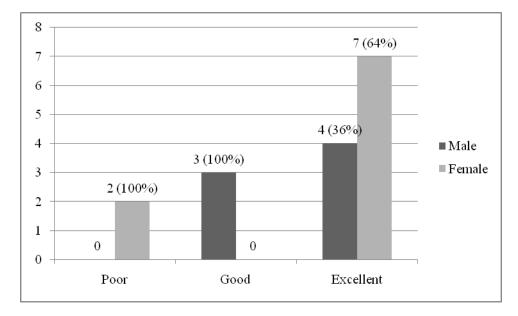


Figure 28. Surgical outcome according to Hoffer's criteria compared to CP form.

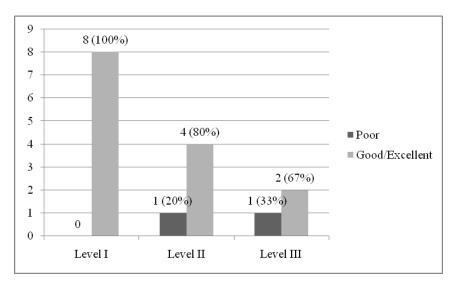


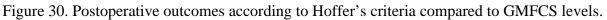
Gender distribution according to procedural outcome is presented in Figure 29.

Figure 29. Surgical outcome according to Hoffer's criteria compared to patients' gender.

There was no significant correlation between the surgical outcome according to Kling's criteria and patients' age at the time of the surgery (ρ =0.136, p=0.615).

Surgical outcome was estimated as good/excellent in all patients with Level I, and in the majority of patients with level II (80%) and level III on GMFCS (67%) (Figure 30).





4.3. SPOTT procedure

SPOTT procedure was performed in 66 patients (76 feet). In 56 patients only one foot was surgically treated, while in 10 patients both feet were treated with the SPOTT procedure. Forty patients (60.6%) were male, and 26 patients (39.4%) were female. CP type distribution is presented in Figure 31. The majority of patients had spastic diparesis (n=22 patients, 33.3%) and spastic hemiparesis (n=21 patients, 31.8%), while 12 patients (18.2%) and 8 patients (12.1%) had spastic paraparesis and spastic quadriparesis, respectively. Four patients (6%) had epilepsy, one patient (1.5%) had hydrocephalus, and one patient (1.5%) had hereditary polyneuropathy.

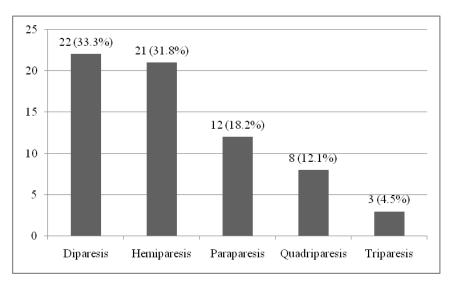


Figure 31. Distribution of CP geographical pattern of involvement in patients treated with SPOTT procedure

The affected side of the body was the right side in 39 feet (51.3%), and the left side in 37 feet (48.7%). Median age at the time of the surgery was 12 years (IQR 7 - 17 years).

4.3.1. Preoperative GMFCS

Distribution of preoperative GMFCS values is presented in Figure 32. The majority of patients had GMFCS level II (36 cases, 47.4%) and level III (34 cases, 44.7%), while level I was recorded in 3 cases (3.9%), as well as level IV in 3 cases (3.9%) level IV.

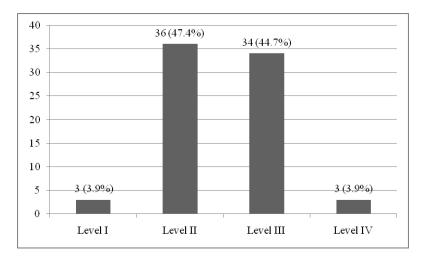


Figure 32. Distribution of preoperative GMFCS levels in patients treated with SPOTT

4.3.2. FMS

Distribution of FMS change postoperatively compared to preoperative values is presented in Figure 33. In the majority of cases (52 feet, 68.4%) there was a positive change in FMS values, while 24 cases (31.6%) remained equal in FMS values. There were no negative changes in FMS in patients treated with SPOTT technique.

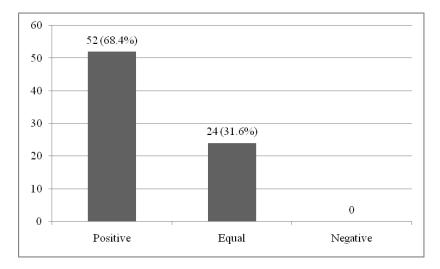


Figure 33. Distribution of FMS change postoperatively compared to preoperative values in patients treated with SPOTT

4.3.3. Postoperative weight-bearing radiographs

Presence of postoperative deformity is presented in Figure 34. In 61 feet (80.3%) there was no postoperative foot deformity. Seven cases (9.2%) had mild deformity (four feet with mild varus and three feet with mild valgus deformity) while eight feet (10.5%) had severe varus postoperative foot deformity.

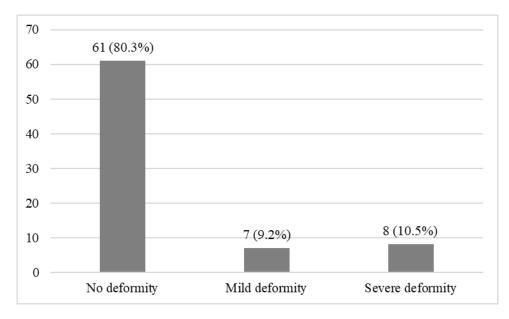


Figure 34. Distribution of postoperative foot deformities in patients treated with SPOTT

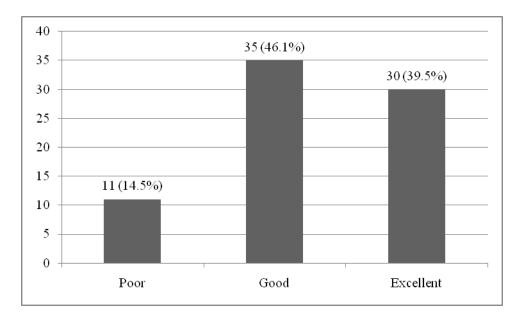
Complete foot contact with the standing surface was achieved in 46 treated feet (60.5%). Outer rim contact was achieved in 21 feet (27.6%), while in 9 cases (11.8%) there was only contact with the inner rim of the foot.

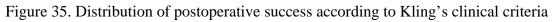
4.3.4. Postoperative fixed skeletal contracture

Postoperative fixed skeletal contracture was recorded in only one patient treated with SPOTT, who had spastic quadriparesis. In this case triple arthrodesis was performed.

4.3.5. Procedural success

Median duration of follow-up was 8 years (IQR 6 - 11, range 3 - 27 years). According to Kling's clinical criteria, excellent and good results were achieved in 30 (39.5%) and 35 (46.1%) patients, respectively. Poor postoperative results were recorded in 11 (14.5%) feet (Figure 35).





Distribution of other surgical procedures performed in patients treated with SPOTT is presented in Table 2.

Additional surgical procedure	Number of patients (%)
Achilles elongation (Z-plasty)	3 (3.9%)
Hoke procedure	55 (72.4%)
Hoke procedure + elongation of flexor digitorum muscle	2 (2.6%)
Strayer's procedure	10 (12.1%)
None	6 (7.9%)

Sixty-one patients (80.3%) used standard/normal shoes after the procedure, while 15 patients (19.7%) required special shoes. The majority of patients did not require orthoses (n=50 patients, 65.8%), 21 patients were wearing orthoses during the night, while 5 patients (6.6%) required orthoses throughout the day. Fifteen patients (19.7%) had callosities postoperatively. Patients were satisfied with the SPOTT outcome in 64 cases (84.2%).

Distribution of surgical outcome regarding patients' CP form is presented in Figure 36. Good/excellent postoperative results were obtained in 18 (85.7%) cases with spastic hemiparesis, 23 (88.5%) cases with spastic diparesis, in all cases with spastic paraparesis and triparesis (17 cases), and in 7 (58.3%) cases with spastic quadriparesis.

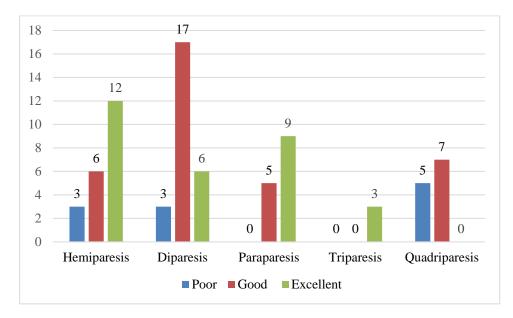


Figure 36. Surgical outcome according to Kling's criteria compared to CP geographical pattern of involvement.

There was no statistically significant difference in surgical outcome (poor/good/excellent) compared to patients' gender ($\chi 2=0.773$, p=0.514, Figure 37).

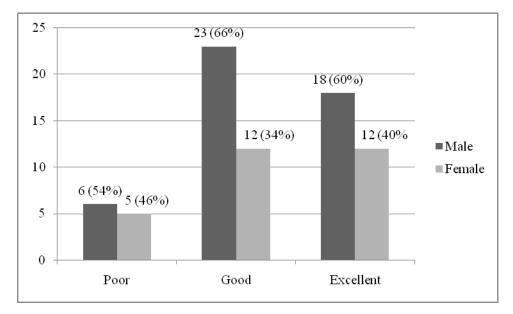


Figure 37. Surgical outcome according to Kling's criteria compared to patients' gender.

Age at the time of the operation showed no correlation with the procedural success (excellent/good vs. poor, ρ =-0.090, p=0.440).

Surgical outcome was estimated as good/excellent in the majority of patients with Level I (100%), level II (92%) and level III on GMFCS (85%). All patients with level IV on GMFCS had poor postoperative outcome according to Kling's clinical criteria (Figure 38).

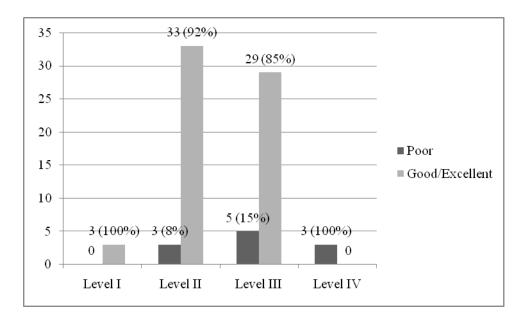


Figure 38. Postoperative outcomes according to Kling's criteria compared to on GMFCS levels.

4.4. SPOTT and SPLATT procedures

SPOTT and SPLATT procedures were performed in 18 patients (18 feet). Ten patients (55.6%) were female, and 8 patents (44.4%) were male. CP type distribution is presented in Figure 39. The majority of patients had spastic hemiparesis (n=10 patients, 55.6%), three patients had spastic diparesis and three patients had spastic diparesis (16.7%), while two patients (11.1%) had quadriparesis. Six patients (33.3%) had epilepsy, two patients (11.1%) had hydrocephalus, and one patient (5.5%) had hereditary polyneuropathy.

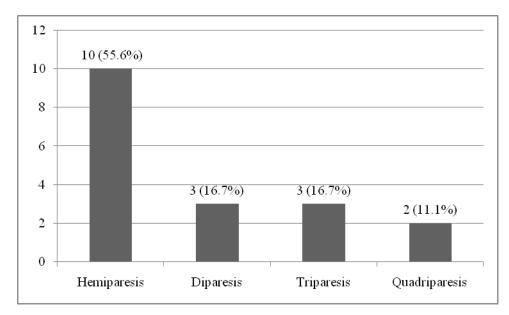


Figure 39. Distribution of CP geographical pattern of involvement in patients treated with SPOTT and SPLATT procedure

The affected side of the body was the right side in 10 patients (55.6%), and the left side in 8 patients (44.4%). Median age at the time of the surgery was 15.0 years (IQR 7.5 - 23.0 years).

4.4.1. Preoperative GMFCS

Distribution of preoperative GMFCS values is presented in Figure 40. The majority of patients had GMFCS level III (8 patients, 44.4%) and level II (6 patients, 33.3%), while level I was recorded in 2 patients (11.1%), as well as level IV in 2 patients (11.1%).

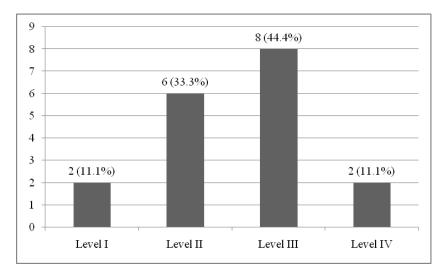


Figure 40. Distribution of preoperative GMFCS levels in patients treated with SPOTT + SPLATT technique

4.4.2. FMS

Distribution of FMS change postoperatively compared to preoperative values is presented in figure 41. In the majority of cases (n=16 patients, 88.9%) there was a positive change in FMS values, while in 2 patients (11.1%) FMS values remained equal. There were no negative changes in FMS in patients treated with SPOTT+SPLATT technique.

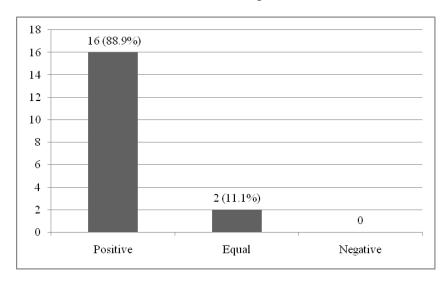


Figure 41. Distribution of FMS change postoperatively compared to preoperative values in patients treated with SPOTT and SPLATT

4.4.3. Postoperative weight-bearing radiographs

Presence of postoperative deformity is shown in Figure 42. In 11 feet (61.1%) there was no postoperative varus foot deformity, five patients (27.8%) had mild deformity (two feet had mild varus and three feet had mild valgus deformity), while two patients (11.1%) had severe postoperative varus foot deformity.

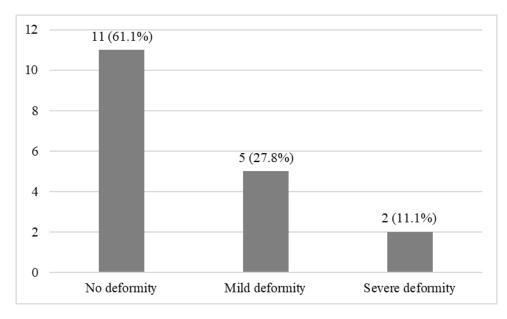


Figure 42. Distribution of postoperative foot deformities in patients treated with SPOTT and SPLATT

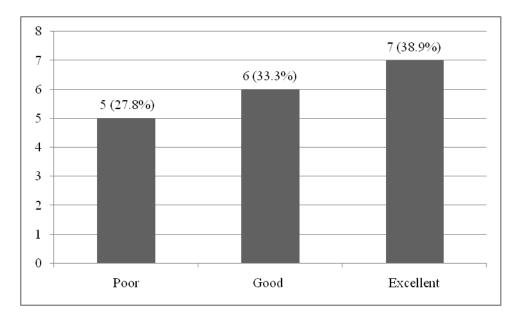
Complete foot contact with the surface while standing was achieved in 11 treated feet (61.1%). Outer rim contact was achieved in 4 feet (22.2%), while in 3 cases (16.7%) there was only contact with the inner rim of the foot.

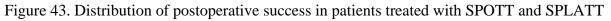
4.4.4. Postoperative fixed skeletal contracture

Postoperative fixed skeletal contracture was recorded in only one patient treated with SPOTT and SPLATT techniques, who had spastic quadriparesis. In this case triple arthrodesis was performed.

4.4.5. Procedural success

Median follow-up duration was 8.50 years (IQR 4.75 - 10.00, range 3 - 17 years). According to Kling's clinical criteria and Hoffer's clinical criteria, excellent and good results were achieved in 7 (38.9%) and 6 (33.3%) patients respectively. Poor postoperative results were recorded in 5 (27.8%) operated feet (Figure 43).





Distribution of other surgical procedures performed in patients treated with SPOTT and SPLATT is presented in Table 3.

Additional surgical procedure	Number of patients (%)
Hoke procedure	4 (22.2%)
Hoke procedure + elongation of flexor digitorum muscle	2 (11.1%)
Elongation of flexor digitorum muscle	2 (11.1%)
Strayer's procedure	3 (16.7%)
None	7 (38.9%)

Table 2 Canaami	itant annai a al mma	and was man farmened as	th CDOTT and CDI	ATT interregion
Table 3. Concom	itant surgical pro	ceaure performed w	vith SPOTT and SPL	AII intervention.

Eleven patients (61.1%) used standard/normal shoes after the procedure, while 7 patients (38.9%) required special shoes. The majority of patients did not require orthoses (13 patients, 72.2%), 2 patients (11.1%) were wearing orthoses during the day, while 3 patients (16.7%) required orthoses throughout the day. Two patients (11.1%) had callosities postoperatively. The majority of patients were satisfied with the outcome of SPOTT + SPLATT intervention (16 patients, 88.9%).

Distribution of surgical outcome regarding the patients' CP form is presented in Figure 44. Good/excellent postoperative results were obtained in all patients with spastic hemiparesis and spastic triparesis. Poor results were recorded after SPOTT + SPLATT intervention in all patients with spastic diparesis and spastic quadriparesis.

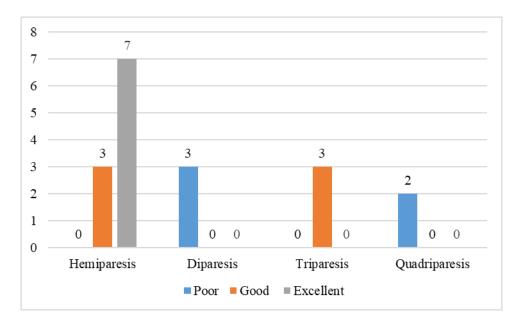
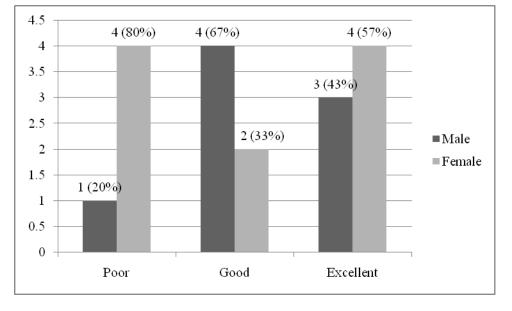


Figure 44. Surgical outcome distribution compared to CP geographical pattern of involvement.



Surgical outcome distribution compared to patients' gender is presented in Figure 45.

Figure 45. Surgical outcome according to Kling/Hoffer's criteria compared to patients' gender.

There was significant correlation between the surgical outcome according to Kling/Hoffer's criteria and patients' age at the time of the surgery (ρ =0.649, p=0.004).

Surgical outcome was estimated as good/excellent in all patients level I, level II, and level IV on GMFCS, as well as in three (37%) patients with level III (Figure 46).

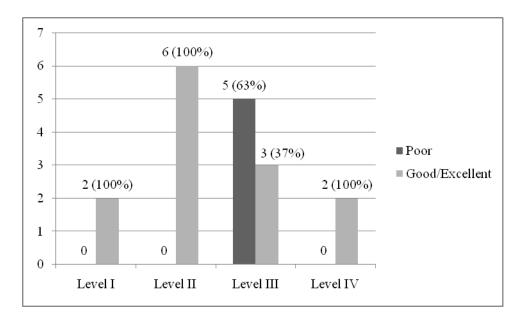


Figure 46. Postoperative outcomes according to Kling's criteria compared to on GMFCS levels.

4.5. Modified SPOTT procedure

Modified SPOTT procedures were performed in 10 patients (12 feet). Six patients (60%) were male, and 4 patients (40%) were female. CP type distribution is presented in Figure 47. Seven (70%) patients had spastic hemiparesis, two (20%) patients had spastic diparesis, while one (10%) patient had spastic paraparesis. Four patients (40%) had epilepsy.

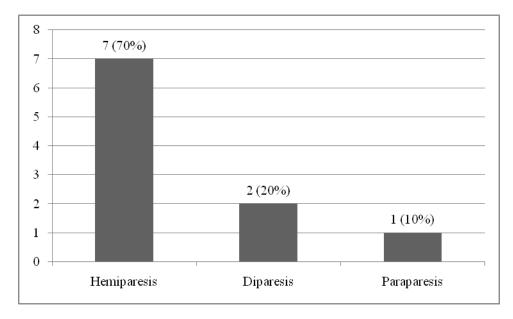


Figure 47. Distribution of CP type in patients treated with the modified SPOTT procedure

The affected side of the body was the right side in 7 cases (58.3%), and the left side in 5 feet (41.7%). Median age at the time of the surgery was 8 years (IQR 5 - 12 years).

4.5.1. Preoperative GMFCS

Distribution of preoperative GMFCS values is presented in Figure 48. There was equal distribution in the GMFCS level from I to III (4 patients per level, 33.3%).

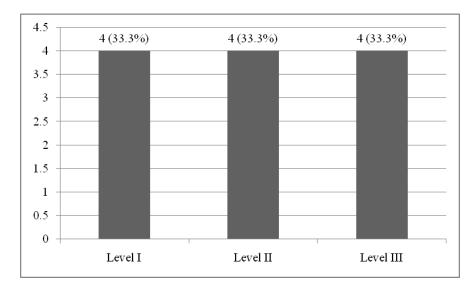


Figure 48. Distribution of preoperative GMFCS levels in patients treated with modified SPOTT technique

4.5.2. FMS

Distribution of FMS change postoperatively compared to preoperative values is presented in Figure 49. In 6 (50%) feet there was positive change in FMS values, and in 6 (50%) feet the FMS value remained equal postoperatively. There were no negative changes in FMS in patients treated with modified SPOTT technique.

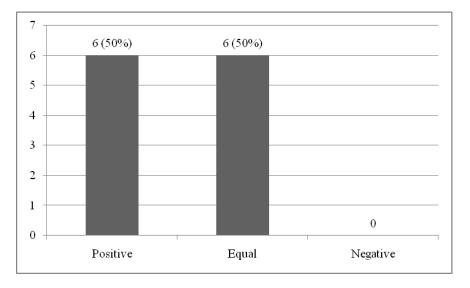


Figure 49. Distribution of FMS change postoperatively compared to preoperative values in patients treated with modified SPOTT

4.5.3. Postoperative weight-bearing radiographs

Presence of postoperative deformity is shown in Figure 50. Ten (83.3%) operated patients had no postoperative foot deformities, while other two feet (16.7%) had mild varus deformity. There were no cases of severe postoperative foot deformity.

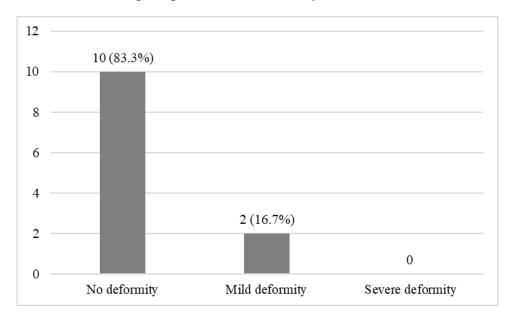


Figure 50. Distribution of postoperative foot deformities in patients treated with modified SPOTT

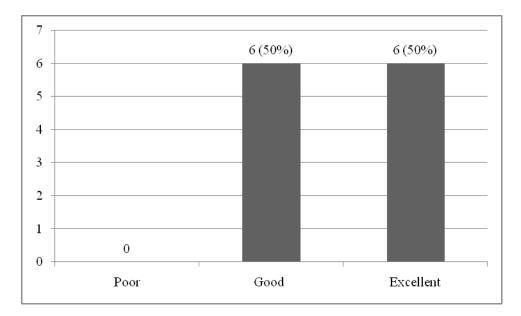
Full foot surface contact during gait was achieved in 6 (50%) feet postoperatively, while other 6 (50%) feet had outer rim foot surface contact during the gait.

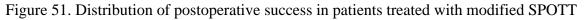
4.5.4. Postoperative fixed skeletal contracture

There were no postoperative fixed skeletal contractures in patients treated with modified SPOTT technique.

4.5.5. Procedural success

Median follow-up in this group of patients was 8.50 years (IQR 5.00 - 13.25, range 4 - 20 years). According to Kling's clinical criteria excellent results were achieved in half the patients 6 (50%), and good results were obtained in other half of the patients 6 (50%). There were no poor postoperative results (Figure 51).





Distribution of other surgical procedures performed in patients treated with SPOTT and SPLATT is presented in Table 4.

Additional surgical procedure	Number of patients (%)
Hoke procedure	1 (8.3%)
Strayer's procedure	5 (41.3%)
Achilles elongation (Z-plasty) + tenotomy of flexor digitorum	1 (8.3%)
Tenotomy of flexor digitorum	1 (8.3%)
None	4 (33.3%)

Table 4. Concomitant surgical procedure performed with modified SPOTT intervention.

All patients used standard/normal shoes after the procedure. Orthoses during the night were required in 5 (41.7%) operated feet, while other 7 (58.3%) operated feet required no orthosis. There were no callosities postoperatively. All patients were satisfied with the outcome of the modified SPOTT intervention.

Distribution of surgical outcome regarding the patients' CP form is presented in Figure 52. Good postoperative results were obtained in 4 (57%), and excellent in three (43%) feet with spastic hemiparesis form. Excellent postoperative results were achieved in all patients with spastic diparesis, while equal number of operated feet (n=2) with spastic paraparesis had postoperative good and excellent results, respectively.

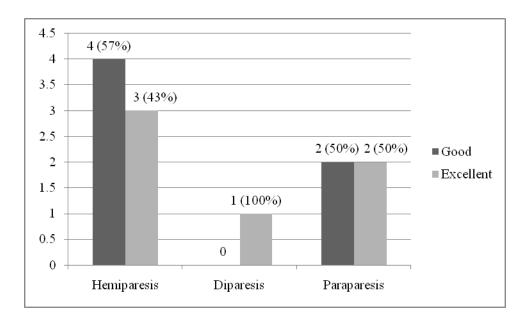


Figure 52. Surgical outcome of patients treated with modified SPOTT according to Kling's criteria compared to CP geographical pattern of involvement.

There was significant correlation between the surgical outcome according to Kling's criteria and patients' age at the time of the surgery (ρ =0.303, p=0.339).

Surgical outcome was estimated as excellent in three (75%) cases with level I, and in one (25%) case with level II on GMFCS (Figure 53). In patients with preoperative level III equal number of patients had good, and excellent postoperative outcome, respectively.

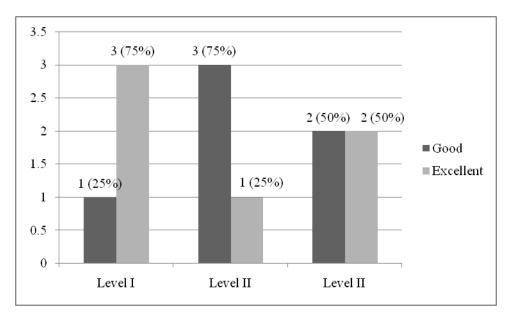


Figure 53. Postoperative outcomes according to Kling's criteria compared to on GMFCS levels.

5. DISCUSSION

This thesis analysed the procedural success of split tibial tendon transfers in patients with spastic varus foot deformity caused by CP. Surgical procedures included SPLATT, SPOTT, combination of SPLATT + SPOTT technique, and the modified SPOTT intervention. Long-term follow up showed that excellent/good postoperative results were achieved in the majority of all treated patients. Postoperative fixed skeletal contractures were recorded in only three out of 122 operated feet.

Foot deformities are frequently recorded in patients with spastic form of CP (51). Cases of mild foot deformities can be treated with orthoses and physical therapy, while severe cases often require surgical interventions. One of the most common foot deformities is pes varus, which represents a challenge for an orthopaedic surgeon. Over the past decades, numerous surgical techniques and their modifications have been developed in order to treat spastic varus foot deformity in children with CP (61-66).

Hindfoot varus in children with CP is caused by the spasticity of the tibialis posterior muscle. One of the surgical methods in reducing its spasticity is split transfer of tibialis posterior tendon on the peroneus brevis tendon. This technique was described by Kaufer et al. (64), and later on developed by Green et al. (65) and Kling et al. (66). One of the first follow-up results of the SPOTT efficacy was reported by Green et al. (65). This procedure was performed in 16 feet (16 children) with equinovarus foot deformity due to spastic CP. Patients were followed-up for at least two years, and the authors reported no recurrent varus deformities, with no equinus present during gait. After two years, Kling et al. (66) reported results from an average of 8 years of follow-up of 37 SPOTT interventions in 31 children. Excellent/good results were recorded in 34 out of 37 (91.9%) patients. Later studies have confirmed the effectiveness of SPOTT in children with CP, which was combined with additional surgical interventions in order to obtain the optimal postoperative results (68,80).

Performing SPOTT procedure along with Achilles lengthening or gastrocnemius recession resulted in excellent/good postoperative results in 40 of 45 treated feet with spastic equino-varus deformity after a median follow-up of 3.5 years (80). Similar study analysed 25 patients with CP and varus foot deformity (34 feet) that were treated with SPOTT intervention (68). Patients were followed for a median time of 5.5 years. According to Green's criteria, very good results were achieved in 23 (67.6%) patients, 8 (23.6%) patients had good results, while remaining 3 (8.8%) patients had poor results. Similar results were recorded in our cohort of 76 patients only treated with SPOTT procedure. Excellent and good postoperative result was observed in 85.6% patients. The most common additional procedures in our cohort were the Hoke procedure (72.5%), followed by Strayer's procedure (12.1%).

The highest procedural success (excellent/good) in our study cohort was seen in patients with spastic diparesis (88.5%) and spastic hemiparesis (85.7%). The highest procedural failure (poor postoperative results) were recorded in patients with spastic quadriplegia (5 of 12 patients, 41.7%). Remaining 7 quadriplegic patients had good postoperative results, with no patients with excellent outcome. Similar results were observed in a report of Vlachou et al. (67), where four patients with spastic quadriplegia were included. After performing SPOTT procedure, there was no excellent postoperative results, with only one (25%) good result, and three (75%) poor postoperative outcomes. Good and excellent results were recorded in all patients with hemiplegia (n=22) and in all patients with diplegia (n=12), comparable to our study group (excellent/good results in 85.7% patients with

hemiparesis, and in 88.4% patients with diparesis). Also, Vlachou et al. (67) performed concomitant Achilles cord lengthening in 18 (47.3%) feet due to equinus hindfoot deformity. Additionally, these authors performed plantar soft tissue releases in 15 feet, Jones procedure in 5, and long extensor tendons transfer to the metatarsals in 2 feet, with transcutaneous flexor tenotomies in 23 feet.

In the analysis of 108 children with CP and equinovarus foot deformity, Chang et al. (59) evaluated the procedural success of different surgical techniques on posterior tibialis tendon. These procedures included split tendon transfer, but also intramuscular lengthening and Z-lengthening. Two possible predictors of successful procedural outcome emerged: age at operation, and preoperative status of ambulation. In our cohort there was no correlation between patients' age at the time of surgery with procedural outcome. Preoperative gross motor function of our patients was assessed by using GMFCS, and patients with better gross motor function (GMFCS I, II and III) more frequently had favourable outcome (100%, 92%, and 85%, respectively). All patients with preoperative GMFCS – level IV had poor postoperative result.

Traditionally, SPOTT procedure is performed with the transposition of the split tendon of tibialis posterior on the peroneus brevis muscle (64,65). Saji et al. (81) have tested the modified SPOTT procedure with the transfer of the split tendon of tibialis posterior to the dorsum of the foot, but through the interosseous membrane. This intervention was performed on 18 children (23 feet). After long-term follow-up of median 8.4 years, postprocedural results were excellent in 14 (60.9%) feet, good in 8 (34.8%) feet, and poor in 1 (4.3%) foot.

Importance of laboratory gait analysis was underlined by several authors (82-84). Moran et al. (82) have explored the dynamic of three different attachment sites of tibialis posterior split tendon. They have measured tibialis posterior muscle moment arms in the original SPOTT procedure with the attachment of split tendon to distal peroneus brevis, in the slightly modified SPOTT where the attachment site was proximal on peroneus brevis tendon, and in the third variation where the split tendon passed through the interosseous membrane and attached to distal peroneus brevis tendon. All three interventions eliminated the extensive hindfoot inversion present with equinovarus, but the interosseous routing showed the weakest reduction. Moment arms of the tibialis posterior split tendon where similar if their attachment location was proximal or distal on the peroneus brevis tendon insertion.

Preoperative dynamic electromyography (EMG) can be useful in choosing the most optimal operative strategy for individual patients, as well as to predict successful postoperative outcome. Varus foot deformity is caused by the disbalance between tibialis muscles (tibialis anterior and tibialis posterior), and peroneal muscles (peroneus longus and peroneus brevis). Pes varus in children with CP has traditionally been associated with the dysfunction of tibialis posterior. However, by using dynamic EMG, Michlitsch et al. (83) have noted higher prevalence of anterior tibialis dysfunction than expected. They have analysed 78 patients (88 feet) with CP and varus foot deformity. Anterior tibialis was the contributor to pes varus in 30 (34.1%) feet, anterior tibialis was the only contributor to pes varus in 27 (30.7%) feet, while posterior tibialis was the only contributor to pes varus in 29 (33%) feet, and another muscle contributed to varus foot deformity in remaining two feet. Therefore, the anterior tibialis (alone or with tibialis posterior) contributed to the development of varus foot deformity in 64.1% of analysed feet.

Dynamic EMG can be of assistance in predicting the success of split tibial tendon transfer in patients with varus foot deformity. In a case series of 25 children with spastic hemiplegic CP and pes varus, surgeons performed SPOTT procedure to correct foot deformity (84). The aim as to predict postoperative SPOTT success by analysing preoperative dynamic EMG status. Postoperative failure was recorded in three patients, with additional five patients having mild residual varus. Unrecognized anterior tibial muscle overactivity and advanced age at operation were associated with under correction of varus foot deformity. On the contrary, our results showed that SPOTT procedure failure was not associated with the age of the children at surgery. Additionally, laboratory gait analysis was not available at our institution, which represented one of the limitations of our study. However, the spasticity of tibialis posterior was clinically tested by our experienced orthopaedic surgeon from out institution.

Group of authors analysed preoperative gait by CODA-3 motion analyser (85). Preoperative analysis was performed in 16 patients with CP and equinovarus foot deformity who were candidates for SPOTT procedure. In the majority of patients (13 of 16 patients), SPOTT intervention was combined with Achilles tendon lengthening. One year after the initial operation gait analysis was repeated, and confirmed the good postoperative outcome, pointing to the significance of preoperative gait assessment. Another group or researchers used surface and implanted electrodes into the muscle for the measurement of muscle activity during gait (86). Surface electrodes were placed on tibialis anterior, triceps, peroneus brevis and peroneus longus, while other electrodes were implanted into tibialis posterior. This study confirmed the impact of laboratory gait analysis on choosing the appropriate surgical tendon transfer strategy.

SPLATT procedure is indicated for the treatment of midfoot and forefoot varus due to the spasticity of anterior tibialis muscle. Initial long-term follow-up results of SPLATT intervention were reported in 1985 by Hoffer et al. (72). Their cohort included 21 patients with CP. Overall 27 feet with spastic equinovarus deformity were treated with SPLATT technique. After 10-years follow-up 19 of 21 treated patients were community ambulators, with improved gait and no need for orthoses. Only one recurrence of deformity was recorded during the follow-up period. Report in which 20 patients with CP treated with 22 SPLATT interventions confirmed the effectiveness of this procedure (74). After an average follow-up of 6.2 years excellent results were achieved in 14 (63.6%) cases, good postoperative results were recorded in 4 (18.2%) feet, while poor result was obtained in 4 (18.2%) feet. Albeit, SPLATT was frequently combined with intramuscular lengthening of the posterior tibial tendon, with/without concomitant lengthening of the Achilles tendon. In our cohort SPLATT procedure was performed in 14 patients (16 feet). Postoperative success was 87.5%. After long-term follow-up of an average 7 years, excellent/good results were noted in 11 (68.7%) and 3 (18.8%) feet, similar to previously published studies (67,73,74,87). Vlachou et al. (67) have analysed three groups of patients with CP and spastic foot deformities with different surgical techniques applied. One group constituted of 11 patients (11 feet) with pes varus caused by anterior tibialis overactivity. Very good results were recorded in 8 (72.7%) feet, and three (27.3%) feet had satisfactory result. CP distribution in this group of patients was following: the majority (10 of 11) of patients had hemiplegia, while one patient had quadriplegia. Satisfactory outcome was noted in a quadriplegic patient. CP distribution in our cohort was different and more diverse, which can explain partially different result from Vlachou et al. (67). Majority of our SPLATT-treated patients had hemiparesis (n=7, 50%), followed by diparesis (n=4, 28%), quadriparesis (n=2, 14.3%) and paraparesis (n=1, 7.1%). Vlachou et al. (67) have also frequently performed supplementary operations due to existing mild cavus. The most common interventions were plantar soft tissue releases, transcutaneous flexor tenotomies and Jones procedure. Concomitant procedures in our SPLATT-cohort were Hoke procedure (performed in half of the patients), while only one patient had Hoke procedure along with elongation of flexor digitorum muscle, and one patient had tenotomy of flexor digitorum muscle.

Group of authors explored predictive role of preoperative GMFSC level on postoperative outcome in SPLATT-treated patients (73). They analysed 45 children with spastic CP and outcomes of SPLATT procedure in 48 equinovarus feet. Favourable outcomes (excellent and good) were registered in 58 (85%) feet. Improvement in GMFCS level was recorded in 34 (75.5%) children, similar to result in our group of patients (GMFCS level improved postoperatively in 13 patients – 81.2%, and remained equal in 3 patients – 18.8%). Although the authors reported no difference in 10-year SPLATT success between patients with GMFCS levels I-II vs. levels III-IV, they indicated that the GMFCS level III-IV might be the predictor of unfavourable outcome. In our SPLATT cohort the majority of patients had GMFCS level I and II (n=13 patients, 81.3%), with three (18.8%) patients having GMFCS level III. Similarly to the results of Limpaphayom et al. (73) all patients with level I and 80% patients with level II on GMFCS had excellent/good postoperative results. Overall results are not comparable since our included patients did not have preoperative GMFCS level IV.

Spasticity of both tibialis posterior and tibialis anterior can present as whole foot varus deformity. In such cases performing both SPOTT and SPLATT techniques is indicated. Vlachou et al. (67) have presented three such patients with CP; two presenting with hemiplegia, and one with diplegia. Postoperative results were excellent in two patients, and satisfactory/good in one patient with diplegia. Our cohort was larger and included 18 patients with CP (18 operated feet). Our results differed from the results of Vlachou et al. (67); excellent/good postoperative outcome was noted in 13 (72.2%) feet, while in five (27.8%) operated feet postoperative result was poor. Explanation for the observed differences may lie different sample size and CP distribution. The majority of our patients (n=10, 55.5%) had hemiparesis, followed by diparesis in 3 (16.7%) patients, triparesis in 3 (16.7%) and quadriparesis in 2 (11.1%) patients.

Treating patients with equinovarus foot deformity due to CP can be quite a challenge for the surgeon. Success of the SPOTT procedure largely depends on achieving adequate tension between the split tendon parts, as well as obtaining the perfect foot position in order to gain long-lasting correction.

Since the previously published trials often presented patients with additional foot deformation along with equinovarus, the reported failure rate was quite variable and even above 50% in cases associated with internal tibial torsion (88). Several authors have tried to modify the SPOTT procedure with an alternative attachment site, or the different route of the split posterior tibialis tendon, in order to get the optimal post-operative results (69,70,81). Medina et al. (70) have performed SPOTT in simplified manner; they have attached the split tendon to the proximal peroneus brevis, avoiding the lateral dissection on the dorsum of the foot. Such intervention was performed on 13 children with equinovarus foot deformity, with 11 having good/excellent post-operative result, and two having a fair result. For patients with tibialis posterior overactivity and dorsiflexor weakness, two additional approaches were developed where the split posterior tibialis tendon was routed through the interosseous membrane.

By attaching the split posterior tibialis tendon to the lateral cuneiform on in 23 feet, Saji et al. (81) achieved excellent results in 61% of feet, good results in 35% of feet and only one (4%) poor result

after 8.4 years of follow-up. Different authors also transferred the split posterior tibialis half through the interosseous membrane to the dorsum of the foot, attaching it to the distal peroneus brevis tendon (69). This procedure was performed on 21 feet, where excellent/good results were obtained in 90%, with only two poor results 2.4 years after the surgery.

In order to avoid the overactivity of the remaining, non-transferred part of the tibialis posterior tendon, experienced orthopaedic surgeon from our institution and the author developed a modified SPOTT technique, elongating the remaining part of the tibialis posterior tendon. Many times, the length of the remaining part of the tibialis posterior tendon does not allow the foot to be placed in apposition of slight valgus which is essential in order to avoid recurrence to varus later on. By performing the Z plasty before anchoring the transferred part of the tibialis posterior muscle, the correct attachment site and foot position are much more easily achieved. Lengthening the fixed arm also allows for easier tensioning and distal attachment positioning of anterior tibialis when the SPLATT procedure is preformed concomitantly. This procedure was performed on 12 feet, where all operated cases had excellent/good results over median 8.5 years of follow-up.

Spastic hemiplegia was the most prevalent CP form in our cohort of patients, present in 70% of cases, excellent postoperative results were achieved in 3 of 7 patients with hemiplegia, and good outcome registered in remaining 4 patients.

This thesis had several limitations. First, this was a retrospective cohort, with no randomization performed. Absence of randomization in our study had the potential to introduce bias. Second, laboratory gait analysis was not available at our institution. The spasticity of tibialis posterior muscle was clinically tested clinically by the experienced orthopaedic surgeon from our institution and all the surgeries were performed by him as well, which could be an additional source of bias. However, the analysed data covered a long period of time and evaluated postprocedural success of different surgical techniques in challenging patients with foot deformity due to CP.

6. CONCLUSIONS

Conclusions of the evaluation of split tibial tendon transfers in correction of varus foot deformity and prevention of skeletal contractures in patients with spastic cerebral palsy are following:

- SPLATT procedure improved functional mobility in the majority of treated patients with pes varus and CP, with no worsening of functional mobility after the intervention.
- After an average follow-up of seven years, favourable postprocedural outcome (excellent/good) was registered in 87.5% of operated feet.
- Postoperative fixed skeletal contracture was recorded in only one (6.3%) patient with spastic hemiparesis.
- Favourable surgical outcome was noted in all patients with GMFCS level I, and in the majority of patients with GMFCS level II and III.
- Age at surgery showed no correlation with the success of SPLATT interventions.
- Improved and equal functional mobility was recorded in all patients treated with SPOTT procedure, with no decline in functional mobility.
- After an average follow-up of eight years, excellent/good postprocedural results were observed in 65 (85.6%) operated feet.
- Postoperative fixed skeletal contracture was recorded in one (1.3%) patient with quadriparesis.
- Favourable result of the SPOTT technique was recorded in all patients with preoperative GMFCS level I, and in the majority of patients with level II and level III. All patients with GMFCS level IV had poor postoperative result.
- Age at time of the surgery did not correlate with the outcome of SPOTT procedure.
- Concomitant SPOTT and SPLATT interventions in patients with varus foot deformity improved functional mobility in the majority of patients (88.9%), with no postoperative worsening of functional mobility.
- After an average follow-up of 8.5 years, favourable result was recorded in 72.2% of treated patients.
- In the group of patients treated with SPOTT+SPLATT interventions, one (5.5%) patient with spastic quadriparesis had fixed skeletal contracture.
- Good/excellent results were recorded in all patients with level I, level II and level IV on GMFCS, and in 37% of patients with level III.
- There was significant correlation between the surgical outcome and patients' age at the time of the surgery.
- Modified SPOTT procedure improved functional mobility in half of the operated patient, while other half of the patients had no change in postoperative functional mobility.
- After an average follow-up of 8.5 years, favourable postoperative outcome was observed in all treated patients.
- There were no postoperative fixed skeletal contractures in patients treated with modified SPOTT technique.
- There was significant correlation between the surgical outcome according to Kling's criteria and patients' age at the time of the surgery.

• Split tibial tendon transfers (SPOTT, SPLATT, SPOTT+SPLATT, and modified SPOTT procedure) are proven successful in treating varus foot deformity in the majority of patients with spastic CP even after long-term follow-up.

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BIOGRAFY OF THE AUTHOR

Marko Aleksić was born on May third 1983. in Belgrade where he finished elementary school. He graduated high school with honors in the class of 2001 at New Burgh Free Academy in the State of New York, USA. On the same year, he enrolled at the Medical School of the University of Belgrade. He graduated in January 2008 with an average grade of 9.69, gaining the title of Medical Doctor. He passed the state examination test for medical doctors in September of 2008. He enrolled in Specialist Academic Studies in Orthopedics in 2008 and defended his final academic paper on "The Influence of Botulinum Toxin Type A in the Treatment of Spastic Deformities in Patients with Cerebral Palsy" in February 2011. gaining the title of Academic Specialist in Orthopaedics. He completed his residency in Orthopedic Surgery and Traumatology at the Medical School of the University of Belgrade on July 12, 2017. He is a doctoral student in the PhD study program "Skeletal biology and orthopedic aspects of bone fragility" at the Medical School of the University of Belgrade. Since June 2010, he has been permanently employed at the Institute for Orthopedic Surgery, "Banjica", where he works as an orthopaedic surgeon in the Spine Surgery Center. He has completed additional education in spine surgery, mainly pediatric spine deformity surgery in University Hospital in Brno, Czech Republic in 2018, and SJD Barcelona Children hospital, Catalonia, Spain in 2019. He has completed many cadaver courses in different aspects of spine surgery that were held in Vienna, Madrid, Budapest, and Prague. He is a member of Europsine and AO Spine. He has published four papers in SCI indexed journals. As a research associate, he has participated in several clinical studies in orthopaedic surgery. He is the author and co-author of numerous papers presented at national and international congresses. His special professional interest is related to the surgical treatment of deformities in children with neuromuscular diseases.

образац изјаве о ауторству

Изјава о ауторству

Име и презиме аутора Марко Алексић

Број индекса 2018/5090

Изјављујем

да је докторска дисертација под насловом

Евалуација успешности парцијалних тибијалних тетивних транспозиција у корекцији варусног деформитета стопала и превенцији коштаних контрактура код пацијената са спастичном формом церебралне парализе

- резултат сопственог истраживачког рада;
- да дисертација у целини ни у деловима није била предложена за стицање друге дипломе према студијским програмима других високошколских установа;
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У Београду, 19.07.2020.

образац изјаве о истоветности штампане и електронске верзије докторског рада

Изјава о истоветности штампане и електронске верзије докторског рада

Име и презиме аутора Марко Алексић

Број индекса 2018/5090

Студијски програм - Докторске академске студије, модул: Биологија скелета

Наслов рада: Евалуација успешности парцијалних тибијалних тетивних транспозиција у корекцији варусног деформитета стопала и превенцији коштаних контрактура код пацијената са спастичном формом церебралне парализе

Ментор проф. др Зоран Башчаревић

Изјављујем да је штампана верзија мог докторског рада истоветна електронској верзији коју сам предао/ла ради похрањивања у **Дигиталном репозиторијуму Универзитета у Београду.**

Дозвољавам да се објаве моји лични подаци везани за добијање академског назива доктора наука, као што су име и презиме, година и место рођења и датум одбране рада.

Ови лични подаци могу се објавити на мрежним страницама дигиталне библиотеке, у електронском каталогу и у публикацијама Универзитета у Београду.

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образац изјаве о коришћењу

Изјава о коришћењу

Овлашћујем Универзитетску библиотеку "Светозар Марковић" да у Дигитални репозиторијум Универзитета у Београду унесе моју докторску дисертацију под насловом:Евалуација успешности парцијалних тибијалних тетивних транспозиција у корекцији варусног деформитета стопала и превенцији коштаних контрактура код пацијената са спастичном формом церебралне парализе

која је моје ауторско дело.

Дисертацију са свим прилозима предао/ла сам у електронском формату погодном за трајно архивирање.

Моју докторску дисертацију похрањену у Дигиталном репозиторијуму Универзитета у Београду и доступну у отвореном приступу могу да користе сви који поштују одредбе садржане у одабраном типу лиценце Креативне заједнице (Creative Commons) за коју сам се одлучио/ла.

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